

PhD thesis 2020

Student ID: 4289861

Study title:

Key stakeholders' perspectives on the development of an early dietary phosphate self-management strategy for children and young people with chronic kidney disease stages 1-3: A mixed-methods study

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Word count: 99368

Acknowledgements

Firstly, I would like to thank Professor Alan Watson for his inspiration which started me on this research journey.

Importantly, I sincerely thank Dr Pippa Hemingway, Professor Gina Higginbottom and Dr Martin Christian for their skilful guidance, ceaseless support and encouragement to the end. You always knew that I could do it – even when I was not so sure!

To the friends who have been a rock and a safe place of refuge when the pressure was great, and the climb was tough – you know who you are. Thank you for being there, your encouragement and friendship has carried me through. This journey has been walked with you.

To the people who are my world, my husband, Ben, I am lost for words! Your strength, ceaseless encouragement and belief in me has been overwhelming. Thank you. To my beautiful children, Abi, Gracie and Reuben, sorry that mummy has been so absent over the past few years, and thank you for insisting on our regular chats Reuben. To my parents and family, your prayers, unfailing encouragement and belief in me has been invaluable. Thank you.

Finally, I am immensely grateful to the many children and young people, parents and healthcare professionals who so willing gave up their time to take part in this study. Without you this study would not have been possible. Thank you.

Glossary

ATP – Adenosine triphosphate

AKI – Acute Kidney Injury

BCT – Boot Camp Translation

CAKUT – Congenital anomalies of the kidney and urinary tract

CD – Chronic Disease

CKD – Chronic Kidney Disease

CKD-MBD – Chronic Kidney Disease-Metabolic Bone Disease

CKiD – Chronic Kidney Disease in Children

CLAHRC – Collaboration for Leadership in Applied Health Research and Care

CYP – Children and Young People

DoH – Department of Health

eGFR – Estimated Glomerular Filtration Rate

EMEESY – East Midlands, East of England and South Yorkshire

EPI – Early Phosphate Intervention

EPSM – Early Dietary Phosphate Self-Management

ERD – Established Renal Disease

ESRD – End-stage Renal Disease

FGF-23 – Fibroblast Growth Factor 23

GFR – Glomerular Filtration Rate

HCP – Healthcare Professional

HRA – Health Research Authority

ID – Interpretive Description

IRAS – Integrated Research Application System

KDOQI – Kidney Disease Outcomes Quality Initiative

MARM – Masters in Research Methods

MRC – Medical Research Council

MMR – Mixed Method Research

NHS – National Health Service

NEM – Network Education Meeting

NIHR – National Institute for Health Research

NSF – National Service Framework

PIS – Participant Information Sheet

PMARP – Per million age-related population

PRiNG – Paediatric Renal interest Group

PTH – Parathyroid Hormone

RCTs – Randomised control trials

RO – Research Objective

RRT – Renal Replacement Therapy (Dialysis)

SM – Self-management

SES – Socio-economic sectors

STP – Sustainability and Transformation Partnerships

UD – Usual diet

UK – United Kingdom

UNCR – United Nations Convention on the Rights of the Child

UoN – University of Nottingham

UPE – Urinary Phosphate Excretion

US – United States

VLP – Very low protein

Abstract

Background: Chronic kidney disease (CKD), classified as stages 1-5, costs the NHS an estimated £1.44-1.45 billion annually, with the cost of provision predominantly in dialysis services when milder CKD progresses to stage 5. Delaying disease progression and the commencement of dialysis, is desirable. CKD alters phosphate metabolism, which is strongly associated with a rise in fibroblast growth factor 23 (FGF-23) and disease progression. The provision of an early dietary phosphate self-management (EPSM) strategy may reduce FGF-23 levels and delay disease progression.

Purpose: To explore the perspectives of children and young people (CYP), parents and key healthcare professionals (HCPs) towards the development of an EPSM strategy for CYP with CKD 1-3, within a managed paediatric renal network.

Methodology: This three-phase study used an exploratory QUALITATIVE-quantitative mixed-methods research (MMR) design. A systematic review informed the development of a topic guide for qualitative semi-structured interviews (n=17) with CYP, parents and HCP (n=21). Interpretive description (ID) provided the qualitative methodology. The co-constructed themes developed from the qualitative interviews informed the first round of the modified online Delphi survey.

Findings: Integration of the findings produced 3 novel MMR themes, emphasising the need for (1) a new paradigm of early dietary intervention to promote kidney health; to be (2) offered by the paediatric renal dietitian as part of an early engagement diagnosis package; and (3) accessible culinary medicine workshops, providing education, social networking, and upskilling to address the practical prerequisites of the strategy.

Recommendations: The testing and implementation of an EPSM, and an action plan to address the challenges and enablers to implementing the strategy.

Conclusion: This MMR produced a collective agreement and some early modelling on the operationalisation of an EPSM strategy. The findings will facilitate developments in the regional paediatric nephrology network, as well as informing national and international policy to improve health outcomes, delay disease progression, and reduce healthcare utilisation and costs.

Publications and dissemination

Publications

- Pugh, P., Christian, M. Higginbottom, G, A. and Hemingway, P. (2020) Children's, parents' and other stakeholders' perspectives on early dietary self-management to delay disease progression of chronic disease in children: a mixed studies systematic review using a narrative synthesis. **Patient Education and Counseling** (peer review).
- McAlister, L., Pugh, P., Greenbaum, L et al. (2020) The dietary management of calcium and phosphate in children with CKD stages 2-5 and on dialysis – clinical practice recommendation from the Pediatric Renal Nutrition Taskforce. **Pediatric Nephrology.** 35: pp. 501-518. https://doi.org/10.1007/s00467-019-04370-z
- Engel, B and Pugh, P. (2019) Paediatric Nutrition. In: Thomas, N. **Renal Nursing: Care and Management of People with Kidney Disease.** John Wiley and Sons Ltd.
- Pugh, P., Hemingway, P., Christian, M. and Higginbottom, G, A. (2018) Children's, parents' and other stakeholders' perspectives on early dietary self management to delay disease progression of chronic disease in children: a protocol for a mixed studies systematic review with a narrative synthesis. **Systematic Reviews,** 7. https://doi.org/10.1186/s13643-017-0671-8

Oral presentations

- (1) Early dietary phosphate intervention. (2) Releasing growth potential in children with CKD and Intercontinental variation in dietetic practice. Paediatric Renal Nutrition Academy and the 38th annual Dialysis Conference, Orlando, Florida 3-6th March 2018.
- Taking Action Early: How do we reduce phosphate without compromising growth? 5th International Renal Dietetic Conference. Glasgow 7-9th September 2017.
- (1) Early Phosphate Intervention. (2) UK Networking Initiatives. 4th International Renal Dietetic Conference. Manchester 3rd- 4th November 2016.

Poster presentations

- Early Phosphate Intervention: Is the consumption of processed food affecting children's kidney function? Nottingham Paediatric research Showcase. 21st June 2018 and the East Midlands Post-Doctoral Research Conference, Bishop Grosseteste University, Lincoln.19 Sept 2018.
- Bagel or Bangers: Are children's food choices affecting their kidney function? East Midlands Doctoral Network Postgraduate Research Conference. Nottingham 13th September 2017.

Table of Content

STUDY TITLE: I

ACKNOWLEDGEMENTS II

GLOSSARY III

ABSTRACT V

PUBLICATIONS AND DISSEMINATION VI

TABLE OF CONTENT VIII

TABLE OF TABLES XVI

TABLE OF BOXES XVIII

TABLE OF FIGURES XX

CHAPTER 1: INTRODUCTION AND CLINICAL CONTEXT TO THE STUDY 1

1.1 INTRODUCTION 1
1.1.1 INSPIRATION BEHIND THE EARLY DEVELOPMENT OF THE RESEARCH 3
1.2 CLINICAL CONTEXT TO THE STUDY 4
1.2.1 CHRONIC KIDNEY DISEASE 4
1.2.2 CLASSIFYING CHRONIC KIDNEY DISEASE 5
1.2.3 STAGES OF CHRONIC KIDNEY DISEASE 5
1.2.4 DISEASE PROGRESSION 6
1.2.5 LIFE EXPECTANCY 7
1.3 DIETARY PHOSPHATE 8
1.3.1 THE NEED FOR EARLY DIETARY PHOSPHATE SELF-MANAGEMENT 10
1.3.2 SPECIALIST RENAL DIETETIC SUPPORT 11
1.3.3 BURDEN OF CARE 11
1.4 DELIVERY OF 'CARE CLOSE TO HOME' 12
1.4.1 THE POLITICAL CONTEXT 12
1.4.2 THE EMEESY MANAGED PAEDIATRIC NEPHROLOGY NETWORK 12
1.5 SELF-MANAGEMENT 14
1.5.1 DEFINITION 14
1.5.2 WHEN IS IT BEST TO ENGAGE CYP IN SELF-MANAGEMENT? 14
1.5.3 THEORETICAL FRAMEWORK 16
1.5.4 CHILD AND YOUNG PERSON-CENTRED APPROACH 17

1.6 SUMMARY 18

CHAPTER 2: RESEARCH AIM AND OBJECTIVES, DESIGN AND METHODOLOGY. 19

2.1 INTRODUCTION 19
2.2 AIM AND OBJECTIVES OF THE RESEARCH 19
2.2.1 RESEARCH AIM 19
2.2.2 RESEARCH OBJECTIVES 19
2.2.3 THE RESEARCH PLAN 20
2.3 ONTOLOGY AND EPISTEMOLOGY 22
2.3.1 PRAGMATISM 23
2.4 MIXED-METHODS RESEARCH 25
2.4.1 THE CHOSEN MIXED-METHODS DESIGN 26
2.4.2 MIXED-METHOD DESIGN TYPOLOGIES 28
2.4.3 RATIONALE FOR USING THE MIXED-METHODS DESIGN 30
2.5 INTERPRETIVE DESCRIPTION METHODOLOGY 31
2.6 SUMMARY 33

CHAPTER 3: PHASE 1 - LITERATURE REVIEW AND A MIXED-STUDIES SYSTEMATIC REVIEW WITH A NARRATIVE SYNTHESIS 34

3.1 INTRODUCTION 34
3.1.1 SEARCH STRATEGY 34
3.1.2 FGF-23 AND DISEASE PROGRESSION IN CYP 35
3.1.3 DIETARY PHOSPHATE RESTRICTION AND FGF-23 37
3.1.4 CLINICAL PRACTICE GUIDELINES FOR CKD STAGES 1-3, AND STAKEHOLDER INVOLVEMENT IN THEIR DEVELOPMENT 41
3.1.5 PATIENT PERSPECTIVE ON CKD SELF-MANAGEMENT 43
3.1.6 HEALTHCARE PROFESSIONALS' UNDERSTANDING OF SELF- MANAGEMENT 45
3.2 GAPS IN THE LITERATURE 48
3.3 THE MIXED-STUDIES SYSTEMATIC REVIEW WITH A NARRATIVE SYNTHESIS 49
3.4 RATIONALE FOR THE REVIEW 49
3.5 WHAT IS A MIXED-STUDIES SYSTEMATIC REVIEW? 50
3.6 WHAT IS A MIXED-STUDIES SYSTEMATIC REVIEW WITH A NARRATIVE SYNTHESIS, AND WHY WAS IT CHOSEN FOR THIS STUDY? 50
3.7 KNOWLEDGE GAP ON EARLY DIETARY INTERVENTIONS FOR CHILDREN WITH A CHRONIC DISEASE OR CKD STAGE 1-3 51
3.8 REVIEW QUESTION 52
3.9 METHODS 52
3.9.1 INCLUSION AND EXCLUSION CRITERIA 52
3.9.2 STUDY IDENTIFICATION AND SEARCH STRATEGY 55
3.10 QUALITY ASSESSMENT 59
3.10.1 QUALITY ASSESSMENT OF PUBLISHED LITERATURE 60
3.10.2 QUALITY ASSESSMENT OF GREY LITERATURE 60

3.10.3 Data extraction 61
3.11 Findings 64
3.11.1 Summary of the included studies 66
3.11.2 Quality assessment findings for each study 70
3.11.3 Structured thematic analysis summary and narrative synthesis themes 73
3.11.4 Theme 1: The role of education in early dietary management 79
3.11.5 Theme 2: Parents/caregivers role in early dietary SM 83
3.11.6 Theme 3: Early dietary intervention in the management of chronic disease 85
3.11.7 Theme 4: Challenges and enablers to dietary change 89
3.12 Discussion of the thematic analysis, data interaction and integration 93
3.13 Summary 94

CHAPTER 4: PHASE 2 – METHODS FOR THE QUALITATIVE DIMENSION 96

4.1 Introduction 96
4.1.1 MRC Framework - Intervention development 96
4.2 Study population, sample size and sampling strategy 98
4.2.1 Study population 98
4.2.2 Sample size and sampling strategy 99
4.2.3 Inclusion criteria 99
4.2.4 Exclusion criteria 101
4.2.5 Research sites 101
4.2.6 Access and recruitment 101
4.3. Research setting 105
4.3.1 Interview location and context 105
4.4 Data collection tools 106
4.4.1 Tools for data collection 106
4.5 Data collection 107
4.5.1 Data collection process 107
4.5.2 Data collection method 107
4.5.3 Semi-structured interviews 109
4.6 Data analysis using Interpretive Description 111
4.6.1 Descriptive data analysis 112
4.6.2 Interpretive data analysis 118
4.7 Ethics application 120
4.7.1 Ethical considerations 121
4.7.2 Ethical consideration when conducting research with children 121
4.7.3 Interviewing children and young people 123
4.7.4 Data management 125
4.8 Summary 125

CHAPTER 5: PHASE 2 – RESULTS OF THE QUALITATIVE DIMENSION 127

5.1 Results 127
5.1.1 Participants interviewed 127

5.1.2 Theme 1: The need for support through timely early engagement and relationship-building 129
5.1.3 Theme 2: The need to take control whilst establishing a new normal 140
5.1.4 Theme 3: The need for early dietary phosphate education 157
5.2 Summary 168

CHAPTER 6: PHASE 3 – METHODS FOR THE QUANTITATIVE DIMENSION 170

6.1 Introduction 170
6.2 The Delphi technique 170
6.2.1 Principle features of Delphi 170
6.2.2 The Delphi process 171
6.2.3 Defining Delphi 172
6.3 Conducting and Reporting of Delphi Studies (CREDES) 172
6.3.1 Rationale for the choice of the Delphi technique 173
6.4 Planning the Delphi Survey 174
6.4.1 Modified Delphi 174
6.4.2 Level of agreement 174
6.5 Study population, sample size and sampling strategy 175
6.5.1 Study population 175
6.5.2 Sample size 176
6.5.3 Sampling strategy 176
6.5.4 Expert panellist 177
6.5.5 Participant anonymity 177
6.5.6 Inclusion criteria 178
6.5.7 Exclusion criteria 178
6.5.8 Recruitment 179
6.6 Research setting 180
6.7 Tool development 180
6.7.1 Demographic questionnaire 180
6.7.2 The level of agreement used 180
6.7.3 Statements that did not reach agreement 180
6.7.4 Development of a Delphi Steering Group 181
6.7.5 Designing the Delphi survey 182
6.7.6 Instructions for use 182
6.7.7 Preparing the statements for the round 1 Delphi survey 183
6.7.8 A 4–point Likert scale 185
6.7.9 Piloting the round 1 Delphi Survey 186
6.7.10 Postal survey 187
6.7.11 Why use an online Delphi survey? 187
6.7.12 The best day to send out the survey 188
6.7.13 Number of Delphi rounds 188
6.8 Data collection and analyses 189
6.8.1 Round 1 data collection 189
6.8.2 Round 1 data analyses 191

6.8.3 Feedback to the panellists 194
6.8.4 Round 2 data collection 196
6.8.5 Round 2 data analysis 197
6.8.6 Round 3 data collection 198
6.8.7 Round 3 data analysis 198
6.9 Summary 198

CHAPTER 7: FINDINGS OF THE DELPHI SURVEY 200

7.1 Introduction 200
7.2 Results of the statement development for Delphi round 1 200
7.2.1 Definition category 201
7.2.2 Rationale for the development of an EPI category 201
7.2.3 Intended users category 202
7.2.4 Delivery of an EPI category 202
7.2.5 Other considerations and potential concerns category 203
7.3 Results of Delphi Survey round 1 205
7.3.1 Round 1 response rates 205
7.3.2 Demographic profiles of the panellists 206
7.4 Results of statements in round 1 208
7.4.1 Round 1 results for the definition category (S1-3) 208
7.4.2 Round 1 results for the rationale category (S4-15) 211
7.4.3 Round 1 results for the intended user's category (S16-18) 212
7.4.4 Round 1 results for delivery of an EPI category (S19-27) 213
7.4.5 Round 1 results for the other considerations category (S28-48) 215
7.4.6 Summary of Delphi round 1 225
7.5 Results of statements in round 2 225
7.5.1 Development of the round 2 survey 225
7.5.2 Round 2 response rate 227
7.5.3 Round 2 results for the definition category 228
7.5.4 Round 2 results for the rationale category 231
7.5.5 Round 2 results for the intended user's category 233
7.5.6 Round 2 results for the delivery category 233
7.5.7 Round 2 results for the other considerations category 235
7.5.8 Stability of agreement across round 1 and 2 242
7.5.9 Summary of Delphi round 2 243
7.6 Results of statements in round 3 243
7.6.1 Development of the round 3 survey 243
7.6.2 Round 3 response rate 244
7.6.3 Round 3 results for the definition category 246
7.6.4 Round 3 results for the rationale category 248
7.6.5 Round 3 results for the delivery and other considerations category 249
7.6.6 Summary of Delphi round 3 250
7.6.7 Statements reaching consensus across Delphi rounds 1-3 250
7.7 Summary 254

CHAPTER 8: INTEGRATION OF THE FINDINGS FROM PHASE 1-3 255

8.1 INTRODUCTION 255
8.2 RESPONSE TO RESEARCH QUESTION AND OBJECTIVES 255
8.2.1 OVERVIEW OF THE KEY STAKEHOLDERS' PERSPECTIVES TOWARDS THE DEVELOPMENT OF AN EPI .. 256
8.2.2 OBJECTIVE 1: ACHIEVE A CONSENSUS DEFINITION FOR EPI 257
8.2.3 OBJECTIVE 2 AND 3: EXPLORATION AND CONSENSUS ON 'HOW' AND 'WHO' SHOULD DELIVER THE EPI STRATEGY 262
8.2.4 OBJECTIVE 4: EXPLORATION AND CONSENSUS ON THE RECIPIENTS OF THE EPI 282
8.2.5 OBJECTIVE 5: EXPLORATION AND CONSENSUS ON 'WHEN' TO DELIVER THE EPI 286
8.2.6 OBJECTIVE 6: EXPLORATION AND CONSENSUS ON 'WHERE' TO DELIVER THE EPI 292
8.2.7 OBJECTIVE 7: IDENTIFYING CHALLENGES AND ENABLERS 296
8.3 SUMMARY OF THE INTEGRATIVE FINDINGS 301

CHAPTER 9: DISCUSSION AND CONCLUSION 303

9.1 INTRODUCTION 303
9.2 MMR THEME 1 – REFRAMING THE EARLY DIETARY PHOSPHATE INTERVENTION (EPI) STRATEGY TO CREATE A NEW PARADIGM OF EARLY CKD MANAGEMENT 305
9.2.1 RENAL HEALTHY-EATING FOR EARLY CKD 305
9.2.2 EPI PROMOTES KIDNEY HEALTH TO DELAY CKD PROGRESSION 307
9.2.3 THEME SUMMARY 308
9.3 MMR THEME 2 – THE EARLY INTERVENTION 'RENAL-HEALTHY-EATING' DIET NEEDS TO BE OFFERED TO ALL CYP WITH PROGRESSIVE CKD, BY THE PAEDIATRIC RENAL DIETITIAN AS PART OF THE DIAGNOSIS PACKAGE, AND ADOPTED BY THE WHOLE FAMILY 309
9.3.1 PART OF THE DIAGNOSIS PACKAGE 310
9.3.2 EPI IS FOR CYP WITH ALL STAGES OF CKD 311
9.3.3 EPI SHOULD BE ENCOURAGED FOR THE WHOLE FAMILY 312
9.3.4 EPI DELIVERED BY THE PAEDIATRIC RENAL DIETITIAN OFFERS TRUSTED EARLY ENGAGEMENT 312
9.3.5. THEME SUMMARY 314
9.4 MMR THEME 3 – CULINARY MEDICINE WORKSHOPS OFFER EDUCATION, SUPPORT AND UPSKILLING 314
9.4.1 EPI WORKSHOPS WOULD TRANSLATE THE COMPLEXITY OF THE DIETARY PRESCRIPTION INTO A SIMPLE, REALISTIC AND ACTIONABLE MESSAGE 315
9.4.2 EPI WORKSHOPS WILL BE INCLUSIVE AND LED BY THE PAEDIATRIC RENAL DIETITIAN 317
9.4.3 EPI WORKSHOPS WOULD BE PART OF AN INDIVIDUALISED PLAN 318
9.4.4 EPI WORKSHOPS OFFER EARLY ENGAGEMENT AND RELATIONSHIP BUILDING 318
9.4.5 FREQUENCY AND LOCATION OF THE EPI WORKSHOPS 319
9.4.6 THEME SUMMARY 320
9.5 CONTRIBUTIONS TO KNOWLEDGE AND PRACTICE 320
9.5.1 CONTRIBUTIONS TO KNOWLEDGE 321
9.5.2 CONTRIBUTIONS TO PRACTICE 322
9.6 REFLEXIVITY AND POWER RELATIONS WHEN UNDERTAKING RESEARCH WITH COLLEAGUES/PATIENTS 324
9.7 MAINTAINING RESEARCH QUALITY (INTEGRITY AND TRUSTWORTHINESS) 326

9.8 STRENGTHS AND LIMITATIONS OF THIS STUDY AND DIRECTIONS FOR FUTURE RESEARCH 328
9.8.1 STRENGTHS OF THIS MMR STUDY 328
9.8.2 LIMITATIONS OF THIS MMR STUDY 329
9.8.3 DIRECTIONS FOR FUTURE RESEARCH 330
9.9 MAIN CONCLUSIONS 331

CITED REFERENCES.....333

APPENDIX 1. DETAILED SEARCH STRATEGY EXAMPLE ON MEDLINE.....373

APPENDIX 2. MIXED METHODS APPRAISAL TOOL 374

APPENDIX 3. TYNDALL'S (2010) AACODS CHECKLIST FOR THE EVALUATION AND CRITICAL APPRAISAL OF GREY LITERATURE.....375

APPENDIX 4. EXAMPLE OF DATA EXTRACTION VARIABLES.....376

APPENDIX 5. EXAMPLE OF CALCULATING THE METHODOLOGICAL QUALITY OF A STUDY 377

APPENDIX 6. RENAL CLINIC AND EMEESY WEBSITE RECRUITMENT POSTER...378

APPENDIX 7. THE INVITATION LETTER SENT TO PARENTS AND HCP TO TAKE PART IN AN INTERVIEW.....379

APPENDIX 8. AGE-APPROPRIATE CYP PARTICIPANT INFORMATION SHEET ...380

APPENDIX 9. AGE-APPROPRIATE INTERVIEW GUIDE FOR CYP, AND PARENT/HCP 382

APPENDIX 10. PHOTOGRAPHS OF THE DEVELOPING DATA ANALYSIS PROCESS (STEPS 2 AND 3).....386

APPENDIX 11. A SECTION OF THE WORD MATRIX SHOWING DOCUMENTATION OF ANALYTICAL THINKING.....387

APPENDIX 12. SUPPORTING RESEARCH DOCUMENTS.....388

APPENDIX 13. A SECTION OF DELPHI ROUND 1 SURVEY.....400

APPENDIX 14. MAPPING PHASE 1 AND 2 DATA AGAINST THE DOMAINS FROM THE TIDIER CHECKLIST (2014) 406

APPENDIX 15. DELPHI RESULTS TABLES 411

APPENDIX 16. CROSS TABULATION OF FREQUENCIES FOR COHEN'S KAPPA 427

APPENDIX 17. LETTER OF HRA APPROVAL 429

Table of Tables

Table 1. The stages of chronic kidney disease 6
Table 2. Causes of CKD in CYP 6
Table 3. The bioavailability of a selection of phosphate-rich foods 10
Table 4. The EMEESY network serves 20 local hospitals 13
Table 5. Self-management skills (Lorig et al. 2003) as applied in this doctoral study 16
Table 6. Mixed-methods designs 27
Table 7. Examples of ID in the healthcare literature 33
Table 8. The categories and number of studies identified in the scoping search 35
Table 9. Key data from Portale et al. (2016) study 35
Table 10. Key adult data on GFR and FGF-23 levels from Isakova et al. (2015) and Tsai et al. (2018) 38
Table 11. Data from Sullivan et al. (2009) and Levey et al. (1996) 39
Table 12. Self-management adult studies recognising the need for diet-specific knowledge 44
Table 13. Search terms and Boolean operators 56
Table 14. Databases searched during stage 3 of the search strategy 57
Table 15. Wildcards applied to the Medline search 58
Table 16. Screening and selection criteria checklist 58
Table 17. McGrath et al. (2006) framework for methodological quality 60
Table 18. AcademyHealth (2006) category of relative importance of grey literature 61
Table 19. The four steps for narrative synthesis 61
Table 20. Tools for narrative synthesis and risk of bias 62
Table 21. Study characteristics of all included article (mixed methods, qualitative and quantitative) 67
Table 22. Methodological quality assessment for each included study 70
Table 23. Matrix providing evidence from the studies to support each theme .. 71
Table 24. The development of Theme 1 – the role of education in early dietary management 73
Table 25. Four themes and their relationship across the studies 74
Table 26. Themes and colour coding in the concept maps 81
Table 27. Quantitative findings regarding early dietary intervention 88
Table 28. The initial (step 1) broad-node "relationship and coping" with its sub-nodes and definitions 116
Table 29. An example of analytic notes and questions 119
Table 30. Participants interviewed 128
Table 31. Schedule for Delphi 191
Table 32. Part of an Excel spreadsheet detailing Delphi round 1-3 results for the statements for 10-14 192
Table 33. Summary of Delphi round one responders and non-responders by region 205
Table 34. The primary role, median age and years of experience with CYP with CKD 207
Table 35. Statements at the end of round 1 225

Table 36. Summary of Delphi round 2 responders and non-responders by region 227
Table 37. Primary role and gender of round 2 participants 228
Table 38. Median LOA rating, % LOA and participant group weighting for the rationale statements in round 2 232
Table 39. Median LOA rating, % LOA and participant group weighting for the delivery statements in round 1 and 2 234
Table 40. Median score, % levels of agreement and participant group weighting for the other considerations statements in round 1 and 2 236
Table 41. Kappa statistic (Landis and Koch 1977) 242
Table 42. Results of Cohen's Kappa interrater reliability analysis 243
Table 43. Statements at the end of round 2 244
Table 44. Summary of Delphi round 3 responders and non-respond by region 245
Table 45. Primary role and gender of round 3 participants 245
Table 46. Median LOA rating, % LOA and participant group weighting, and Fisher's Exact test for the definition S2a and b in round 1-3 247
Table 47. Median LOA rating, % LOA and participant group weighting, and Fisher's Exact test for the rationale S12 in round 1 and 3 248
Table 48. Median LOA rating, % LOA and participant group weighting, and Fisher's Exact test for the other considerations: potential concerns S44a and b in round 1-3 249
Table 49. Statements reaching consensus 251
Table 50. Summary of EPI education strategies for early CKD 274
Table 51. Summary of EPI skills strategies for early CKD 277
Table 52. Summary of EPI support strategies for early CKD 282
Table 53. Summary of the recipients of the EPI strategy 286
Table 54. Summary of when the EPI should be delivered 292
Table 55. Summary of the best locations for delivering the EPI 296
Table 56. Comparison between the current and the proposed new paradigm of early CKD 'renal-healthy-eating' dietary intervention 309

Table of Boxes

Box 1. PICO tool 52
Box 2. Inclusion and exclusion criteria 53
Box 3. Key messages informing the interview guide development in Phase 2 ... 94
Box 4. The eight-stages of interpretive description (Thorne, 2008) 111
Box 5. Example of nodes developed in NVivo 12 during description data analysis 114
Box 6. Delphi Steering Group 182
Box 7. Adapted Hoffman's TIDieR (2014) checklist items that informed round 1 DS categories 183
Box 8. An example of labelling all 4-response categories on the Likert scale ... 186
Box 9. Example of information provided to panellists 195
Box 10. An example of a statement that had reached consensus requiring no further response in the round 2 survey 196
Box 11. An example of a misunderstood instruction 197
Box 12. Delphi round 1 survey categories as informed by the TIDieR checklist (2014) 200
Box 13. Median LOA rating, % LOA and participant group weighting for the 3 definition statements in round 1 209
Box 14. Definition S2 210
Box 15. Median LOA rating, % LOA and participant group weighting for the delivery statements in round 1 213
Box 16. Median LOA rating, % LOA and participant group weighting for the other considerations statements in round 1 215
Box 17. Other considerations: building relationship, S28 217
Box 18. Other considerations: support, S32 218
Box 19. Other considerations: early tests undertaken in the out-patient clinic, S38 219
Box 20. Other considerations: early tests undertaken in the out-patient clinic, S39 220
Box 21. Other considerations: potential concerns, S44 222
Box 22. Other considerations: potential concerns, S46 223
Box 23. Other considerations: potential concerns, S48 224
Box 24. Median LOA rating, % LOA and participant group weighting for the definition statements in round 1 and 2 229
Box 25. Definition S2 in round 1-3 230
Box 26. Rationale S12 round 1-3 233
Box 27. Other considerations: support S33b 237
Box 28. Other considerations: potential concerns S42 239
Box 29. Other considerations: potential concerns S44 240
Box 30. Other considerations: potential concerns S45 241
Box 31. The seven research objectives 256
Box 32. Definition S3 411
Box 33. Median LOA rating, % LOA and participant group weighting for the 12 rationale statements in round 1 412
Box 34. Rationale S7 413
Box 35. Rationale S13 414

Box 36. Rationale S14 415
Box 37. Intended user's S18 416
Box 38. Median LOA rating, % LOA and participant group weighting for the 3 intended user's statements in round 1 417
Box 39. Delivery: where to deliver the EPI, S20 418
Box 40. Delivery: how should the EPI be delivered, S23 419
Box 41. Other considerations: support, S34 420
Box 42. Other considerations: SM S35 420
Box 43. Other considerations: SM S37 421
Box 44. Other considerations: early dietary blood test, S41 421
Box 45. Other considerations: potential concerns, S42 422
Box 46. Other considerations: potential concerns, S45 422
Box 47. Other considerations: potential concerns, S47 423
Box 48. Median LOA rating, % LOA and participant group weighting for the intended users statements in round 1 and 2 423
Box 49. Delivery S24 round 1-3 424
Box 50. Median LOA rating and % LOA and participant group weighting for the delivery S24 in round 1-3 425
Box 51. Median LOA rating, % LOA and participant group weighting for the delivery S33b in round 3 425
Box 52. Median LOA rating, % LOA and participant group weighting for the other considerations: potential concerns S42 in round 1-3 426
Box 53. Median LOA rating, % LOA and participant group weighting for the other considerations: potential concerns S45a and b in round 1-3 426

Table of Figures

Figure 1. The three-phases of this MMR study 1
Figure 2. Mixed-methods research design 21
Figure 3. Schematic of Sequential Exploratory Mixed-Methods Research Design (adapted from Creswell et al. 2003) 28
Figure 4. Chronic disease and CKD search strategy 57
Figure 5. PRISMA flow chart sequencing the review process (2015) 65
Figure 6. Key sources and facets of education and their relationship with other themes 82
Figure 7. Key role and profile of parents and caregivers in the early dietary self-management of children with chronic diseases and the relationship with other themes. 85
Figure 8. Factors influencing early dietary self-management and their relationship with other themes 87
Figure 9. Challenges and enablers to dietary change and their relationship with other themes 92
Figure 10. MRC Framework intervention development 97
Figure 11. Research process, access and recruitment 102
Figure 12. Evolving group signifiers in Step 1, 2 and 3 117
Figure 13. Ethics application timeline 120
Figure 14. Summary of theme 1 findings 129
Figure 15. Summary of theme 2 findings 141
Figure 16. Summary of theme 3 findings 158
Figure 17. The Delphi process 171
Figure 18. CONSORT (2010) flow diagram for Delphi round 1-3 206
Figure 19. Statements reaching consensus across 1-3 Delphi rounds 226
Figure 20. Objective 1 themes 258
Figure 21. Objective 2 and 3 education themes 263
Figure 22. Objective 2 and 3 skills themes 274
Figure 23. Objective 2 and 3 support themes 278
Figure 24. Objective 4 themes 283
Figure 25. Objective 5 themes 287
Figure 26. Objective 6 themes 292
Figure 27. Objective 7 themes 296
Figure 28. Concept model of the novel MMR themes 304

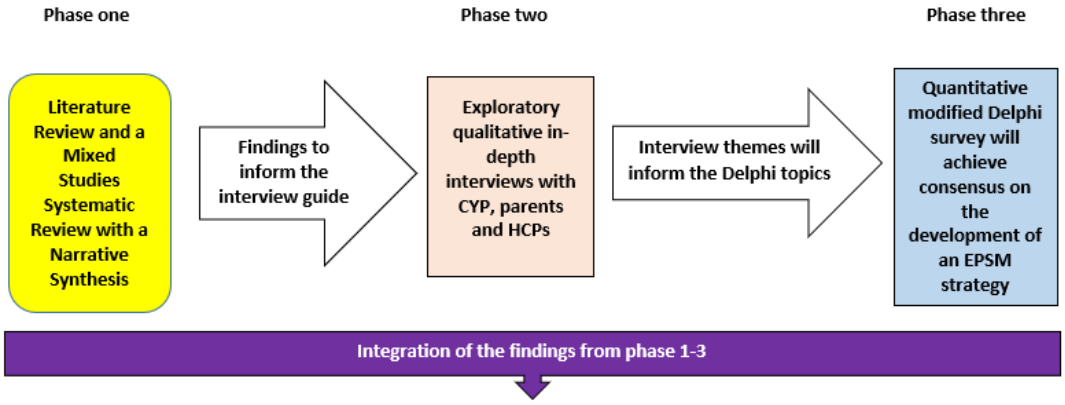
Chapter 1: Introduction and clinical context to the study

1.1 Introduction

This thesis presents a sequential exploratory QUALITATIVE–quantitative three phase MMR study (Creswell et al. 2003). The perspectives of CYP, parents and HCPs on the development of an EPSM strategy (used interchangeably with the term early phosphate intervention – EPI), to delay disease progression, for CYP with CKD stage 1-3 within a managed paediatric renal network was explored. The thesis comprises nine chapters. Chapter 1 sets the scene, providing an introduction to the thesis, the inspiration behind the study, and positions the study within a biomedical and self-management (SM) clinical context.

Chapter 2 presents the ontological and epistemological underpinnings to the study. Followed by the three-phase MMR model (see Figure 1 below), the methodology of interpretive description (Thorne, 2013), and the aims and objectives of this study.

Figure 1. The three-phases of this MMR study



Next, the three study phases are separated into sections, firstly presenting Phase 1, then the methods and results for Phase 2 and 3. Other MMR researchers have used this approach (Ruhi, 2010; West, 2011). Chapter 3 presents Phase 1 of the study, a scoping search on early dietary phosphate SM in CYP with CKD; the findings explore the debates on FGF-23 and CKD progression in CYP, dietary phosphate restriction and FGF-23, the patient's perspective on CKD SM, and the HCP's understanding of SM. Next, evidence gaps in the literature are highlighted to inform a mixed studies

systematic review with a narrative synthesis. The rationale, process and findings of the systematic review are detailed. The main findings and recommendations, which were integrated to shape the qualitative semi-structured interview guide in Phase 2 are discussed.

Chapter 4 presents the qualitative methods used in Phase 2 of the study. The chapter begins with a description of the research method, semi-structured interviews, and how it addressed the research question. The study population, sample size, sampling strategy, and the data analysis procedures are presented, before examining the research ethics with a particular emphasis on undertaking research with CYP.

Chapter 5 presents the Phase 2 qualitative findings as three key themes, and multiple sub-themes. Chapter 6 presents the quantitative method used in Phase 3 of the study. The chapter begins with a description of the Delphi technique (Dalkey and Helmer, 1963), and how this research method connected a group of CKD management experts and enabled the use of the co-constructed data from Phase 1 and 2 to build consensus on the development of an EPSM strategy. The study population, sample size and sampling strategy are described, before outlining the membership and functions of the Delphi Steering Group. Each stage of the Delphi survey is described, from the pilot survey to the three Delphi consensus rounds with their respective data collection and analysis. Chapter 7 presents the findings of the Delphi survey, with a statistical analysis detailing the 56 statements reaching consensus, and the four that did not.

Chapter 8 draws together the findings from Phase 1, 2 and 3 of the study, integrating these empirical research outputs to answer the research question and objectives that formed the basis for this study. Chapter 9 presents a discussion of the integrative findings from this MMR study as three novel MMR themes. It also highlights the contributions of this research to theory and practice. The chapter then provides a reflexive account of the author's positionality and recognition of power relations while undertaking the research with colleagues/patients. Research integrity is presented, before discussing the strengths and limitations of this study and direction for future research. Finally, the conclusion marks a culmination of the ideas, insights and outcomes of this thesis.

This thesis is written in the third person, except for the next section in which the researcher describes the inspiration behind the genesis of the study, where she uses the first person (Nalubega, 2017).

1.1.1 Inspiration behind the early development of the research

The origins of this PhD lie in my extensive experience as dietitian. I have been employed by the NHS as a dietitian for over 23 years, specialising in paediatric nephrology for the past 18 years. Allied to medicine, paediatric renal dietetics provides a crucial and complex toolkit to support the management of childhood renal disease, to optimise growth and minimise the complications of renal disease (Rees and Jones, 2013). Over the years, I have had many conversations with anxious parents seeking ways to help their child, in particular, wanting to know if early dietary change can delay the progression of CKD. Hereafter the term child will be used interchangeably with CYP.

As much of my time in clinical practice focuses on managing poor phosphate control during the later stages of CKD management, predominantly influenced by dietary habits established during the early stages, I am particularly interested in the proactive early management of dietary phosphate. Convinced that dietary phosphate management is more than a reactive tool that a dietitian uses to urgently address deranged serum phosphate levels in the later stages of CKD (Rees et al. 1989; Rigden et al. 1987), this led me to explore the preventative role of phosphate management in early CKD. On further reading, it became clear that much attention was paid to identifying FGF-23 as an early marker of abnormal phosphate metabolism (discussed in detail in section 3.1.2 and 3.1.3). FGF-23 holds the potential as a clinical therapeutic target. Identifying those patients in the early stages of CKD at the greatest risk of disease progression, who may benefit from a strategy such as the dietary control of phosphate (Kumar et al. 2012; Siomou and Stefanidis, 2012) to positively alter phosphate metabolism. Potentially delaying the associated complications of childhood CKD, such as poor growth and CKD-mineral bone disease (CKD-MBD) (Fernández-Iglesias et al. 2018; Hyoungnae et al. 2019), FGF-23 has been shown to have a direct positive correlation to disease progression. However, this important

emergent relationship between dietary phosphate intake and FGF-23 is under investigation (Ix et al. 2019), and remains unclear due to the complex interaction between FGF-23, the bones and other organs (Fukumoto, 2019).

It became clear that, whilst the scientific community moved forward with investigating the relationship between early dietary phosphate intake, FGF-23 and disease progression (Ix, 2019; Santamaria et al. 2018; Isakova et al. 2011), what remained was the need to explore how service users and providers view early phosphate management and its implementation in clinical practice.

These conversations evolved into a research proposal for a Masters of Arts in Research Methods (MARM) that I completed in 2015-16. I conducted a small empirical study which explored parents' views towards, and knowledge of dietary phosphate management in CYP with Stage 3 CKD in a specialist paediatric nephrology clinic. The key qualitative findings emphasised the "burden of care" of diet on the family, and its "disruptive" impact on family life, creating a constant undercurrent of anxiety. This was juxtaposed by a desire for relevant, tailored, accessible information to advance "knowledge to improve control" in the early stages of CKD, all within the context of trusted relations with professional staff (Pugh, 2016, unpublished thesis). This led to a debate with clinical peers regarding the next step – an exploration of the multiple stakeholder perspectives, namely CYP's, parents' and HCPs' perspectives on the development of an EPSM strategy for CYP with early stage CKD to delay disease progression. I secured a NIHR (National Institute of Health Research) CLAHRC (Collaboration for Leadership in Applied Health Research and Care) PhD studentship to complete this doctoral study. The next chapter provides the clinical context to the study, placing it within a biomedical and SM clinical framework.

1.2 Clinical context to the study

1.2.1 Chronic kidney disease

Chronic kidney disease has been defined by the global body, Kidney Disease Quality Outcome Initiative (KDOQI), as any kidney impairment causing a decrease in the glomerular filtration rate (GFR) for 3 months or more (Levey, 2005). Early abnormal signs leading to a diagnosis of CKD may include blood (haematuria) and protein (proteinuria) in the urine or elevated blood pressure (hypertension) (Byrne et al. 2018).

1.2.2 Classifying chronic kidney disease

Kidney function is measured using the GFR by assessing blood flow through the glomeruli per minute (Soveri et al. 2014). It is expressed as a rate corrected for the body surface area of the average adult ($1.73m^2$). Glomeruli are a mesh of capillaries in the kidneys that filter the blood as the first step in the nephrons' homeostatic elimination of waste function. Healthy kidneys have a GFR of >90 ml/min/$1.73m^2$ (Denic et al. 2016), therefore making it accessible to patients to consider GFR as a percentage kidney function. In CYP, the gold standard for measuring GFR involves the clearance of the complex sugar inulin from the kidneys by applying tests to timed urine samples (Schwartz and Work, 2009). In clinical practice other tracers are used, most commonly radioisotopes, such as ^{51}Cr-EDTA and more recently DTPA (Soveri et al. 2014). Such techniques involve repeated blood sampling after intravenous injection of tracer and are not suitable for frequent repeated measurements. An easier strong surrogate marker is to estimate GFR (eGFR) using the serum creatinine level and height (as a measure of muscle mass). The Schwartz formula defines eGFR as a constant multiplied by height and divided by serum creatinine (Schwartz et al. 1976).

1.2.3 Stages of chronic kidney disease

CKD is classified as stages 1-5, from mild kidney disease at stage 1 to severe at stage 5. In this study the exploration of stakeholder perspectives towards the development of an EPSM strategy is in relation to CYP with CKD stage 1-3b (Table 1, p. 6), shaded in green. During this early stage, there are no overt symptoms, or feelings of ill health, however underlying bone changes are taking place (Wesseling-Perry and Salusky, 2013).

Table 1. The stages of chronic kidney disease

CKD Stage	Treatment required	Description	GFR ml/min/1.73m^2
1	Kidney damage with normal or reduced GFR. Asymptomatic, but require annual blood pressure monitoring	Don't usually feel unwell	<90
2	Mildly reduced kidney function. Asymptomatic, but require routine blood pressure monitoring		89-60
3a	Moderately reduced kidney function. Likely asymptomatic, with possible early signs of fatigue and fluid retention		59-45
3b			45-30
4	Severely reduced kidney function. Biochemical changes, anaemia, fluid retention and hypertension may present. Preparing for established renal disease and dialysis	May feel tired and symptomatic	30-15
5	Established renal failure. Advancing of the above symptoms. Preparing for established renal disease and dialysis		<15
5d	Established renal failure. Receiving dialysis		Dialysis

Adapted from KDIGO (2013)

1.2.4 Disease progression

All kidney function will eventually deteriorate and today the parents of all CYP with CKD are given advice about the long-term outlook and the factors that can influence disease progression. The variable rate of decline in CKD function depends on the underlying disease (see Table 2).

Table 2. Causes of CKD in CYP

Some of the causes of CKD in descending order	
Aetiology	*Description*
Congenital anomalies of the kidney and urinary tract (CAKUT)	a group of abnormalities affecting the kidneys or other structures of the urinary tract
Reflux nephropathy	kidney damage due to urine flowing backward from the bladder towards the kidneys
Obstructive uropathy	structural or functional interference of normal urine flow
Glomerulonephritis	acute inflammation of the kidney due to an immune response
Hereditary nephropathy	a genetically varied disorder characterised by blood or protein in the urine, and hypertension
Cystic kidney disease	a range of hereditary developmental and acquired kidney conditions such as autosomal dominant polycystic kidney disease

Adapted from Harambat, J., Stralen, K, J, V., Kim, J, J. and Tizard, E, J. (2012).

Early intervention can delay disease progression, decreasing the risk of poor bone health and growth (Fernández-Iglesias et al. 2018), delaying the commencement of dialysis (Peng et al. 2019) and related cardiovascular complications (Hyoungnae et al. 2019). Additionally, early intervention offers many psycho-social benefits: an increased early understanding of the disease and treatment options provide clarity and guidance on the practical application of what may be viewed as difficult and often conflicting dietary advice (Lopez-Vargas et al. 2014). It promotes a positive attitude that can help patients to cope better, enjoy life, allaying anxieties and improving mental health (Lopez-Vargas et al. 2014; Ormandy, 2008; Havas et al. 2016a). Early intervention builds relationships with HCPs, fostering trust, building patients' and parents' confidence to ask questions, voice concerns and take control (Lopez-Vargas et al. 2014; Swallow et al. 2013; Williams and Manias, 2013; Clarkson and Robinson, 2010).

When people reach CKD stage 5 they need renal replacement therapy (RRT) (dialysis) or a kidney transplant. Globally, the prevalence of RRT in 0-19-year-olds ranges from 18 to 100 per million age-related population (pmarp) (Harambat et al. 2012). In the UK the prevalence of RRT in CYP (<16 years) is 60.4 pmarp (Hamilton et al. 2016). This may reflect early screening and improved access to treatment. By contrast, the prevalence in the United States is 85 pmarp, while much lower in Japan (34 pmarp) and Brazil (23 pmarp) (Harambat et al. 2012). CKD, with its many underlying causes (see Table 2, p. 6) is often identified antenatally and parents are educated about CKD when their child is newborn. The mean annual cost for a patient on dialysis in 2009-10 was £27,000, and for a patient who had been transplanted was £12,000 (Kerr, 2017). In stark contrast, the annual cost of providing conservative management, including dietary therapy, was £235 (Kerr, 2017); suggesting significant economic benefits as well as health benefits if CKD progression can be delayed.

1.2.5 Life expectancy

Globally, life expectancy for a healthyCYP is 72 years (WHO, 2018), and between 73.7-83.2 years in the UK depending on the location (Office for National Statistics, 2015). Comparatively, mortality rates in CYP receiving dialysis are 30 times higher (Chong et

al. 2017) than the age-matched general population (Office for National Statistics, 2018), with children <5 years having the highest risk of death, with the risk of death reported to be more than 120 times greater still in children under 1 year of age (McDonald and Craig, 2004; Mitsnefes et al. 2013). This increased dialysis related mortality reflects the mineral dysregulation causing vascular calcifications, which intensifies the progression of cardiovascular risk factors (Shroff, 2011). Expected *remaining* lifetime (years) for a 0-19 year old with CKD receiving dialysis is 19.3-24.1 years (Neild, 2017), equating to a life expectancy of approximately 22-40 years of age.

1.3 Dietary phosphate

Phosphorus which occurs most commonly in the salt phosphate (hereafter referred to as phosphate) is vital for life (Houston et al. 2013), and as a primary component of adenosine triphosphate (ATP) it is central to energy transport in all cells (Gandy, 2014). Bones expend 85% of the body's phosphate during skeletal mineralisation (Gandy, 2014), making phosphate crucial for the growing CYP. Organic phosphates, bound to carbon-containing molecules are naturally consumed in the form of meats, fish, dairy, vegetables, grains and nuts. Inorganic phosphate salts are appended to food in the form of additives and preservatives when food is processed (Eckberg et al. 2015).

Phosphate bioavailability

Bioavailability – the amount of organic and inorganic phosphates entering the circulation to have an active effect on the body varies considerably (Eckberg et al. 2015). Phosphate naturally occurring in plant-based foods, stored in the form of phytate or phytic acid, has a bioavailability of 20-50% (Noori et al. 2010). Since humans do not express the phytase enzyme required to break down phytate, this phosphate remains largely unavailable for absorption (Uribarri and Calvo, 2003). Fibre-rich food consumption (typically high in phytate) by CYP <18 years is significantly lower than recommended (Public Health England, 2018). Crucially, as plant-based foods have a low phosphate bioavailability but a nutrient-rich canvas of vitamins and minerals, they make an ideal contributor to the diet of individuals with early stage CKD (Gluba-Brzózka et al. 2017).

Protein-rich dairy produce, meat and fish provide an extensive supply of organic carbon-bound phosphate (Calvo et al. 2013). Due to its dependence on enzymatic digestion in the gastrointestinal tract to release phosphate from its carbon-bound element, absorption is slow and inefficient, yielding a moderate bioavailability of between 30-70% (D'Alessandro et al. 2015; Houston et al. 2013; Calvo et al. 2014). Since meat and dairy products are key contributors of high biological value proteins in the diet of CYP (Uribarri and Calvo, 2003), and are essential for growth, an adequate, but not excessive, intake during the early stages of CKD should be encouraged. High biological value protein foods are those containing many of the essential amino acids in a proportion similar to that required by the body (Hoffman and Flavo, 2004).

It is of significant concern that "free" - not organically bound or associated with phytate - phosphate salts added to processed foods as preservatives are rapidly and efficiently absorbed into the gastrointestinal tract with a bioavailability of up to 100% (Calvo et al. 2014; Ritz et al. 2012). Given the ubiquitous exposure of CYP to processed foods (Rauber et al. 2015) (see Table 3, p. 10), there is significant potential for excessive intake, making inorganic phosphate the main target for action in the battle to manage phosphate metabolism in CYP during the early stages of CKD (Evenepoel and Vervloet, 2016). Table 3, highlights the variable bioavailability of phosphate-rich foods. The traffic light code provides a guide to the food groups to be encouraged and discouraged in the early CKD diet.

Table 3. The bioavailability of a selection of phosphate-rich foods

Key: The traffic light colours reflect the least to the highest bioavailability

ORGANIC – Plant and plant derived	Bioavailability: Low	Early CKD diet
Seeds, beans, legumes, peas, peanuts, almonds, lentils, wholegrain bread and pasta, rice, cous cous, salad, fruit, vegetables	10-30%	Encourage in the diet
ORGANIC – Animal and dairy derived	**Bioavailability: moderate**	**Early CKD diet**
Chicken, lamb, pork, veal and ham with no preservatives, tuna fish, trout, cod, hake and sole. Soft cheese, meat and poultry (unprocessed), fish, egg yolk and egg white, milk	40-60%	Moderate intake, sufficient to support growth
Red meat, sausages, offal, salmon, shrimps and squid, hard cheeses	50-70%	Moderate intake, sufficient to support growth
INORGANIC – Additives in processed foods	**Bioavailability: high**	**Early CKD diet**
Bacon, deli meats, canned meat, canned fish, hot dogs, chicken nuggets, meat pies, processed cheese, instant puddings and sauces, refrigerated baked products e.g. cakes and muffins, ready to eat cereal, colas, fruit juices, liquid non-dairy creamer, flavoured milk, dry powered beverages and take-away or fast-foods.	100%	Significantly reduce intake

(Table 3 adapted from Ritz et al. 2012; Houston et al. 2013; Adema et al. 2014)

1.3.1 The need for early dietary phosphate self-management

When the kidneys are functioning well their efficient homeostatic mechanisms allow the body to use the dietary phosphate needed for health, any excess is excreted in urine (Kuro-o, 2013). If kidney function is impaired, their ability to effectively excrete phosphate deteriorates as the disease progresses (Kuro-o, 2013). Consuming a phosphate-rich diet during the early stages of CKD (1-3) stimulates both the regulation of serum phosphate by the kidneys and the release of the phosphaturic hormone FGF-23 from the bones (Sinha et al. 2012; Yoshiko et al. 2007). As the FGF-23 levels increase, phosphate excretion increases in the urine, suppressing the synthesis of 1, 25-dihydroxyvitamin D^3 (Calcitriol) and increasing release of parathyroid hormone (PTH) by the parathyroid glands (Sinha et al. 2012; Portale, 1984); resulting in a near neutral phosphate balance. This reflects a balance between the phosphate absorbed in the intestine and the amount excreted in the urine and

faeces (Houston et al. 2013), which over time triggers detrimental bone changes and the early signs of CKD-MBD. Raised FGF-23, an early marker of phosphate overload, is significantly associated with an increased risk of progression to established renal disease (ERD) (stage 5) in CYP (Portale et al. 2016) (discussed in more detail in section 3.1.2).

Reducing dietary phosphate in early CKD (1-3) is a possible therapeutic strategy to attenuate FGF-23 levels, potentially delaying disease progression (Siomou and Stefanidis, 2012; Portale et al. 2016; Kumar et al. 2012; Sinha et al. 2012; Komaba and Fukagawa, 2016). This study will focus on exploring the development of an EPSM strategy during the early stages of CKD 1-3, where bone changes take place earlier than dietary phosphate SM interventions are currently offered.

1.3.2 Specialist renal dietetic support

Phosphate management is complex and difficult (D'Alessandro et al. 2015). The NICE hyperphosphataemia guidelines (2013) recognise the need for the specialist paediatric renal dietitian to provide bespoke dietary advice when serum phosphate levels start to rise during CKD 4-5. There is currently no routine recognition for this specialist support during the early stages of CKD. Despite recognition of the need for proactive dietary phosphate education by adopting healthier lifestyle choices, specialist dietetic support is not readily available (National Kidney Foundation, 2015; Evenepoel and Vervloet, 2016). Failure to provide dietary phosphate intervention until the late stages may be a missed opportunity to delay disease progression (Evenepoel and Vervloet, 2016), hence the need to explore stakeholders' perspectives towards the development of an EPSM strategy.

1.3.3 Burden of care

Prescribing a specialised diet during the early stages of CKD may be met with resistance, adding to the 'burden of care' (Watson, 2014, p. 319), disrupting family life, and augmenting family stress (Geense et al. 2017). Similarly, encouraging an increased consumption of unprocessed, organic derived phosphate sources may be problematic for families in terms of cost, as well as food preparation skills and time

(Ritz et al. 2012; Geense et al. 2017). These important stakeholder views were explored during the qualitative interviews in this study.

1.4 Delivery of 'care close to home'

1.4.1 The political context

Under the leadership of Tony Blair (1997-2007), the New Labour Government in England made an explicit commitment to improving CYP services. Sir Ian Kennedy (2001), who was significantly influenced by the poor outcome of paediatric surgical cases in Bristol, compiled a significant report highlighting the desperate need to reform CYP services. The United Kingdom (UK) National Service Framework (NSF) for children, young people and maternity services published *Getting the Right Start* (2003), followed shortly by the Green Paper on children entitled *Every Child Matters* (2004) set out a vision to improve children's services for the future. The UK National Service Framework Standard 6 (2004, p. 4) identified a vision for the healthcare of sick children and their families that would see CYP receiving timely, high quality and effective care as close to home as possible; a vision the EMEESY network is striving to implement. The development and implementation of an EPSM strategy will support this vision, seeking to provide care close to home in the EMEESY shared-care clinics, or at a venue most convenient for the family. Promoting healthy-eating in childhood offers a strong start in life, and is a recognised tool for early prevention and management of chronic disease (*Five Year Forward View*, 2014; *NHS Next Steps,* 2017; *The NHS Long Term Plan*, 2019), affirming the focus of this study.

1.4.2 The EMEESY managed paediatric nephrology network

The Children's Kidney Network for the East Midlands, East of England, and South Yorkshire (EMEESY) was established in 2012. This managed paediatric nephrology network operates from a tertiary university children's hospital in the East Midlands, UK. It is one of 13 specialist centres for paediatric nephrology based in major cities across the UK. The EMEESY Network is aligned to models described in the two Royal College of Paediatrics and Child Health (RCPCH) documents: *Improving the standards of care of children with kidney disease through paediatric nephrology networks* (RCPCH,

2011) and Bringing networks to life (RCPCH, 2012) aimed at bringing specialist CKD care close to the patient's home. Through the establishment of formal links, EMEESY shared-care clinics take place in 20 local hospitals across the region. The network, serves a population of approximately 6 million (RCPCH, 2011).

A purposive sample of participants was recruited from across the three EMEESY regions (see Table 33, p. 205 for Delphi responders) by virtue of their suitability for the purpose of the study (see inclusion criteria 4.2.3 and 6.5.6). The majority were based in the East Midlands, within close proximity to the tertiary hospital. Children and young people living a distance from the tertiary centre receive care close to home by attending the EMEESY shared-care clinic at their local hospital (RCPCH, 2011; The NHS Long Term Plan, 2019). A visiting paediatric nephrologist, supported by the local paediatrician, routinely leads the clinic 1-3 monthly, with occasional support from a specialist renal dietitian and/or nurse. If families prefer regular access to the whole multi-disciplinary team, they are given the opportunity to attend the tertiary centre for routine appointments. This offer is frequently taken up when the CYP progress to more severe renal disease at CKD stage 4-5, leaving early-stage patients to make up a significant number of those attending shared-care clinics (see Table 4). Crucially, the lack of access in shared-care clinics to the specialist multi-professional renal team means the majority of early stage patients have limited opportunity to receive specialist renal dietary education. This study explores stakeholders' perspectives towards accessible educational platforms.

Table 4. The EMEESY network serves 20 local hospitals

East Midlands	8 tertiary/distric general hospitals
East of England	7 tertiary/district general hospitals
South Yorkshire	5 tertiary/district general hospitals

1.5 Self-management

1.5.1 Definition

Self-management is recognised by adults living with chronic disease (Richard and Shea, 2011), including CKD (Havas, 2016a; Havas, 2016b; Peng et al. 2019), and by CYP with CKD (Ferris et al. 2015) as an important and preferred method for managing the actual or potential daily impact of their chronic disease. Havas et al. (2018) argue that CKD SM for adults is about tailoring advice and support to their individual needs. Likewise, Modi et al. (2012) attested that SM involved CYP developing a wide range of attitudes, health behaviours and skills to help minimise the impact of their condition on all aspects of life for themselves and their families/caregivers. Saxby et al. (2019) recently affirmed the importance of CYP taking an active role in their SM by mapping SM to the child's age and developmental milestones.

In the literature SM is often used synonymously with or as a subset of self-care (Lorig and Holman, 2003). Self-care refers more generally to the actions taken by individuals or communities to achieve, preserve or promote optimal health (Lipson and Steiger, 1996), whereas SM is specific to chronic disease management (Lorig and Holman, 2003). In this study, the term SM is used to capture the complex nature of chronic disease management, in particular the desire for and application of dietary-specific knowledge, which is well recognised across the SM literature (Ormandy and Hulme, 2013; Costantini et al. 2008; Griva et al. 2012; Curtin et al. 2004; Lewis et al. 2010; Ormandy, 2008; and Lopez-Vargas et al. 2014).

1.5.2 When is it best to engage CYP in self-management?

Although most of the CKD SM literature has focused on adults (Bonner et al. 2014; Havas et al. 2016a; Choi and Lee, 2012; Peng et al. 2019), chronic disease SM is crucial for CYP to embrace, making them partners in disease management (Modi et al. 2012; Ferris et al. 2015; Saxby et al. 2019; Huby et al. 2016), and supporting their transition to adult-care (Nightingale et al. 2019). In fact, CYP want their expertise in their own condition to be recognised, and often report feeling left out when it comes to shaping the services they access (Ormandy and Hulme, 2013; Farrant and Watson, 2004; Zack et al. 2003).

Additionally, as young people are vulnerable to poor health outcomes (Ferris et al. 2015), increased ownership and involvement in their SM is associated with better health outcomes (Ellis et al. 2005; McPherson et al. 2006). Unlike with young children who generally accept parental requests around health, young people will frequently not be dictated to (Sawyer and Aroni, 2005), exhibiting denial, depression, low literacy, poor coping strategies and a poor understanding of their condition (Ferris et al. 2015). But as young people mature, and are encouraged and supported, they learn to take on more responsibility and become more involved in their own SM (Coleman et al. 2009; Ferris et al. 2015).

Based on Piaget's theory of cognitive development (1925), adolescence is arguably characterised by risk-taking and increased abstract thinking. He proposes that cognition develops over the life cycle in response to biological maturation, peer influence and the environment. Piaget believes these influences create a tendency for young people to focus on the present, and are not readily persuaded to consider the long-term health risks; preferring to choose a high phosphate-containing cola-based drink for example, rather than lemonade, or better still, water. By contrast, Erikson's theory of psychosocial development (Munley, 1975) proposes that progress occurs in response to conflict, with the key conflicts arising during adolescence. As conflict times are navigated successfully, virtues associated with these periods are established. Neurological development during adolescence is different to at any other time in the life cycle (Blakemore et al. 2010), providing insight to the complexity and importance of peer influence, risk taking and poor impulse control during this period.

Anticipating future events or consequences not personally experienced diverges from their risk-taking tendencies. This conflicts with the goals of SM, which tries to be proactive or pre-empt untoward events. As a result, problems with SM typically occur both during the early stages of an illness and during early adolescence (14-16 years) (Urbach et al. 2005), making this an important time to engage both CYP and families in education and skills training strategies. This supports the rationale for including adolescents in addition to other age ranges in this study.

With evolving skills and the slow maturation of the cognitive control system (McRae et al. 2012), CYP become increasingly able to appraise their emotional response and defer immediate gratification in the light of the future impact. In partnership with the CYP and family, HCPs need to develop strategies that recognise the developmental level of the CYP and determine their readiness to engage in SM (Ferris et al. 2015; Nightingale et al. 2019). Together, they should seek to nurture the CYP to become their own best advocates. CYP's risk taking characteristics and vulnerability to poor health outcomes (Ferris et al. 2015) further strengthen the rationale in this study for exploring the development of an EPSM strategy in this group with early CKD.

1.5.3 Theoretical framework

Lorig et al.'s (2001) US (United States) Chronic Disease SM Programme, adapted as the Expert Patient Programme (Roger et al. 2008) in the UK, provides the theoretical framework for this study (see Table 5). Prochaska's (1979) model of change provided important holistic steps that could be adapted to support changes in lifestyle and eating behaviour, and Bronfenbrenner (1977) captured the evolving complexity of structures and agencies required to support change across the life span.

Table 5. Self-management skills (Lorig et al. 2003) as applied in this doctoral study

Core self-management skills	Explained	Application in this study
Problem-solving	Recognition of the problem(s) involves defining the problem(s) and upskilled to problem-solve	• Defining EPI • Recognise where dietary upskilling is required e.g. cookery workshops • Exploration of when, where and how dietary education and support is best delivered • Identify enablers that will empower CYP and families to seek advice and support
Decision-making	Equipped with the knowledge to make informed decisions	• Recognise when and where dietary education and support is best delivered • Explore how much and the best mode to deliver information
Resource utilisation	Skilled to find and utilise resources	• Recognise the value of early dietary education and support to build confidence, voice concerns and seek advice • No recognition of community resources and health promotion programs
Development of patient-HCP partnership	Developing a partnership with healthcare providers	• Exploration of CYP and parents desire to receive dietary advice/support during the early stages of CKD • Exploration of the HCP views towards the provision of early dietary education/support • Exploration of how and when patient-HCP partnership can be built
Action planning	Learning how to change a behaviour	• Recognise the importance of simple, clear dietary education, 'burden of care' and other challenges to EPI • Explore stakeholders' views on changing behaviour

However, it was necessary to identify a framework that recognised the multifaceted, dynamic and evolving nature of chronic disease. Recognised in the CKD SM literature (Welch et al. 2015; Sorat, 2018), Lorig et al.'s (2001) five core SM tenets provide a holistic framework that can be tailored to the individual needs of CYP living with all stages of CKD. In particular, it focuses on supporting the development of an individual's knowledge and skills in order to share power with the HCPs over the management of their disease. These three important components: education, skills and support are directly addressed in this study. Table 5, p.16 details the application of these five SM tenets (Lorig et al. 2003) to this study. These five core SM tenets are illuminated in the integration of the findings (Chapter 8). Lorig et al. (2003) gives credence to Bandura's social cognitive theory of behaviour (1989) as a significant underpinning to SM strategies. Self-efficacy (Bandura, 1997) recognises a person's belief in their ability to plan and carry through with an action to achieve a change in behaviour. This important facet of SM underpinned the themes that developed during the integration of the MMR findings.

1.5.4 Child and young person-centred approach

The *Expert Patients*' *Programme* (Tattersall, 2002) and the ten-year plan set out in the *Children's National Services Framework* (DoH, 2004) focused on championing the child's own views (Barlow and Ellard, 2004). Although this vision is yet to be fully realised, strongly affirmed by the NHS Youth Forum in their disappointment 'that we weren't highlighted as a specific group' (NHS Youth Forum, 2016) in the *Six principles for engaging people and communities* (2016) document, momentum is building. As the NHS seeks to respond to the changing and evolving complexity of individual's health needs, early intervention and preventative action in childhood is becoming a priority (The *NHS Long Term Plan*, 2019). Guided by the NIHR INVOLVE standards for CYPs (2016), the voices of CYP with long-term chronic conditions are being listened to, and their involvement in research is informing the development, delivery and commissioning of local and national services (*Healthy Children: Transforming Child Health Information,* NHS England, 2016; Lambert et al. 2013; RCPCH Voice Matters, 2020). *National Voices,* a coalition of charities, to champion the individuals' health and care values and preferences, is testament to the vision of a child and young person-

centred approach being realised. For example, using a range of new training and online resources, Martin (2016) as part of the *Me first* project, in her Twitter based Common Room, sought to improve communication between CYP and HCPs, placing them at the centre of the conversation about their health and healthcare. The Young People 4 Change (YP4C) forum, which started as an initiative in the children's kidney unit at Nottingham Children's Hospital, has become a national example of how young people are shaping health services (GIFT nd).

The United Nations' *Convention on the Rights of the Child* (UNCRC) (1989) recognises participation in matters that affect children's future health as an important component of a child's right to protection and well-being. Indeed, many HCPs understand the importance of involving CYP in decision-making about their treatment and service provision, especially those with a chronic disease (McPherson, 2010). The active involvement of CYP in this study recognises their crucial participation in healthcare decisions (Greig et al. 2017; Söderbäck et al. 2011), to foster a sense of control and confidence when following the EPSM diet and lifestyle. Additionally, the researcher is facilitating partnership building, and 'attempting to see the world through their [CYP] eyes' (DoH, 2004, p. 87), rather than just engaging in rhetoric (Coad and Shaw, 2008).

1.6 Summary

This study was conceived from real-life conversations with parents of CYP with CKD, seeking ways to slow down the progression of their CYP's disease. Self-management is vital to the management of CKD, and dietary issues and phosphate control are becoming an important emergent issue in the early stages of CKD. Children and young people with CKD should be encouraged and supported to take an active age-appropriate role in the SM of their chronic disease. Developing an EPSM strategy for CYP with CKD stages 1-3, during a period when bone changes are taking place but dietary support to manage phosphate intake is not currently routinely available, may delay disease progression. The next chapter outlines the development of the research aim and objectives, and the three-phase research plan, which are original in their content and design.

Chapter 2: Research aim and objectives, design and methodology

2.1 Introduction

This chapter first presents the research aims and objectives, before examining the ontological and epistemological underpinnings of the study. Next, the mixed methods research design and the methodology of interpretive description that was employed to address the research question are presented.

2.2 Aim and objectives of the research

2.2.1 Research aim

In order to answer the research question:

'What are the perspectives of children and young people, parents and key health care professionals towards the development of an early dietary phosphate SM strategy, to delay disease progression for children and young people with CKD stages 1-3, within a managed paediatric renal network?,' the primary aim of the research was to:

> Explore the perspectives of children and young people, parents and key healthcare professionals towards the development of an early dietary phosphate self-management strategy, to delay disease progression for children with CKD stages 1-3, within a managed paediatric renal network

2.2.2 Research objectives

This aim was addressed through the following seven research objectives:

1. To achieve a consensus definition for Early Phosphate Intervention (EPI)
2. To explore and gain consensus on how we should deliver the dietary phosphate education, skills and support to CYP and their families
3. To explore and gain consensus on who should deliver the EPI education, skills and support

4. To explore and gain consensus on who should receive the EPI strategy
5. To explore and gain consensus on when the EPI education and support should be delivered
6. To explore and gain consensus on where the EPI education, skills and support should be delivered
7. To identify the challenges and enablers to implementing an EPI strategy

2.2.3 The research plan

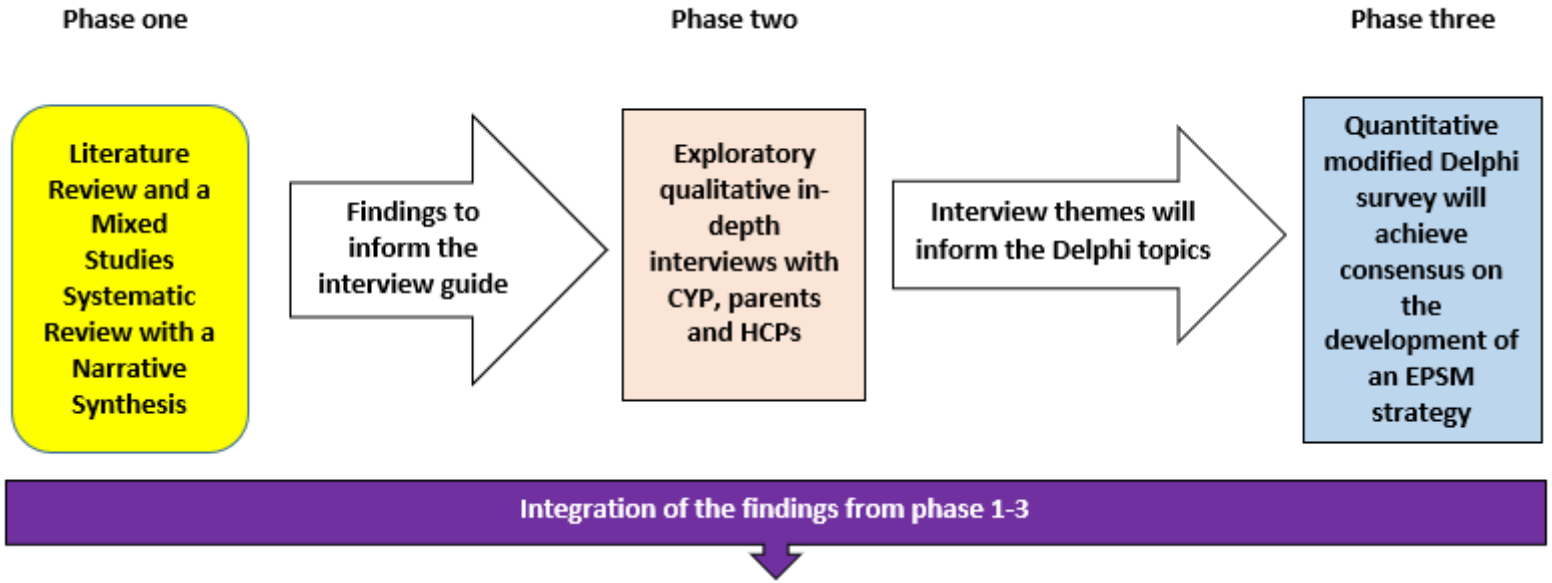
With the goal of exploring the perspective of CYP, parents and HCPs towards the development of an EPSM strategy for CYP with CKD stage 1-3b; this doctoral research involved a three-phase study.

Phase 1 – a mixed studies systematic review with a narrative synthesis systematically explored the literature on the CYP, parents and HCPs views concerning the use of diet as a tool for the management of early chronic disease. The results and recommendations from the systematic review informed the interview guide in Phase 2.

Phase 2 – semi-structured interviews were undertaken with CYP, parents and HCPs. Using the methodology of interpretive description to guide the data analysis (refer to section 4.5), qualitative themes were developed from the interviews. The integration of Phase 1 and 2 findings informed the development of Delphi round 1 consensus survey in Phase 3.

Phase 3 - over three consecutive Delphi rounds the expert panel of parents and HCP built consensus on the issues they felt were most important when planning the development of an EPSM strategy. The integration of the findings from Phase 1, 2 and 3 produced 3 novel MMR themes, recommendations for practice development with the essential groundwork and the emergence of some early modelling for the EPSM strategy. Refer to Figure 2, p. 21 for the MMR design.

Figure 2. Mixed-methods research design



When undertaking healthcare research, the underlying ontological and epistemological assumptions must be clear, as this informs the aggregation of information and analytical methods that best serve the research question (Carter and Little, 2007).

2.3 Ontology and epistemology

Ontology, a branch of metaphysics, describes the ultimate nature of things as they are and what constitutes social reality (Rawnsely, 1998). This provides an understanding of whether social reality is external to social actors or whether they are intrinsically engaged in the process of creating it (Bryman, 2016). By contrast, epistemology seeks to understand how knowledge is produced, acquired and communicated, especially how the researcher chooses to approach the knowledge (Bryman, 2016). The researcher's ontological and epistemological assumptions will inform the choice of methodology and methods that best serve to answer the research aims and objectives as detailed in section 2.2.1 and 2.2.2 (Carter and Little, 2007).

Ontology

Healthcare research has traditionally chosen a positivistic approach, employing quantitative methods alone (Muncey, 2009), with the emergence and increasing appreciation of the need for realignment focusing not only on the observable, but also on the human interpretation and meaning behind social actions (Snape and Spencer, 2003).

Positivism's ontological stance is realism, which asserts that 'social facts' are 'out there' and have an existence autonomous from the participant and researcher activity (Silverman, 2014, p. 23; Scotland, 2012). This stance places an objective requirement upon the researcher to describe and explain the phenomenon in as neutral and objective a way possible (Justesen and Mik-Meyer, 2012). By contrast, the ontological stance of interpretivism is relativism, which seeks to embrace the various ways individuals interpret reality (Guba and Lincoln, 1994), recognising that an individual incrementally constructs their own reality of the world through subjective engagement with social systems, language, and concepts in their world (Ross, 2012). Through her extensive clinical experience (refer to section 1.1.1) the researcher

recognised the value of the objective clinical stance, while esteeming the importance and uniqueness of the individual's perspective in managing CKD. If the findings of the study are to improve clinical practice, it was essential to capture the service user and providers' perspectives, which directly informed her need to take an interpretivist's ontological view point.

Epistemology

Objectivism underpins the positivist epistemology, attempting to discover absolute knowledge of an objective reality that resides not in the consciousness of the researched or the researcher, but solely in objects (Scotland, 2012; Crotty, 1998). The researcher recognises the importance of objective markers such as clinical reference ranges, and diagnostic pathways to direct CKD management. By contrast, the interpretivist epistemology is subjectivism based on participants 'real world' experiences (Scotland, 2012), thus the researcher sought to capture the CKD stakeholder's distinctive viewpoint and experience. Allowing each individual to interpret phenomena uniquely produced multiple realities, each 'valid' and 'truthful' in their own right (Ross, 2012, p.86). During the study the researcher was conscious that a transformative process was taking place at every juncture of interpretation, as the individual submitted their own variation of events through the lens of their own worldview. Gudmundsdottir (1996 p. 304) related this process of transformation to the philosophical image of 'an endless hall of faulty mirrors'. These paradigms of inquiry – objectivism and interpretivism – have traditionally been linked with quantitative and qualitative research methodologies in healthcare studies (Mesel, 2013).

In order to improve clinical practice the researcher recognised the need to harness both the positivist and interpretivist viewpoint. Methodological pluralism and pragmatism which is engendered in this doctoral study are gaining acceptance by researchers (Webb et al. 2016; Mesel, 2013).

2.3.1 Pragmatism

Pragmatism is a paradigm built on the premise that the best way to judge a piece of knowledge is to determine how useful it is to solve a given problem (Muncey, 2009).

William James, one of pragmatism's early proponents, expounded this idea when he coined the term "cash-value," elevating the notion that a hypothesis is merely worth its practical usefulness. He clarified,

> *"Pragmatism unstiffens all our theories, limbers them up and sets each one at work"*
>
> *(James, 1975, p. 27)*

Instead of pursuing a finding that closely mirrors reality or truth, pragmatism aims to use "what works" in order to seek answers to the research question, thus providing "real-world practice with real-world evidence" (Neta and Johnson, 2018; Creswell and Piano Clark, 2011). This approach aligns with the fundamentals of interpretive description (see section 2.5), the chosen methodology for this doctoral study, which values the subjective and experiential knowledge of the researched and the researcher, seeking to capture 'as naturalistic a context as possible' (Thorne, 2008, p. 74).

Pragmatism sees the research problem as the most important, valuing the use of multiple perspectives (Creswell and Piano Clark, 2011), and esteeming both the positivist and interpretivist paradigm in order to gain a practical understanding of everyday experiences to reveal the answers (Tashakkori and Teddlie, 2010; Muncey, 2009). Therefore, the paradigm of pragmatism, with its pluralistic and action-driven nature will underpin this mixed methods research, utilising the strengths of both qualitative and quantitative research methods (Tashakkori and Teddlie, 2010). The paradigmatic principles of pragmatism align with the ethos of this study, and its aims and objectives (see 2.2.1 and 2.2.2) designed to explore (qualitative) and gain consensus (qualitative and quantitative) on the development of an EPSM strategy to delay disease progression by eliciting the multiple perspectives of key stakeholders involved in the real-world experience of managing CYP with CKD.

As an experienced practitioner, with a drive to improve clinical practice, pragmatism aligns with the researcher's own world view and view of knowledge, allowing the findings and application of the study to become more contextually relevant and meaningful to practice.

Using the worldview of pragmatism as an underpinning for the entire study, the researcher drew upon the subjective and objective as necessary in order to use "what

works" to answer the research question (Creswell and Plano Clark, 2011). The viewpoint utilised during each phase of the study (refer to 2.2.3) will now be explained.

Despite lacking an ontological or epistemological stance (Schryen et al. 2015), Phase 1 of this doctoral research involved a systematic review of the evidence to "first understand the literature in the field", to determine what has already been done and how to learn from it (Boote and Beile, 2005, p. 3).

Underpinned by an interpretivist viewpoint, Phase 2 of the study, which involved semi-structured interviews with CYP, parents and HCPs, explored their perspectives towards the development of an EPSM strategy. The in-depth qualitative interview is a valuable and powerful tool and a recognised feature of the pragmatic paradigm (Farquhar et al. 2011; Johnson et al. 2007), enabling the researcher to probe, follow up questions to comprehend the participants perspective, get closer to what participants would actually like to see happening in clinical practice and discover an actionable solution to the problem.

The modified Delphi employed in Phase 3 of the study using an objective single statistical measure to grade the expert panels' level of agreement (LOA) is representative of the positivist paradigm (Keeney et al. 2011). By contrast, the acquisition of panellists' subjective free-text comments on the Delphi statements during rounds 1-3, allowed the researcher to utilise an interpretative stance (Engles and Kennedy, 2007); the 'ongoing iterative dance of discovery and interpretation' that ensues as individual's feedback on group opinions, with the opportunity for respondents to change their views, embraces the social constructivism viewpoint (Miller and Crabtree, 1992, p. 11).

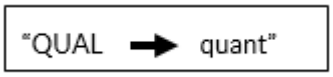
2.4 Mixed-methods research

The rationale is now provided for using mixed-methods. The origins of MMR go back to the ancient Western philosophers Plato and Protagoras. Plato argued for a universal approach to viewing the world (quantitative), whereas Protagoras saw multiple truths (qualitative) (Johnson et al. 2007). Mixed-methods research attempts to fully respect the wisdom of both of these viewpoints. It adopts a research design in

which one or more quantitative or qualitative methodological strategies access some part of the phenomena of interest, and integrates it into a single study to provide a better understanding of a research problem than could be provided by the use of one approach alone (Morse and Niehaus, 2009).

2.4.1 The chosen mixed-methods design

In order to address the research question, aims and objectives (see section 2.2.1 and 2.2.2), which focused on the exploration of stakeholders perspectives and building consensus, qualitative methods constituted the dominant paradigm in this study, with quantitative methods less dominant. Using the notation suggested by Morse (1991), the research design for this study can be abbreviated as:



The upper and lower case distinguish between the dominant and less dominant methods to be used in the study, and the arrow indicates the sequence and direction of the planned investigation procedures. Table 6, p. 27 provides an example of this mixed–methods design typology (shaded in blue).

Table 6. Mixed-methods designs

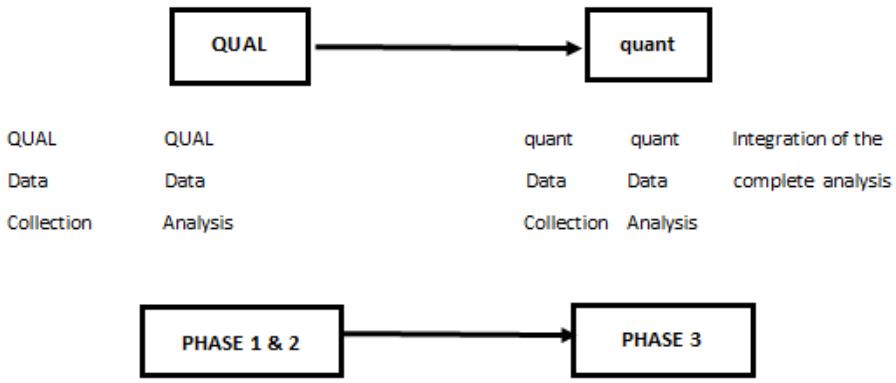
Research design	Process	Purpose	Level of interaction	Priority	Example from healthcare literature
Convergent parallel (simultaneous)	Qual Quant	Different but complimentary data is collected to address a single research question	Data is collected and analysed independently	Equal	Bussing *et al.* (2012) simultaneously collected data eliciting ADHD treatment perceptions from four stakeholder groups using a postal survey. The inferential statistical and qualitative data from the open-ended survey responses were analysed separately.
Sequential explanatory	QUANT → qual	The qualitative data gives greater explanation to the quantitative findings	Quant data frames qualitative data collection	Quantitative data is dominant	Jellesmark *et al.* (2012) used face-to-face survey to assess the fear of falling amongst community-dwelling elderly people, followed by in-depth interviews with 4 participants.
Sequential exploratory	QUAL → quant	The qualitative data informs the quantitative data to strength generalizability and confidence in the findings	Qualitative data informs quantitative data collection	Qualitative data is dominant	Barber *et al.* (2011) used two-rounds of a Delphi survey, which informed the follow up interviews to investigate the feasibility of evaluating the impact of public involvement on health and social research.
Embedded/nested	Qual (quant) Or Quant (qual)	This approach is used to collect different data to answer a complementary research question	The data sets are embedded	Quantitative or qualitative can be dominant	Grocke *et al.* (2014) determined whether group music therapy positively affected the quality of life of participants with severe mental illness. Qualitative and quantitative data were generated during focus groups and song lyric analysis.

Table 6 was adapted from Halcomb and Hickman (2015).

2.4.2 Mixed-method design typologies

The literature reports various typologies of mixed-methods designs (Farquhar et al. 2011; Halcomb and Hickman, 2015). 7 provides examples of these, and their use in the healthcare literature. The mixed–method research design and sequential modelling for this doctoral study is detailed in Figure 2, p. 21 and Figure 3.

Figure 3. Schematic of Sequential Exploratory Mixed-Methods Research Design (adapted from Creswell et al. 2003)



As informed by Halcomb and Hickman (2015) there are four main characteristics which define these designs and give rationale to the chosen design:

Interaction – the extent to which the qualitative and quantitative data interact (or not). The chosen design, shaded in blue (in Table 6, p. 27) saw the data interact at several junctions in the study (refer to Figure 2, p. 21).

Implementation sequence – the concurrent or sequential collection of qualitative and quantitative data. The former is resource intensive but can reduce the time required for data collection (Halcomb and Hickman, 2015). The latter, the sequential approach, which was applied in this study, involved sequentially collecting the qualitative and quantitative data separately; with the benefit that findings from the systematic review informed the semi-structured interviews, which informed the statements in round 1 of the Delphi survey. A disadvantage of the sequential approach is the longer time needed to complete data collection (Halcomb and Andrew, 2009).

Relative priority - the relative priority given to the qualitative and quantitative data is usually determined by the exploratory or explanatory nature of the study. Both the qualitative and quantitative methods are essential to the study, as the insights from one complement the other, but the CAPITALISED component of the study indicates the principal method for data collection (see Table 6, p. 27). Exploratory designs, such as this doctoral study, where a key objective was to explore stakeholder perspectives, privilege the qualitative data collection component. Priority was given to the qualitative (QUALS) component to uphold the exploratory elicitation of the stakeholder perspectives, to inform the statements in the round 1 Delphi survey. The explanatory designs, which seek affirmatory data often prioritise the quantitative data (Andrew and Halcomb, 2009).

Integration - the integration or 'conversation between the qualitative and quantitative components', considered to be the most important aspect of any mixed-methods study, as this is where the two approaches become 'greater than the sum of the parts' (O'Cathain et al. 2010; Farquahar et al. 2011), can occur at any point in the study (Andrew and Halcomb, 2009). Integration ideally occurs during the data collection, analysis, interpretation and discussion of the findings (Andrew and Halcomb, 2009); and can take three possible mutually inclusive approaches in health services research: integration, connection or embedding (Zhang and Creswell, 2013). Integration took place at the end of each phase of this doctoral study (see section 3.12 and 6.7.7), particularly during the integration and discussion of the findings (Chapter 8 and 9). Connection, as applied to this doctoral study, involved the 'mixing' of data from early in the study, with the findings from one phase informing the next phase of the study (see Figure 2, p. 21), and the final connection occurred when all of the phases were brought together in the integration of the findings in Chapter 8 (Zhang and Creswell, 2013). Lastly, embedding involves the analysis of one set of data embedded within another (Zhang and Creswell, 2013), which was not used in this doctoral study.

As an alternative to a MMR design, a qualitative research design was considered, utilising face-to-face semi-structured interviews or focus groups. The challenges of the latter method are highlighted in section 4.5.2. Additionally, as the core focus of

the study comprised the exploration of stakeholders' perspectives, and the achievement of consensus on the 'how', 'who', 'when' and 'where' facets, and challenges to the development of an EPSM strategy, the research required both qualitative and quantitative research techniques. A method was sought which brought together a geographically disparate group of expert stakeholders' differential perspectives to drive a consensus process, with an objective statistical measure to grade their LOA. Before deciding on the Delphi survey, focus groups were considered as detailed in section 4.5.2. The doctoral study inductively utilised the literature (Phase 1) and CYP, parents and HCPs perspectives (Phase 2) to shape the original statements in round 1 of the modified Delphi. This required the integration of qualitative and quantitative findings, and this provided a sound basis for using a MMR design for this doctoral study, with the pragmatic underpinning.

This doctoral mixed-methods design took advantage of both qualitative and quantitative research methods and paradigms to access the key stakeholders' multiple viewpoints. This provided a more detailed understanding of the problem, with a comprehensive 'yield' unavailable to an independent qualitative or quantitative study, and an output that is 'a whole greater than the sum of the parts' (Tashakkori and Teddlie, 1998; O'Cathain et al. 2007).

This need for multiple viewpoints, with in-depth understanding, sits well within the complex intervention development framework (Farquahar et al. 2011; Campbell et al. 2000) (see section 4.1.1), informing the process of development, through a greater knowledge of the 'active ingredient' (Hoffman et al. 2014), and identification of the potential enablers and challenges to implementation.

2.4.3 Rationale for using the mixed-methods design

As informed by Greene (2007), some of the key reasons for using this MMR research design include:

Corroboration - the findings of CYP, parents and HCPs perspectives, elicited during the semi-structured interviews were corroborated (triangulation) (Morse, 1991) with the free-text comments and statements reaching consensus during the Delphi survey. This increased the confidence in the conclusions that were reached.

Development - the results of one method informed another.

Instrument development - the findings of the systematic review informed the development of the interview guide employed to support the shape and flow of the interview.

Sampling - the parents and HCPs recruited for the interviews facilitated sampling for the Delphi survey.

Expansion - the semi-structured interviews/dyads (see section 4.5.3) elicited the perspective of a specific group of participants (CYP), expanding the depth and breadth of the study.

2.5 Interpretive Description methodology

The dominant exploratory focus of this MMR design required a qualitative methodology that would allow access to real-world practice (Thorne, 2008), while harnessing the researcher's positionality and expertise. Interpretive description (ID), an applied qualitative research methodology that uses an inductive analytical approach to help researchers understand human health and individual's experience of managing disease and the consequences for the clinical practice (Thorne, 1997), provided this tool. Conventional content analysis was considered as an alternative to ID, and rejected as discussed in section 6.6.2. A key reason for choosing ID reflected the inspiration behind the study (see section 1.1.1) and the researcher's positionality. With over 23 years of experience as a dietitian, working with CYP, parents and HCPs in paediatric nephrology for the past 18 years, it was important to capture this wealth of insight and experience, allowing and enabling its lens to challenge, guide and shape the qualitative findings in this study. ID encourages the researcher to be transparent about her positionality and *a priori* knowledge and experience (Thorne, 2008). ID embraces this "inseparable relationship", acknowledging that as these 'way[s] of thinking' guided the research process (Thorne, 2013), and influenced interactions with the researched, the findings and the application of the study become more contextually relevant and meaningful to practice (Thorne, 2008). Mindful of her positionality, when approaching study participants she did so as a researcher, and not as an expert practitioner (Thorne, 2103). Although theoretical models are drawn upon

throughout the research process (Modi et al. 2012; Lorig et al. 2001), they did not drive the analysis and interpretation of the data. Rather, the researcher's professional viewpoint or 'way of thinking' guided the process (Thorne, 2013). This made the findings and the application of the study more contextually relevant and meaningful to practice, allowing new knowledge to be turned into action (Thorne, 2008). For detailed application of ID in this study, refer to the section on data analysis in 4.6.

For the following reasons, the foundational underpinnings of ID align to those of pragmatism and MMR:

Allied to the "real-world practice" of pragmatism (Neta and Johnson, 2018; Scotland, 2012) ID has a desire to capture 'as naturalistic a context as possible' (Thorne, 2008, p. 74), explicitly valuing the subjective and experiential knowledge of the researched and the researcher as a fundamental source of clinical insight. This insight holds a 'practice mandate' with a pragmatic obligation to ensure the findings are 'usable' in clinical practice (Thorne, 2008, p. 227). ID, with its prerequisite to capture 'individual expressions of variance' mirrors pragmatism's desire to view a problem from multiple perspectives (Creswell and Piano Clark, 2011), by using the relevant methods to access the phenomena of interest (Morse and Niehaus, 2009). Likewise, just as pragmatism esteems the interpretivist stance in order to gain a practical understanding of everyday experiences to reveal answers (Tashakkori and Teddlie, 2010; Andrew and Halcomb, 2009), ID embraces the socially 'constructed' elements of human experience and recognises that they cannot be meaningfully separated. Rather, every 'reality' consists of multiple 'constructed realities' (Thorne, 2008, p. 74).

Originally designed for qualitative nursing researchers, ID has evolved through its application to MMR research by a variety of HCPs. Refer to Table 7, p. 33 for examples of ID in the healthcare literature.

Table 7. Examples of ID in the healthcare literature

Author	Study context
Oliver & Charles (2016)	ID was used in their mixed-methods study to strengthen child protection relationships.
Clark *et al.* (2011)	Sports scientists used ID in their mixed-methods to explore adolescent girl's experience of physical activity.
Ampofo-Addo *et al.* (2016)	An ID approach was used to understand family physician's location decisions.
Wong Riff *et al.* (2017)	ID was used in the development of a complex tool for children and young people with cleft lip and/or palate.

Similarly, in this study, ID will provide a tool to access the subjective, experiential knowledge of CYP, parents' and HCPs perspectives towards the development of an EPSM strategy.

2.6 Summary

This chapter has presented the ontological and epistemological underpinnings for healthcare research, with justification for the selection of the pragmatist paradigm for this study. Strongly aligned with pragmatism and interpretive description methodology, a mixed-method research design was asserted to be the best approach to solving the research question. Chapter 3 presents Phase 1 of the study – a llitertaure review, which informed a mixed studies systematic review with a narrative synthesis.

Chapter 3: Phase 1 - Literature review and a mixed-studies systematic review with a narrative synthesis

3.1 Introduction

This chapter begins by presentings the findings of an initial scoping search of stakeholders' perspective on the use of an early dietary phosphate SM strategy in children with CKD; identifing gaps in the literature, which informed a mixed-studies systematic review with a narrative synthesis. The chapter then moves into the rationale for conducting the systematic review, providing an outline of the systematic review research question. This is followed by a detailed account of how the systematic review was conducted, including the search strategy and selection criteria, and a comprehensive description of the narrative synthesis. The chapter concludes with the first 'integration' point (Box 3, p. 94), where key recommendations informed the development of the Phase 2 interview guides for CYP, parents and HCPs.

3.1.1 Search strategy

Informed by the methods of Levac et al. (2010), and Arksey and O'Malley (2005), a scoping search of existing literature was mapped to inform a systematic review. The scoping search question directed the identification and selection of relevant literature, manual collation of the printed studies, and a summary of the findings. Additional relevant papers were identified by searching the reference lists, hand searching and database alerts.

Scoping search question:

What are the perspectives of children, parents and other stakeholders towards the use of an early dietary phosphate SM intervention, to delay disease progression for children aged 0-16 years with early stage CKD?

An initial specific scoping search of Medline, CINAHL, COCHRANE, EMBASE, Scopus, PsycINFO and OpenGrey identified literature relevant to the research question. The full-text copies of relevant studies were printed, and manually collated into the five categories as detailed overleaf in Table 8, p. 35.

Table 8. The categories and number of studies identified in the scoping search

Topic	Number of studies
FGF-23 and CKD progression in children	12
Dietary phosphate restriction and FGF-23	23
Clinical practice guidelines on CKD stages 1-3b and stakeholder involvement in guideline development	19
The patient's perspective on CKD self-management	30
The HCP's understanding of self-management	27

The evidence for each category is presented in turn below.

3.1.2 FGF-23 and disease progression in CYP

Plasma FGF-23 levels, an early marker of phosphate overload, increases during the early stages of CKD in CYP, and progressively increases as GFR declines (Portale et al. 2016; Santamaria et al. 2018). There is a paucity of published data on FGF-23 in the paediatric population with CKD. In the largest known paediatric longitudinal study to date with early and pre-dialysis CKD, Portale et al. (2016) found FGF-23 to be a self-governing risk factor for CKD progression. Portale et al. looked at deterioration of renal function in a sub-sample of children (n=419) enrolled for the Chronic Kidney Disease in Children (CKiD) (Warady et al. 2015) prospective observational cohort study, between 2005-2009, who had baseline measures of plasma FGF-23, PTH and vitamin D metabolites. Portale et al's (2016) study (Table 9) demonstrated a mean plasma c-terminal FGF-23 (cFGF-23) of 132 RU/ml in children with early stage CKD (median GFR 44ml/min 1.73m^2, CKD 3), which is 2.5 x those found in healthy children (Gkentzi et al. 2014).

Table 9. Key data from Portale et al. (2016) study

Author	Baseline GFR (median)	Age (years)	Plasma cFGF-23 (mean)
Portale *et al.* (2016) n=419	44ml/min/1.73m^2 (IQR 33-57) 82% CKD 1-3	1-16	132 RU/ml (IQR 88-200) 2.5 x normal level 65% had cFGF-23 >normal upper limit

There were no differences in age, gender, background disease and GFR between those included in the sub-sample and those not. Progression of GFR over a 5.5-year follow up period demonstrated that 32.5% of the children reached an end-point of end-stage renal disease (ESRD) (commencement of dialysis) or a 50% reduction in GFR. Higher FGF-23 concentrations were independently associated with higher risk of achieving the end-point. Time to progression was 40% shorter for the tertile with the highest FGF-23 levels compared to the tertile with the lowest FGF-23 levels. After multivariate adjustments (for baseline GFR, blood pressure, underlying glomerular disease, proteinuria and medication use) children with higher FGF-23 levels at baseline were significantly associated with CKD progression.

These findings concur with adult studies (Tsai et al. 2018; Kendrick et al. 2011). However, the correlation between rising phosphate trends and CKD progression is a much-debated topic, with some paediatric studies demonstrating a positive association (Warady et al. 2015; Staples et al. 2010) and other adult studies indicating no association (Mehrotra et al. 2013; Titan et al. 2011; Fliser et al. 2007). The CKiD data (Warady et al. 2015), and the retrospective cohort study of 4166 paediatric patients with CKD 2-4 by Staples et al. (2010) found hyperphosphataemia be a risk factor for disease progression in children. The retrospective nature of this study was a recognised limitation. These early changes in phosphate metabolism affirm the importance of a strategy to target phosphate in the early stages of CKD.

Interestingly, in the large adult cohort study by Mehrotra et al. (2013), the lack of a correlation between serum phosphate and disease progression led them to question the validity of serum phosphate as a biomarker in early stage CKD. Given that <1% of the body's phosphate is found in the serum, they suggest it may be more appropriate to use an alternative measure, such as urinary phosphate excretion (Mehrotra et al. 2013) or FGF-23 levels to predict those at risk of disease progression (Portale et al. 2016). Although the debate about the most appropriate measure of whole body phosphate continues, there have been no conflicting data about the role of FGF-23 as an early biomarker of disease progression (Takashi and Fukumoto, 2018; Kim et al. 2019). This potential for an early dietary clinical marker to guide phosphate

management was explored by the CYP, parent and HCPs perspective as part of the EPSM strategy, during this doctoral study.

3.1.3 Dietary phosphate restriction and FGF-23

In the early stages of CKD, despite an altered phosphate intake, FGF-23 modifies serum phosphate levels via a cascade of endocrine activity in the kidney, gut and parathyroid glands, maintaining serum phosphate at a near neutral level (Sinha et al. 2012) (see 3.1.2). As a result, FGF-23 is a favourable biomarker to monitor response to interventions aimed at lowering dietary phosphate uptake (Isakova et al. 2015).

In this emergent field, to date, definitive evidence demonstrating an efficient and safe strategy to control FGF-23 is yet to be identified. Diet alone is the preferred first line management, followed by the introduction of a phosphate binder medication (NICE, 2013; Evenepoel and Vervloet, 2016). As dietary phosphate intervention is a key responsibility of the paediatric renal dietitian (McAlister and Pugh et al. 2020), albeit only during the later stages of CKD, it is the central SM focus for this study. Thus, the evidence presented in this section will primarily focus on the use of diet alone. Studies using phosphate binder medication to control phosphate were excluded from this doctoral study, as they are predominantly a medical (not dietetic) prescription.

Isakova et al. (2015) and Tsai et al. (2018) conducted a review, and systematic review with a meta-analysis, to examine the rationale and approaches to phosphate and FGF-23 reduction, and the effects of lower versus high phosphate diets on FGF-23 levels, respectively. Both of these studies were adult based. Key data from the Isakova et al. (2015) and Tsai et al. (2018) reviews involving dietary manipulation alone in early stage patients can be found in Table 10, p. 38. Changes from a meat to a grain/plant-based diet consistently reduced FGF-23 levels (Moe et al. 2011; Di Iorio et al. 2012).

Table 10. Key adult data on GFR and FGF-23 levels from Isakova et al. (2015) and Tsai et al. (2018)

Author(s)	Study design	GFR (mean) ml/min/1.73m²	iFGF-23 (pg/ml)	Country
Moe *et al.* (2011) n=9	Crossover trial	32 (CKD 3)	Grain-based vs meat-based diet Pre/post meat: 72 & 101pg/ml Pre/post grain: 84 & 61pg/ml (paired *t* test <0.008)	US
Di Iorio *et al.* (2012) n=32 (n=16 for each 7 day diet)	Prospective randomised controlled crossover study	30 (CKD 3)	Plant/animal-based low protein diet (167.8 ±44.1pg/ml) Plant-based very low protein diet (111.6 ±85.pg/ml) (*p<0.001*)	Italy

In the small crossover study by Moe et al. (2011), the impact of changing the protein source of phosphate was compared using a grain-based diet, then a meat-based diet. Each diet was followed for one week, with no wash out period between the crossovers - when patients receive no intervention to eliminate the effects of the previous diet. Authors recognised this as a potential limitation of the study. The grain-based diet demonstrated a statistically significant reduction in FGF-23 levels compared with the meat-based diet, affirming the FGF-23 lowering potential of a plant-based diet, with its rich content of low bioavailable organic phosphate (see section 1.3).

Similarly, in the randomised crossover study by Di Iorio et al. (2012), phosphate consumption was altered using a very low (all plant-based protein source), and a low protein diet (1:3.6g ratio plant to animal-based protein source). Then, acting as their own control, patients switched to the alternative diet. The reduced consumption of phosphate from the plant-based very low protein diet resulted in a significant fall in FGF-23 levels. Once again this affirms the rationale behind the EPSM strategy with its encouragement of more plant-based unprocessed foods.

An altered dietary phosphate intake (Levey et al. 1996), and the reduced exposure to inorganic phosphate additives (Sullivan et al. 2009) demonstrated a varied impact on serum phosphate levels (a conventional marker of phosphate metabolism) and disease progression. Neither of these studies measured FGF-23 levels. Although the Sullivan et al. (2009) study involved adults on dialysis (CKD 5), lessons can be drawn from their educational approach, to inform the EPSM strategy. Sullivan et al. (2009) delivered an intensive face-to-face dietetic education and food labelling advice

programme to reduce consumption of inorganic phosphate food additives. Practical eating out resources, and a follow-up telephone call reinforced the message, achieving a reduction in serum phosphate levels, and a statistically significant increase in interest and knowledge of food labels/ingredients (Sullivan et al. 2009) (see Table 11).

Table 11. Data from Sullivan et al. (2009) and Levey et al. (1996)

Author(s)	Study design	GFR (mean) ml/min/1.73m^2	Serum phosphate	Country
Sullivan *et al.* (2009) n=279	Cluster randomised controlled trial	CKD 5	Decline in serum phosphate of 0.6mg/dL in intervention vs control group (95% CI-1.0 to-0.1mg/dL)	US
Levey e*t al.* (1996) n=585 (Newsome *et al.* 2013; Selamet *et al.* 2015)	Randomised multicentre clinical trial (3 year follow up)	**Early stage:** CKD 3b GFR 25-55ml/min/1.73m^2	Study A: Mean serum phosphate level change over 0-4 months: **Usual protein diet** 0.01mg/dL (95% CI -0.05 to 0.06) **Low protein diet** -0.05 (95% CI -0.10 to 0.01) Mean serum phosphate level remained at 3.7mg/dl over the 3 years	US
		Established Renal Disease: CKD 4-5 GFR 13-24 mL/min/1.73 m^2	Study B: Mean serum phosphate level change over 0-4 months: **Low protein diet** -0.17 (95% CI -0.29 to −0.04) **Very low protein diet with ketoacids** -0.25 (95%CI -0.38 to −0.12) Mean serum phosphate increased from 3.8 to 4.8 mg/dL (VLP) and from 4.1 to 5.2mg/dL (LP)	

Conversely, in a recent *post hoc* analysis of data from the Modification of Diet in Renal Disease (MDRD) study (Levey et al. 1996), Selamet et al. (2016) and Newsome et al. (2013) examined the clinical significance of dietary phosphate restriction across CKD stages 3b-5, on the risk of progression to ERD. GFR determined participants allocation to parallel randomised control trials, study A (GFR 25-55ml/min/1.73m2 CKD3b) and study B (GFR 13-24ml/min/1.73m^2 CKD 4/5). Early stage CKD (Study A) participants were randomised to either a usual (UP) (1.3g/kg/day) or low (LP) (0.575g/kg/day) protein diet, and established renal disease patients (Study B) to the LP or very low protein (VLP) diet (0.28g/kg/day). The alteration of dietary phosphate intake (assessed

by 24-hour urinary phosphate excretion [UPE]) during the early stages of CKD (Study A) was negatively associated with progression to end-stage renal disease. Additionally, there was found to be no association between dietary phosphate intake and serum phosphate concentration. Irrespective of diet in the early stage, mean serum phosphate remained stable across the 3 year study period. By contrast patients at CKD stage 4-5 experienced a rise in serum phosphate, which was more marked for those on the VLP diet.

However, *post hoc* analysis of data from the MDRD study has raised legitimate concerns regarding the internal and external validity of the observational data (Evenepoel and Vervloet, 2016). Critically, the source of the protein was not detailed, limiting clarity regarding the phosphate origin and bioavailability. Long-term dietary compliance, and therefore protein intake was problematic, greatly reducing confidence in the outcomes (Menon et al. 2009). An individual's 'usual' phosphate intake was measured using a single urinary phosphate excretion measurement. Although 24-hour UPE lacks both the recall bias inherent in 24-hour dietary recall methods and the inaccuracy of nutrient databases with regard to phosphate content of popular food items (Kuro-o, 2013), the accuracy of a single measure is debated (Cupisti and Gallieni, 2019) and takes no account of the variation in dietary phosphate exposure, compromising internal validity. Additionally, the highest 'usual' 24-hour UPE reported in the MDRD study was comparable to the mean baseline 24-hour UPE in a trial of healthy participants (Vervloet et al. 2011). Moreover, the 'usual' intake suggests an ordered eating pattern, which is contrary to the growing trend for 'grazing' on nutrient-poor processed snacks rather than three proper meals a day (Bellisle, 2014). Since the MDRD data was collected in the late 1990s phosphate additive use by the food industry has increased significantly. The demand from society for 'healthy convenience' foods has resulted in an estimated increase in phosphate consumption from additives alone from 500 mg/d to over 1000mg/d (Kalantar-Zadeh et al. 2010). These shifts in dietary phosphate consumption raise genuine concerns regarding the external validity of the MDRD findings. The contrary findings of the MDRD study can be explained by its design faults.

Based on these studies involving participants with CKD stage 3-5, there is evidence to support the use of dietary phosphate restriction alone as having a beneficial effect on CKD progression. The absence of randomised hard-end-point studies providing definitive evidence on the efficacy of a phosphate-restricted diet in pre-dialysis CKD patients (1-3), where serum phosphate is normal or near-normal, does not negate its potential 'preventative' role (Bover and Cozzolino, 2019). Rather, it identifies a clear gap in the literature, a 'new paradigm of phosphate restriction' (Kuro-o, 2013) and a need to explore stakeholders' perspectives towards such an intervention.

To date, no study has examined the impact of modified dietary phosphate sources alone in children with early stage CKD. This may relate to concerns regarding the compromising of protein intake for growth (Chaturvedi and Jones, 2007). However, a focus on reducing inorganic phosphate additives and not on protein would reduce these concerns (Evenepoel and Vervloet, 2016).

Although the inclusion of phosphate education is largely absent from early dietary strategies and guidelines, the studies detailed in the next section suggest it has a potential 'preventative' role.

3.1.4 Clinical practice guidelines for CKD stages 1-3, and stakeholder involvement in their development

A recent systematic review by Lopez-Vargas et al. (2013) identified 15 guidelines and one consensus statement between 1980 and 2011, on the management of adults and children with CKD stages 1-3. Lifestyle modifications were recognised in all of the guidelines, however, the extent of coverage varied significantly. Despite all guidelines advocating lifestyle change, dietary modifications to delay CKD progression were included in just 10 (67%) guidelines, with only one guideline (Flores et al. 2009) placing it as first line therapy, with a key focus on weight management. Dietary changes described included increasing fruit and vegetable intake, inclusion of specific lipid-lowering advice, reducing salt, and fluid, protein and energy restrictions. Phosphate did not feature in the advice, and only three guidelines advocated referral to a dietitian. This verified a gap in the literature, and a need to examine perspectives on early dietary phosphate management, with early specialist renal dietetic involvement, as addressed in this doctoral study.

The recent KDIGO (2017) clinical practice guideline update for the diagnosis, evaluation, and treatment of CKD-MBD in adults (Burton et al. 2018) implicated a role for diet in the treatment of adults with early CKD 3a, using 'phosphate-lowering therapies'.

Due to its more recent publication, the adult 'KHA-CARI Guideline: Early chronic kidney disease: Detection, prevention and management' by Johnson et al. (2013) did not feature in the Lopez-Vargas et al. (2013) above review. However, early modification of lifestyle habits and dietary change featured as an important component of early disease management to delay disease progression (Johnson et al. 2013), affirming the key focus of this doctoral study. Based on the lack of current clinical evidence of interventions relevant to lifestyle, Johnson et al. (2013) discouraged the restriction of dietary phosphate intake in the early stages of CKD (stages 1–3). The recent clinical practice recommendations from the Pediatric Renal Nutrition Taskforce (McAlister and Pugh et al. 2020) for the dietary management of calcium and phosphate in CYP with CKD stages 2-5, elevates the importance of targeting dietary phosphate in the early stages of CKD. It provides a practical tool to support the regular assessment and management of dietary phosphate intake in the early stages of CKD.

Stakeholder involvement in the development of practice guidelines

To improve methodological quality, ensuring greater relevance, endorsement and uptake, which should serve to improve patient outcomes, there is a need to move away from HCP solely writing practice guidelines, towards the involvement of patients (Lopez-Vargas et al. 2013). Using the AGREE II instrument to assess methodological quality, Lopez-Vargas et al. (2013) reported stakeholder involvement scored low. Furthermore, applicability, consideration of the challenges and facilitators, and the advice and tools to implement a change in practice also scored low (Brouwers et al. 2010). Authors did not mention the involvement of stakeholders in the development of the KHA-CARI guidelines (Johnson et al. 2013). Likewise, although the Pediatric Renal Nutrition Taskforce comprised a team of international paediatric nephrology experts, representing the voice of the CYP/carer, no service users were involved in the

development of the practice recommendations. This highlights the need to capture patient/carer perspectives.

3.1.5 Patient perspective on CKD self-management

Despite recognition of the importance of including the CYP perspective on SM in CKD, there is a paucity of data in this area (for example, Lee and Shin, 2018; Nightingale et al. 2017). The Korean paper by Lee and Shin (2018) was available with only small sections translated into English. Lee and Shin (2018) undertook in-depth interviews with thirteen adolescents with CKD, identifying the need to take control by integrating disease experience into their life through SM. CYP spoke of their efforts to regain normalisation by reorganising their daily life. Access to motivational resources and support from parents and friends provided mechanisms to support SM (Lee and Shin, 2018).

Eliciting the CYP's perspective was central to the development of a digital care-management app for CYP with CKD. During seventeen interviews with CYP, Nightingale et al. (2017) recognised CYP's desire for a variety of interactive media and content, specifically adapted to their age and condition. This would help reduce feelings of isolation, support care-management, and enable them to live "normal" lives. In this doctoral study, stakeholders were encouraged to consider the EPSM strategy's educational platforms.

Drawing on what adults with CKD want from a SM strategy, Havas et al. (2016a) conducted an integrative review of SM support from an adult patient's perspective. Twelve studies met inclusion criteria, with 12 topics defined as being significant to people living with CKD. The desire for diet-specific knowledge was identified in seven studies (see Table 12, p. 44). Additionally, the recent scoping review identifying and describing SM interventions for adults with CKD (Donald et al. 2018), affirmed the most common intervention topic was diet/nutrition. Patients in these reviews requested support and help to understand complex or confusing dietary issues (Lewis et al. 2010; Ormandy and Hulme, 2013; Griva et al. 2013), and practical resources to support implementation and establish a daily routine (Costanini et al. 2008). These studies highlight the desire of patients for more dietary information to inform the

daily management of their chronic disease, which underpins a key focus of this doctoral study.

Table 12. Self-management adult studies recognising the need for diet-specific knowledge

Lead authors/year/location	Diet-specific knowledge
Costantini *et al.* (2008) Canada	Lifestyle, diet and fluids
Curtin *et al.* (2004) America	Controlling diet
Griva *et al.* (2013) Singapore	Lifestyle, diet and fluids
Lewis *et al.* (2010) America	Lifestyle and diet
Lopez-Vargas *et al.* (2014) Australia	Lifestyle, diet and fluids
Ormandy (2008) England	Lifestyle, diet and fluids
Ormandy & Hulme (2013) England	Lifestyle, diet and fluids

Self-management was recognised as taking place in the context of everyday life. In one study (Harwood et al. 2005) individuals (mean age 72.7 years) described their anxieties around being a burden, due to their dependence on others. Peer support (Griva et al. 2013; Ormandy and Hulme, 2013), and signposting to available resources and support groups (Lopez-Vargas et al. 2014) were recognised. Children and young people with chronic disease are known to value the social support of those in their chronic disease peer group, as well as support from their wider peer group (Lee and Shin, 2018; Sawyer and Aroni, 2005). Additionally, maintaining normality in life was recognised as important (Ormandy and Hulme, 2013; Lopez-Vargas et al. 2014).

A lack of confidence and self-assurance affected their ability to self-manage (Lopez-Vargas et al. 2014). Individuals wanted the skills to respond to their changing health needs, and build confidence to assume more control. Many recognised the importance of looking after their mental health (Costantini et al. 2008) – a pertinent issue in CYP, when maturity and confidence are developing, and mental health issues are rising (RCPCH, 2014).

Adults with all stages of CKD recognised the building of effective and supportive relationships with HCPs as a key facilitator of SM (Curtin et al. 2004; Lopez-Vargas et al. 2014), including encouragement to voice their views and opinions on practice and service development (Curtin et al. 2004; Lopez-Vargas et al. 2014). Finally, four studies

(Clarkson and Robinson, 2010; Costantini et al. 2008; Harwood et al. 2005; Williams and Manias, 2013) highlighted a desire to start SM early in the disease, expressing disappointment at not being given information after diagnosis (Costanini et al. 2008). Each of these important facets of SM informed and shaped the interview guide for Phase 2, which explored stakeholders' perspectives towards the development of an EPSM strategy in this doctoral study.

Patient engagement in the design of self-management interventions for CKD

Similar to the development of practice guidelines, many authors in the integrative review by Havas et al. (2016) recognised the lack of the patient's perspective in the design of SM CKD interventions, with the majority designed by HCPs. Likewise, Donald et al. (2018) recognised how much person-centred care is shaping the way HCPs deliver care to patients and the growing need to elicit the perspective of patients and families, involving them in the co-production of SM strategies to give credence to their values, culture and psychosocial needs. This affirmed the decision to elicit the CYP and parent perspective on the development of an EPSM strategy, alongside the HCP, in this doctoral study.

3.1.6 Healthcare professionals' understanding of self-management

HCPs managing chronic disease are increasingly aware of the need for a concerted focus of attention on early intervention, delaying disease progression and improving outcomes (Parker et al. 2004; Havas et al. 2016a). HCPs are best situated to provide early SM support for CYP with CKD and their family. However, research suggests HCPs, CYP and their parents have differing views concerning what a SM strategy looks like (Sadler et al. 2014).

A systematic review by Sadler et al. (2014) with a narrative synthesis of lay and HCPs understanding of SM comprising 55 studies, reported a broad coverage of perspectives from allied health professionals, nurses, carers, psychologists and other HCPs. The perspective of individuals with a range of chronic diseases, including, asthma, arthritis, diabetes and heart disease were included. Three core conceptual themes were generated, as follows:

Models of professional-patient relationship

HCPs and adult patients routinely interpret SM as a vehicle to promote compliance with prescribed specialist advice or regimens, and many adult patients preferred being told what to do (Yen et al. 2011) – the medical model. Where compliance was poor, HCPs ascribed this to adults' negative psychosocial attributes, such as poor motivation and a lack of willingness to engage or take responsibility, a recognised area of contention in adolescents (Ferris et al. 2015). Parents are held to account for the adherence of their CYP; simultaneously adolescents are encouraged to take on board more responsibility for their own disease management (Ferris et al. 2015; Sawyer and Aroni, 2005). Compliance was felt to be impeded by a lack of skills, knowledge, or social constraints and emotional issues. Improved education was advocated to increase competency and compliance with advice (Yen et al. 2011). Crucially, a disparity of information or lack of joined up services affected compliance (Kirby et al. 2012).

By contrast, some adults perceived SM as a shared venture with HCPs, an opportunity to gain increased control through active engagement in decision-making. Where adolescents are encouraged to share their disease management with the HCP (Ferris et al. 2015; Lorig et al. 2003; Nightingale et al. 2019) they report feeling motivated (Kyngäs et al. 1998). The shared SM approach is explored in this doctoral study.

Quality of relationship between HCP and adult patients

For many adult patients SM involved building quality collaborative relationships with HCPs, and receiving personalised support tailored to their changing needs. When HCPs actively nurtured mutual respect and trust, communicated well and offered psychosocial support and choice, individuals felt they were being treated more holistically (Sadler et al. 2014).

Although collaboration was recognised by HCPs, it was regularly referred to in different terms (Sadler et al. 2014). HCPs recognised the importance of tailored advice but acknowledged the challenges of delivering continuity and consistency when community services were fragmented, and skilled HCPs were limited (Sadler et al. 2014). The researcher recognises that a collaborative ethos is constantly challenged

across the EMEESY network, due to a lack of joined up care and a diminishing workforce.

Contextualising self-management

Adults with CKD desired biomedical and psychological strategies to support the SM of their disease, and signposting to identified structures that helped them make sense of and deal with the challenges of living with a chronic disease. Social support from family and friends, and peer influences are essential (Sadler et al. 2014). These factors are either not acknowledged or taken-for-granted by HCPs.

Sadler et al. (2014) concluded that the dichotomy of adult patients' and HCPs' perspectives regarding the purpose and implementation of SM might explain why many SM strategies lack effectiveness. The focus needs to move away from compliance, towards empowering individuals to manage the everyday challenges of living with a chronic disease. HCPs need to understand the diversity of patients' expectations and account for a patient's social setting, nurturing a collaborative partnership when planning SM strategies. No mention was made of the HCPs' views towards SM in the early stages of a chronic disease, and the CYP's perspective was not presented.

The nearest studies to this doctoral research

The literature was searched to identify any similar studies to this doctoral research project involving stakeholder engagement (with the inclusion of CYP) in the exploration of early CKD dietary SM strategies. No studies were found. However, a US study on stakeholder engagement in a diabetes SM study (Kwan et al. 2017) provided valuable insights.

Kwan et al. (2017) engaged primary care practitioners, community resource representatives, adult patients with diabetes and family members in a community-based participatory research approach known as Boot Camp Translation (BCT). The purpose of the project was to translate evidence about diabetes and SM support into an intervention strategy that addressed multiple stakeholder needs. Boot Camp Translation involved an initial lengthy meeting (6 hours) with multiple subsequent conference calls (x 4) and individual meetings (2-2.5 hours each).

The following lessons from this study were carried forward to the doctoral study. Stakeholder engagement facilitated co-learning, to develop, refine and implement the new diabetes SM programs. The onerous time commitment of BCT was not realistically feasible in this doctoral study due to the limitations of EMEESY stakeholder participants' busy schedules and geographical spread. However, the project proved helpful in 'meeting people where they are at', yielding valuable insights, which were often different to what the research team expected. The study highlighted the value of multiple stakeholder engagement using a pragmatically relevant approach, which provided flexible guidance to maintain the research focus, while allowing participants to harness their own expertise.

3.2 Gaps in the literature

The aforementioned evidence has identified a clear need to target dietary phosphate management in the early stages of CKD as a potential 'preventative' tool to delay disease progression (Bover and Cozzolino, 2019; Moe et al. 2011; Di Iorio et al. 2012). In this emergent field, to date, definitive evidence demonstrating an efficient strategy to control FGF-23 in CYP is yet to be identified. FGF-23 proposes an early clinical marker of phosphate overload, and a self-governing risk factor for disease progression. The inclusion of dietary phosphate education by the Pediatric Renal Nutrition Taskforce, in the early management of CYP with CKD (McAlister and Pugh et al. 2020) highlights the growing importance of this issue and the need to be proactive.

Furthermore, the need for an early dietary phosphate intervention is affirmed in the SM literature with an implicit desire for dietary education and support to begin early to delay disease progression and improve outcomes. Stakeholder engagement in the development of SM strategies will ensure greater relevance, approval and uptake of the strategy in clinical practice. CYP, parents and HCPs understand SM differently. This incongruity needs to be addressed particularly when planning an EPSM strategy for CYP, where both the CYP, parents and HCPs need to have a clearer understanding of each other's perspective if a positive partnership is to be established to improve health outcomes. Therefore, future developments in SM strategies require preliminary engagement with the CYP, adult and HCP perspective.

This scoping search has identified a clear gap in the evidence for an early dietary phosphate self-management strategy for CYP with early stage CKD (1-3), to delay disease progression. The scoping search demonstrated that to date, no published research has been undertaken to elicit the perspective of CYP, parents and HCP towards the development of such an EPSM strategy. This doctoral study will make a valid contribution to knowledge, providing a multi-stakeholder perspective towards development and operationalisation ('how', 'who', 'when', 'where' and 'challenges and enablers') of an EPSM. This scoping search directly informed the mixed-studies systematic review with a narrative synthesis.

3.3 The mixed-studies systematic review with a narrative synthesis

The initial scoping search of stakeholders' perspective on the use of an early dietary phosphate SM strategy in children with CKD, yielded a paucity of data (see section 3.1.1 for the initial search of the databases). However, due to the significance of diet in the early management and prevention of progressive chronic disease, the findings suggested that sufficient literature was available to inform a systematic review if the search of 'early dietary intervention as a form of SM' (excluding the term 'phosphate') was extended to all chronic diseases during childhood.

3.4 Rationale for the review

Health science researchers engage in a diverse range of knowledge-making practices, including randomised control trials (RCTs), questionnaires, and qualitative interviews (Bryman, 2016), among others. The comparative virtues of these different approaches are debated, some strive for optimal reliability and validity, while others desire reflexivity and trustworthiness (Bryman, 2016). At the core of these debates is the non-pragmatic quest: to uncover knowledge that takes us nearest to the 'truth'.

As described by Hemingway and Brereton (2009), a systematic review uses a level of rigour to bring together all published and unpublished evidence in a single area, assessing its quality, and providing an unbiased synthesis of the findings in a balanced and impartial summary.

The 'hierarchy of evidence' (Evans, 2003) states that systematic reviews of RCTs are one of the most powerful tools currently available (Kuehn, 2011). Indeed, some would

say RCTs are the 'gold standard' research method, providing the optimum level of evidence (Barry, 2006, p. 2648); with quantitative experimental methods having the highest perceived value (Benton and Craib, 2011) and qualitative methods ranked much lower. However, the advent of MMR in the mid-to-late 1980s (Creswell and Clark, 2011) and the progressive emergence of qualitative research, saw qualitative and mixed-studies systematic reviews gain more prominence (Dixon-Woods and Fitzpatrick, 2001; Pluye and Hong, 2014, Hong et al. 2018). Critiques in the health literature argue that RCTs are insensitive where psychosocial health issues are implied, favouring the subjective, 'real-world' insights that the qualitative literature can bring (Clark, 2007; Deaton and Cartwright, 2018). The pragmatist argues that no outright 'gold standard' method exists; rather each method best serves a given function - its usefulness being determined by its relevance to practice (James, 1975). Therefore, the type of knowledge sought needs to best serve the interest of the project.

3.5 What is a mixed-studies systematic review?

A mixed-studies systematic review involves a concomitant examination of qualitative, quantitative and mixed-methods primary studies to address a complex question (Pace et al. 2012). It provided a rich, comprehensive, and highly practical understanding of complex health interventions, better than a single approach to a systematic review (Higginbottom et al. 2017), giving clinicians and decision-makers a more relevant and useful result (Pace et al. 2012).

3.6 What is a mixed-studies systematic review with a narrative synthesis, and why was it chosen for this study?

As described by Popay et al. (2006), a mixed-studies systematic review with a narrative synthesis is an,

> '*approach to the systematic review and synthesis of findings from multiple studies, primarily based on the use of words and text to summarise and explain the results of the synthesis'*
>
> *(Popay et al. 2006, p. 5).*

Text and illustrations are used to describe, compare, and combine heterogeneous qualitative findings and quantitative results (Pluye and Hong, 2014; Evans, 2002). Qualitative, quantitative and mixed-methods studies can utilise this approach, as the focus is on the interpretive synthesis of the narrative findings of the research as opposed to a meta-analysis of the data (Pluye and Hong, 2014; Higginbottom et al. 2012).

Due to the complex and highly context-sensitive nature of dietary interventions for chronic disease management, a mixed studies systematic review with a narrative synthesis was deemed particularly relevant for this topic (Pluye and Hong, 2014; Higginbottom et al. 2017). As the perspective of the CYP with CKD, their families, and the HCPs involved in the delivery of care are central to this study, this approach accessed the multidisciplinary perspective of stakeholders. Providing a dynamic viewpoint to assess the plurality of health-related knowledge about early dietary SM in chronic diseases using various methodologies, it offered an objective appraisal of the evidence, and the findings provided a platform to inform Phase 2 of this doctoral study.

3.7 Knowledge gap on early dietary interventions for children with a chronic disease or CKD stage 1-3

There is a systematic review of carers' and adult patients' views on the needs and treatment decision-making of adults with CKD (Morton et al. 2010; Tong et al. 2008), the valued experience of parents of children with CKD (Tong et al. 2008), and an integrative review of SM interventions in adults with stages 1-4 CKD (Welch et al. 2015). However, a scoping search of the databases (Medline, CINAHL, COCHRANE, EMBASE, AMED, Scopus and PsychoINFO, and the JBI Database of Systematic Reviews and Implementation Reports) to date has not revealed a systematic review or mixed-studies systematic review on the perspectives of CYP, parents and HCPs towards the use of an early dietary phosphate intervention as a form of SM, to delay disease progression, for children aged 0-16 years with early CKD (see section 3.1.1). As a result, the question for the systematic review was changed to capture 'early dietary intervention in chronic disease'.

3.8 Review question

The review question was framed using the patient-specific evidence-based PICO model (Richardson, 1995), which recognises the patient/population, intervention or phenomena or interest, comparison or context, and outcome as key components (see Box 1).

Box 1. PICO tool

Population – CYP aged 16 years or less, parents or carers and HCPs involved in the provision of care and support of a child aged 0-16 years
Intervention – dietary intervention as a form of self-management during the early stages of a chronic disease or CKD (stage 1-3)
Comparison – qualitative and mixed-methods studies may not have a comparison group. Quantitative studies may compare the given dietary intervention with usual care or standard practice
Context – in-patient and out-patient hospital and community care in any country
Outcome – to delay disease progression

Review question: What are the views of children, parents and other stakeholders towards the use of an early dietary intervention as a form of SM, to delay disease progression, for children aged 0-16 years with an early chronic disease?

3.9 Methods

The protocol for this systematic review was published with BioMed Central (Pugh et al. 2018).

3.9.1 Inclusion and exclusion criteria

The inclusion and exclusion criteria are detailed in Box 2, p. 53.

Box 2. Inclusion and exclusion criteria

Inclusion criteria – the review considered studies that included one or more of the following :

- CYP aged 16 years of age or younger
- Parents or caregivers, HCPs but not exclusively the paediatrician, paediatric nephrologist, nurse or dietitian involved in providing care and support for CYP with early chronic disease or the early stages of chronic kidney disease (stage 1-3) (see section 2.1.2) in a hospital, home or community setting
- Reported CYP, parents' or other stakeholders' perspective towards dietary self-management
- Publication date January 2000 to January 2018
- Reported the inclusion of a diet/nutrition/food or lifestyle changes as part of a self-management strategy
- Empirical qualitative, quantitative and mixed-methods research and findings
- Available in English

Exclusion criteria

- Studies with a primary diagnosis of acute kidney injury (AKI) or damage, or where the chronic disease was caused by solid-organ tumours were excluded from the review
- The review did not consider the perspective and views of CYP, parents and other stakeholders where phosphate binder drugs were used as the primary form of self-management

The following section provides the rationale behind these criteria.

Inclusion criteria

Population - As discussed in section 1.3.2, there is recognition of the need to provide early dietary phosphate intervention for CYP during the early stages of CKD (National Kidney Federation, 2015; Evenepoel and Vervloet, 2016). Furthermore, as discussed in section 1.5.2, SM problems typically occur during the early stages of an illness or during early adolescence (Urbach et al. 2005), making this a good time to engage both CYP and families in education and skills training strategies. For these reasons, the age group 0-16 years and the early stage of chronic disease were included in this systematic review.

Intervention - In order to identify the challenges and enablers to providing an early dietary intervention, the review examined how the dietary SM intervention was carried out, and any challenges encountered.

Design - The review examined studies using research methods of qualitative, quantitative or MMR where the narrative description was reported separately. All descriptive and analytic research study designs were included (CEBM, 2017).

Comparison - Quantitative RCTs (Shroff et al. 2012; Stapleton et al. 2001; Niggemann et al. 2001) studies compared the given dietary intervention with usual standard care or practice.

Context - The review included in-patient and out-patient hospital and community care in any country, which recorded the perspectives of CYP, parents or other stakeholders on early dietary SM for CYP with early chronic disease or CKD (stage 1-3).

Outcome - As discussed in section 1.2.2 and 1.2.3, chronic disease morbidity begins in the early stages of a disease, and the provision of a dietary intervention that will delay the progression of the disease is necessary.

The review included publications from the year 2000 up to the date the protocol was published in January 2018; marking the period when MMR and mixed studies reviews began to emerge (Pluye and Hong, 2014).

Exclusion criteria

As this systematic review included early intervention strategies for CKD, studies with a primary diagnosis of acute kidney injury (AKI) or damage, where there is a potentially reversible short-term decline in renal function (Thomas et al. 2015), were rejected. Studies including solid-organ tumours, defined as an abnormal mass of tissue that usually does not contain cysts or liquid and can be benign, and are not always classified as a progressive chronic disease (Lazebnik, 2010), were also excluded from this review. Phosphate binders are medicines taken with meals and snacks to reduce the phosphate absorption in the digestive system (Gandy, 2019). Since this study focused on dietary intervention and not drug therapy, studies that included phosphate binder drugs as a primary form of SM were excluded. Because of the linguistic abilities of the reviewers, only English-language studies were included. Studies referring to lifestyle change, without specifying a dietary component were excluded due to ambiguity. Reviews, editorials, commentaries and expert opinion papers were excluded, although the reference lists of reviews were searched for relevant literature.

3.9.2 Study identification and search strategy

The following section details the process for identifying studies for inclusion in the systematic review. Scoring system methodologies for systematic reviews as detailed in the Centre for Reviews and Dissemination Guide for Undertaking Reviews in Health Care (CRD, 2009) were utilised. In order to obtain the maximum literature coverage, to identify published and unpublished studies, the search strategy was applied to several databases, using a four-stage approach.

3.9.2.1 Stage 1 – initial search of the databases

The initial scoping search of Medline, CINAHL, COCHRANE, EMBASE, Scopus, PsycINFO and OpenGrey did not identify sufficient early dietary phosphate SM literature in CYP with CKD (see section 3.1.1). To allow lessons to be drawn from the early dietary management and prevention from other disciplines outside of CKD, the search was changed to early dietary SM in CYP with early CKD or early chronic disease.

3.9.2.2 Stage 2 – application of validated filters

The validated Patient Issues search filters on the Scottish Intercollegiate Guidelines Network (SIGN) (www.sign.ac.uk) were applied to Medline. However, despite providing a broad coverage of patient issues, it did not deliver the required specificity to address the review question. With the support of an information scientist, a bespoke chronic disease and CKD search strategy reflecting the review question (section 3.8) was devised. A detailed search strategy for Medline (January 2000- 2018) is shown in Appendix I. The key search terms were the result of a detailed tabulation and analysis of the search terms and Medical Search Headings (MeSH) identified and applied during Stage 1 and 2 of the search strategy. The key search terms and Boolean operators applied are detailed in Table 13, p. 56.

Table 13. Search terms and Boolean operators

Concepts	Key words	Boolean operators
Condition	Chronic Disease	OR
	Chronic Illness	OR
	Chronic Disorder	OR
	Chronic Kidney Disease	OR
	Chronic Kidney Failure	OR
	Renal Insufficiency	OR
	CKD	
		AND
Population	Patient	OR
	Adolescent	OR
	Child	
		AND
Care provider	Parent	OR
	Carer	OR
	Nurse	OR
	Consultant	OR
	Physician	OR
	Dietitian	OR
	Allied Health Personnel	
		AND
Early Intervention	Self-management	OR
	Self-care	OR
	Early intervention	OR
	Secondary prevention	
		AND
Progression of disease	Disease progression	OR
	Time	OR
	Endpoint	
		AND
Diet	Diet	OR
	Nutrition	OR
	Diet treatment	OR
	Food	
		AND
Views	Perspectives	OR

3.9.2.3 Stage 3 – chronic disease and CKD search strategy

Stage 3 – chronic disease and CKD search strategy

The bespoke search strategy was applied to seven databases (see Table 14, p. 57).

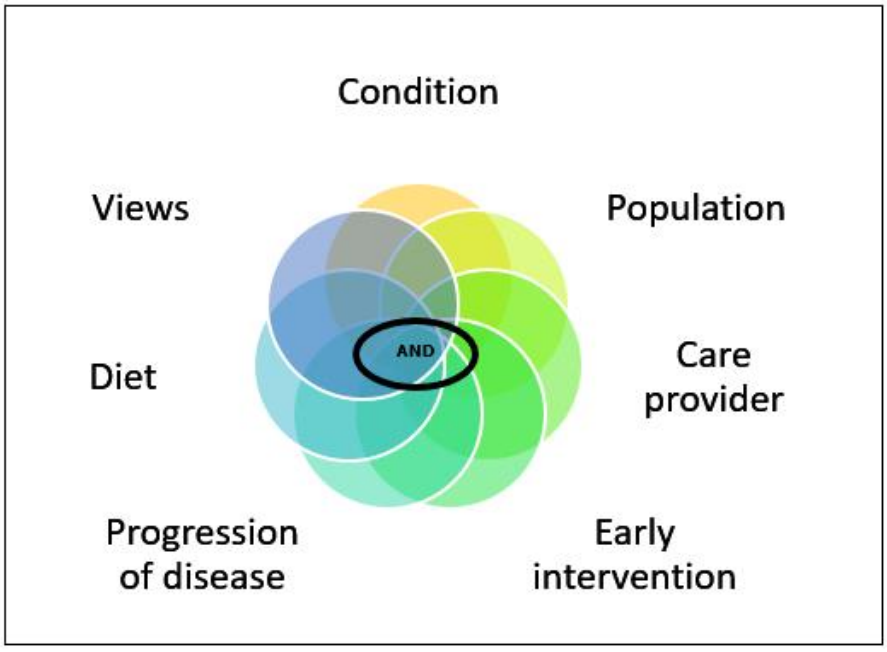
Dates - The date in brackets reflects the individual databases start date.

Table 14. Databases searched during stage 3 of the search strategy

Database	Provider
MEDLINE – In-Process & Non-Indexed Citations and Ovid Medline 1946	Ovid (1946 onwards)
EMBASE – Excerptra Medica Database	Elsevier (1986 onwards)
COCHRANE Library	(1993 onwards)
CINAHL – Cumulative Index of nursing and Allied Health literature	EBSCO (1982 onwards)
PsycINFO	(1995 onwards)
Scopus	Elsevier (1970 onwards)
OpenGrey (System for Information on Grey Literature in Europe) www.opengrey.eu	INIST-CNRS (1997 onwards)

Figure 4 depicts the search strategy. The black circle where all the individual circles/concepts overlap, shows the studies that have been retrieved and screened for inclusion in the review.

Figure 4. Chronic disease and CKD search strategy



Wildcards

Wildcards customised to each database maximised search outcomes. For an example of the wildcards used in Medline, see Table 15.

Table 15. Wildcards applied to the Medline search

Wildcard	Meaning
Asterisk *	When used at the end of a root word as a "truncation" it tells the database to look for all possible endings to the root word.
Proximity operator adj2	Chronicadj2 (illness* or disease* or disorder*) The proximity operator finds one keyword within a specified number of words. The word chronic was found within 2 words of the words in the bracket. The proximity operator can also be used with the truncation symbol to find chronic illnesses, chronic kidney disease or chronic renal disorders, for example.

Screening and final selection

In accordance with the inclusion/exclusion criteria, all studies retrieved during the Stage 3 search strategy underwent a two-phase study selection process: (a) screening; and (b) retrieval and final selection (Higginbottom et al. 2012). The studies were imported into EndNote X8, a reference management software package provided free of charge by the University of Nottingham (UoN). Duplicates were removed and the title, abstract and full-text of the remaining studies were examined against the screening and selection criteria checklist (see Table 16).

Table 16. Screening and selection criteria checklist

Item	Yes	No	Can't say
1. Involved children (≤16 years) with an early chronic disease or CKD (1-3)			
2. Reported children, parents' or other stakeholders' perspective towards dietary self-management			
3. Publication data January 2000-April 2018			
4. Reported the inclusion of diet/nutrition/food or lifestyle changes as part of a self-management strategy			
5. Available in English			
6. Empirical research and findings			

Adapted from Higginbottom et al. (2012)

A secondary reviewer decided on the final inclusion of articles for full retrieval. Where it was not possible to confirm that the literature met all the criteria, by marking with 'can't say', this literature passed to the next stage for full retrieval and further screening.

3.9.3.4 Stage 4 – hand search

Finally, the reference lists of papers identified in stage 3 were searched by hand for additional studies. Author searches and contacts were carried out as necessary.

3.9.3.5 Grey literature

Due to the utilisation of digital technologies in education and training, and the need to provide a more complete and relevant view of the available evidence (Mahood et al. 2014), there is an increasing recognition of the importance of including grey literature in systematic reviews (Tyndall, 2010; White et al. 2012). Health service researchers regularly produce and use grey literature to defend their inquiry (Leroy et al. 2016), and Chronic Care and Chronic Care Delivery are priority subject areas (AcademyHealth, 2006) for grey literature. The OpenGrey database, a multidisciplinary system for information on Grey Literature in Europe (www.opengrey.eu) was used. Alternative grey literature databases were considered such as WorldCat (www.worldcat.org) and BASE (www.base-search.net); however, the multidisciplinary feature of OpenGrey provided access to a wide range of health related literature. Grey literature includes empirical research (using qualitative, quantitative and MMR), doctoral thesis, conference papers and official publications in print and electronic formats, which are not controlled by commercial publishers (Tyndall, 2010). In accordance with the checklist of screening and selection criteria as shown earlier in Table 16, p. 58, grey literature items were selected as appropriate.

3.10 Quality assessment

To reduce bias, all relevant studies retrieved through the search strategy were critically appraised for methodological quality, by the researcher and a supervisor, before inclusion in the review.

3.10.1 Quality assessment of published literature

The Mixed Methods Appraisal Tools (MMAT) (Souto et al. 2015; Hong et al. 2018; Popay et al. 2006) (Appendix 2) and scoring system, and the Centre for Evidence-Based Management (CEBM, 2017) were used to review the quality of published literature. The MMAT is recognised as an efficient and validated tool (Souto et al. 2015), and the only available tool for the concurrent quality assessment of qualitative, quantitative and mixed methodological studies (Hong et al. 2018). Its use in systematic reviews is well recognised (Brooks et al. 2018; Kerins et al. 2018). Disagreements between reviewers were resolved by discussion and, if necessary reached consensus with a third reviewer.

3.10.2 Quality assessment of grey literature

In order to critically appraise the trustworthiness and relative importance of the grey literature, a quality evaluation tool was sought. Authors including grey literature in systematic reviews have either developed a tool to meet their review requirements (Van Eerd et al. 2010) or used the Tyndall's checklist (Dawson et al. 2014) (see Appendix 3). In this review, Tyndall's checklist (2010), a recognised tool for assessing the methodological quality and relative importance of grey literature was used alongside McGrath et al. (2006) framework (see Table 17) and the AcademyHealth advisory committee categories (see Table 18, p. 61). Tyndall's AACODS checklist takes into account the following aspect of grey literature: Authority, Accuracy, Coverage, Objectivity, Date and Significance (see Appendix 3). The McGrath et al. (2008) framework judged the quality of information/data description. The two reviewers accepted any reports matching category A for further screening (Table 17).

Table 17. McGrath et al. (2006) framework for methodological quality

Category A (Accepted)	Reports judged to have a sufficient quality to be included in the next stage of selection process
Category B (Background)	Reports judges to provide background or contextual information
Category R (Rejected)	Reports judged to be of no use for purpose of the review

Table 18. AcademyHealth (2006) category of relative importance of grey literature

5	4	3	2	1
Working papers Testimony Committee reports Conference proceedings	Data Evaluations Foundation reports Government reports Grantee publications Non-commercially published conference papers Reports Special reports	Speeches Annual reports Presentations Grantee reports Webcasts Theses Technical specifications & standards	Newsletters Bibliographies Bulletins PowerPoint presentations Foundation financial statements	Pamphlets Protocols Guidelines Poster sessions Meeting agenda Translations

The AcademyHealth (2006) advisory committee identified categories 4 and 5 as being of sufficient relative importance (see Table 18). The grey literature that passed the evaluation had to present a clear research question(s), with key findings, and provide adequate details on the population studies, interventions, and study design, analysis method and evaluation outcomes (CEBM, 2017).

3.10.3 Data extraction

The type of data that was extracted was determined by the nature of the studies, with an emphasis on how best to answer the research question. The Centre for Review and Dissemination Guidance (CRD, 2009) and Popay *et al's* work (2006) provided a suggested focus for the data extraction variables. Appendix 4 lists data extraction variables. Data extraction was primarily conducted by the researcher using a Microsoft® Word 2016 tabulation, with checks conducted by the supervisory team.

3.10.3.1 Analysis using narrative synthesis

Elements of Popay et al. (2006, p. 5) four step narrative synthesis methodology were used (see Table 19). The process involved a cyclical interweaving of the steps, drawing out the narrative from the text in order to "tell a trustworthy story".

Table 19. The four steps for narrative synthesis

A narrative synthesis comprises four key steps:

1. Developing a theory of how the intervention works, why and for whom;
2. Developing a preliminary synthesis of findings of included studies;
3. Exploring relationships in the data;
4. Assessing the robustness of the synthesis

As suited to the nature of the evidence, this process used a variety of narrative synthesis and risk of bias tools within each step (see Table 20) (Popay et al. 2006). The four steps (Popay et al. 2006), with rationale for choosing specific tools, are described next.

Table 20. Tools for narrative synthesis and risk of bias

Element	Tools for narrative synthesis and assessment of bias
2	Evans et al. (2002) The synthesis process/Tabulation A textual summary of the results in a tabular format with headings such as setting, participants, research aim, sampling technique, analysis, results
2 and 3	Clarke & Braun; Braun & Clark (2013; 2006) Thematic analysis The data presented in tabular format allowed papers to be grouped according to themes that appeared between and within studies. Themes related to particular features such as dietary intervention, stakeholder groups or main findings. Recurrent themes were analysed using thematic analysis.
3	Popay et al. (2006) Narrative synthesis/Investigate heterogeneity/Conceptual mapping. Identify and investigate key characteristics that vary between the studies in order to elucidate the different findings. Clinkenbeard (1991) ideas or concept maps Explore connections reported across the studies
4	Popay et al. (2006) Robustness of the synthesis Lincoln & Guba (1985) Assessment of bias

3.10.3.2 Step 1: developing a theory

Theory development did not play a role in this synthesis because the review was of an exploratory nature.

3.10.3.3 Step 2: developing a preliminary synthesis

The preliminary synthesis involved a tabulation of the findings (Evans, 2002) by extracting the descriptive characteristics of the studies with headings to identify the research aims, methodology, sample characteristics and key outcomes/findings. The tabulation process provided an important initial visual mapping of the studies, and a key building block for the next element of the synthesis process (Evans, 2002). Additionally, differences in study populations, methods of data collection and data analysis were easier to identify, allowing a review and comparison of results between

studies. Where there was a lack of clarity with regard to methodology, for example, contact was made with the primary author.

Using thematic analysis, a recognised analytical method, the researcher was able to identify and analyse preliminary patterns across the studies (Clark and Braun, 2013; Braun and Clarke, 2006). Thematic analysis was chosen because of its suitability to explore people's views and experiences. It is also adaptable to large or small data sets, and has the flexibility to examine qualitative patterns across different research designs without the need for an exploratory meaning framework (Clarke and Braun, 2013).

3.10.3.4 Step 3: exploring relationships within and between studies

The tabulation of the textual description enabled the researcher to explore relationships within and between studies, identifying the challenges and enablers to early dietary intervention, with a focus on how and why a component had a particular impact. The type of nutritional interventions used and the beliefs behind early dietary intervention were identified. The clinical variation in the results of the various research methods, methodologies and participant characteristics were given careful attention throughout the synthesis. After interrogating the literature using thematic analysis and examination of heterogeneity, a conceptual model provided a visual representation of the many factors influencing the CYP with chronic disease and their family, and their early dietary SM of chronic disease. Concept maps can be effectively applied across disciplines to represent changes in knowledge, to help organise and understand new findings (Novak, 1990). Concept mapping gave the researcher control and learning became more meaningful and effective (Novak, 1990).

After several drafts to compare and contrast the views of the stakeholders, concept maps were developed for each theme (Novak, 1990; Clinkenbeard, 1991) (see sections 3.11.4 to 3.11.7). Concept mapping provided a further opportunity to iteratively analyse the data, emphasising the cross-relevance of the many factors.

3.10.3.5 Step 4: assessing the robustness of the synthesis

In order to assess the robustness of the studies included in the review and narrative synthesis, the methodological quality of each research study was appraised using the

Mixed Methods Appraisal Tools (MMAT) (Hong et al. 2018) (Appendix 2), and the Centre for Evidence-Based management (CEBM) critical appraisal checklist (CEBM, 2017). The rationale for using these tools is detailed in section 3.10.1 (Quality assessment of published literature).

In recognition of the need to minimise bias in the included papers, Lincoln and Guba (1985) provide a framework for assessing bias. Qualitative authors (Morrow, 2005; Pandey and Patnaik, 2014) have used this tool. As informed by Lincoln and Guba (1985), critical reflection of each study included in the review sought to address the following:

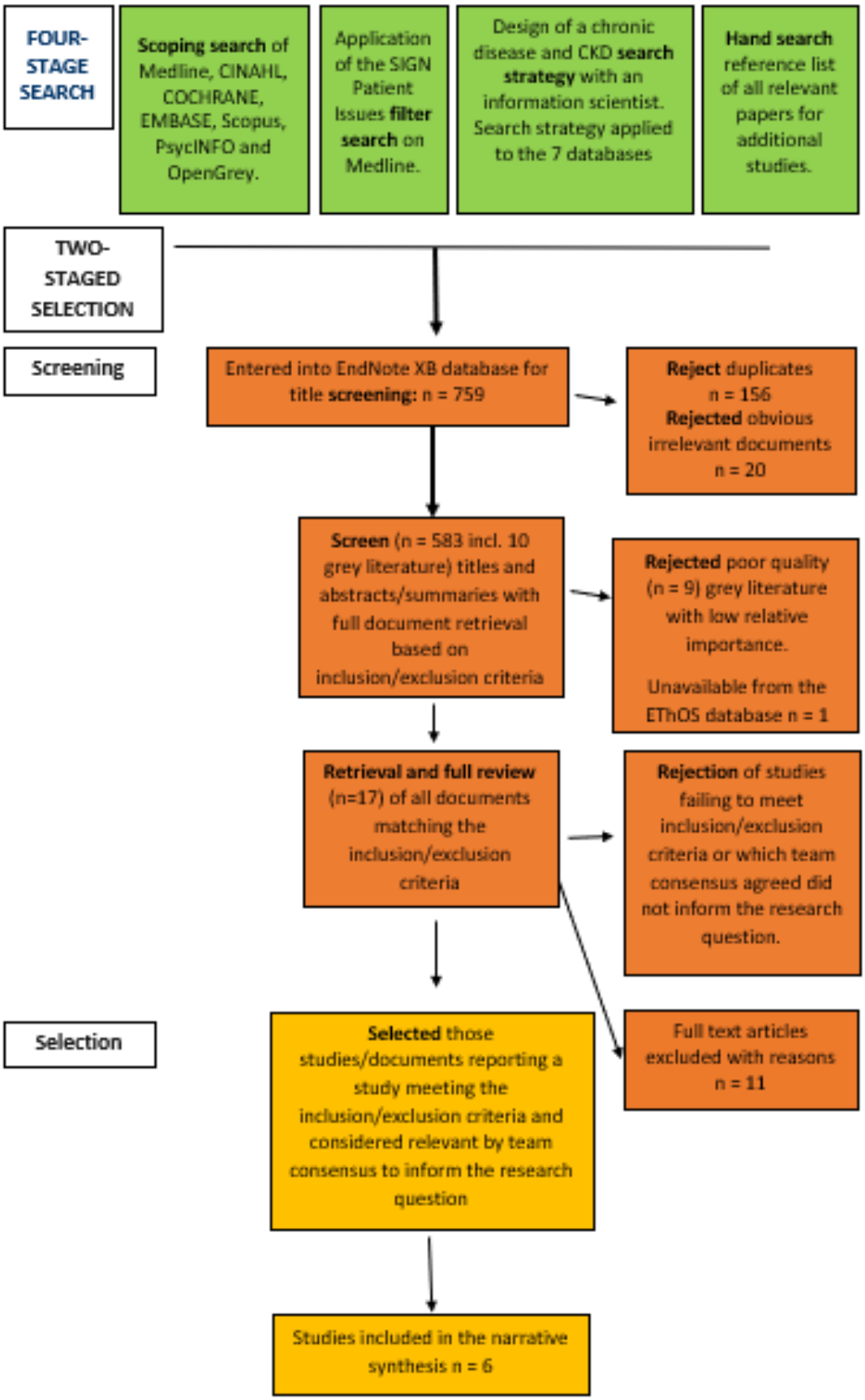
> (1) Identify limitations and their impact on the findings;
>
> (2) Risk of bias by focusing on the quality, validity and generalisability of quantitative research, and focusing on confirmability, transferability, credibility and trustworthiness for qualitative research;
>
> (3) Differences and uncertainties in the findings and how they were addressed; (4) identifying aspects of the research where the evidence was weak or absent;
>
> (5) Suggestions for future research, and finally;
>
> (6) The evidence was synthesised and presented to highlight the 'thick' and 'thin' findings, key differences and strengths. Providing a 'thick description' in the study proceedings means that sufficient detail is given to evaluate the extent to which the findings can be transferred to other situations, settings and populations (Lincoln & Guba, 1985).

3.11 Findings

The search identified 759 articles. When initially screened against the screening and selection criteria checklist in Table 16, p. 58, a total of 176 (156 duplicates and 20 clearly irrelevant documents) were excluded. The remaining 583 articles were screened for eligibility against the inclusion and exclusion criteria, and a total of 566 articles were excluded. A retrieval and full-text review was undertaken for 17 articles. A total of 6 articles (1 mixed method, and 5 quantitative) were selected for the final narrative synthesis.

The PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) flow diagram (Figure 5, p. 65) presents the results of the search process (PRISMA, 2015).

Figure 5. PRISMA flow chart sequencing the review process (2015)



Grey literature

Ten of the 759 articles retrieved from the database searches were grey literature. Initial screening excluded eight articles, with two articles screened against the inclusion and exclusion criteria. A full-text copy of the one article, a PhD thesis (Poustie, 2003), which met the inclusion/exclusion criteria and was considered relevant by the reviewers, was requested from the library but was not available from the EThOS database.

3.11.1 Summary of the included studies

Key characteristics of the six studies included in the narrative synthesis are detailed in Table 21, p. 67. The studies comprised a heterogeneity of settings: one in an elementary school (Coleman et al. 2010), three in a specialist clinic for chronic disease (Shroff et al. 2012; Stapleton et al. 2002; Ball et al. 2005), one of which included an educational home visit (Stapleton et al. 2002), and two in a children's in-patient ward (Niggemann et al. 2001; Totadri et al. 2017). The study groups included Hispanic (Coleman et al. 2010), Caucasian (Shroff et al. 2012), Australian (Stapleton et al. 2002), European (Niggemann et al. 2001), and American and Indian populations (Ball et al. 2005; Totadri et al. 2017).

Table 21. Study characteristics of all included article (mixed methods, qualitative and quantitative)

Author, Pub year	Study aim	Methodology	Sample characteristic	Key outcomes/findings
Study characteristics of the mixed method study				
1. Coleman *et al.* 2010	Families with at least one child aged 8-12 years at risk of developing type 2 diabetes mellitus attended ten 90-minute sessions with exercise, cooking demonstrations, and healthy lifestyle lessons. Weight and height were recorded at the start and finish of the 10-week program. Parents and children completed a quantitative health behavior and program knowledge survey, and qualitative interviews were undertaken with a selection of parents. The sequential mixed method study sought to test: (a) the effectiveness of a minimal, tailored diabetes prevention program that could be delivered in a school setting; and understand: (b) the parents' perceptions of their child's behavior and risk of disease; and (b) how their child's risk of disease and health habits changed as a result of the program.	Self-reported questionnaires/structured interviews to complete the questionnaires. Questionnaire was based on the International Physical Activity Questionnaire, the Behavioural Risk Factor Surveillance Survey, and the San Diego Health and Exercise Survey. Structured interviews with parents. Interviews were transcribed and analyzed into themes.	62 children aged 7.5 ± 3.1 years old attending an elementary school, and their parents (n=82) aged 37.5 ± 8.6 years (98% women) enrolled in the program. As all families were Hispanic, classes were delivered in Spanish.	Thirty-eight families completed at least 5 of 10 classes. Parents had an average weight loss of 1.5lbs (61% lost some weight) from baseline to the end of the program. Parents were more likely to believe that their child's weight led to serious chronic illnesses after participating in the program. There was a large increase in the number of parents who self-reported engaging in leisure-time physical activity and eating more servings of vegetables as a result of the program. Women had a greater understanding of their control over what they gave their children and felt empowered to change their family's health. Women enjoyed the hands-on cooking sessions and social support. Children worked with parents to make change happen in their family.

Author, Pub year	Study aim	Methodology	Sample characteristic	Key outcomes/findings
Study characteristics of quantitative studies				
2. Shroff *et al.* 2012	The study aimed to compare the effect of a daily dose of ergocalciferol (or a matched placebo) in children with CKD 2-4, on the time to development of hyperparathyroidism	A randomized, double-blinded, placebo-controlled study. Experimental intervention consisted of ergocalciferol (2000IU)/ml in sunflower oil). The control group were given a placebo of sunflower oil only which was matched for colour, odour and taste for 3 months, with a 12 month follow up period	72 children were screened and 42 aged <18 years with an estimated GFR between 15-70ml/min per 1.73m^2, and normal intact PTH levels but low 25(OH)D levels were enrolled in the study from a specialist renal clinic. 20 in each arm completed the study.	Nine of 20 children on placebo and 3 of 20 children on ergocalciferol developed hyperparathyroidism. The time to development of hyperparathyroidism was significantly longer in children with ergocalciferol treatment compared with placebo. All of the early stage (CKD 2-3) patients in the treatment group, except one child with CKD 4 achieved a normal 25(OH) D level.
3. Stapleton *et al.* 2001	The study aimed to assess the impact of a 'Go and Grow with CF' intervention program on nutrition and pancreatic enzyme knowledge and self-management skills in children with cystic fibrosis.	A randomized controlled trial. Separate validated questionnaires were used for face-to face interviews with children and for telephone interviews with carers on three occasions: pre-intervention, immediately post-intervention and 12 months post-intervention.	59 children aged 2-11 years and their primary carer were enrolled in the clinical trial from a specialist CF clinic. 42 children and 55 carers participated in the program.	Children's knowledge in the intervention group improved significantly immediately post-intervention, but this did not remain at the 12-month follow-up. Children's knowledge, both in the intervention and control group was related to their appropriate self-management score immediately and 12 months post-intervention. The program was found to be effective in increasing children's knowledge in the short-term.
4. Niggemann *et al.* 2001	The study aimed to assess the effectiveness of an amino-acid-based infant formula versus an extensively hydrolyzed whey formula in infants with cow's milk allergy/intolerance and atopic dermatitis.	Prospective, controlled, multi-center trial. Experimental intervention consisted of receiving an amino-acid formula for 6 months. The control group received an extensively hydrolyzed whey formula. Clinical symptoms of atopic dermatitis, dietary intake, and weight were monitored.	73 infants between 1-10 months were enrolled in the study from in-patient ward. All infants suffered from atopic dermatitis and had a cow's milk allergy/intolerance confirmed by double-blind, placebo-controlled food challenge.	All children experienced an improvement in their eczema symptoms in response to dietary change. The use of an AA base formula achieved a statistically significant increase in length.

Author, Pub year	Study aim	Methodology	Sample characteristic	Key outcomes/findings
5. Ball *et al.* 2005	The study aimed to determine the nature and prevalence of dietary supplement use among chronically ill children. Questions inquired about the current and previous year's dietary supplement use, and frequency. Participants expected and observed benefits and side effects of their child's supplement use. Cost of supplements, and the health providers awareness of their supplement use.	A quantitative descriptive design involved parents completing a questionnaire consisting of 26 open-and closed-ended questions during their child's clinic visit.	Parents of 505 chronically ill children and adolescents with a mean age of 9.9 ±5.3 years were recruited from the medical clinics in Salt Lake City. UT. Chronic illnesses included asthma, type 1 diabetes, cancer, leukemia, renal and liver transplantation, seizure and rheumatological disorders, cystic fibrosis, and neurobehavioural disorders.	Sixty-two percent of the population used dietary supplements, and 30% used non-prescribed supplements. Use varied amongst the diagnosis groups, with the highest non-prescribed use amongst children with cancer, neurobehavioral and rheumatological disorders. Supplements were primarily given to improve general health, supplement the diet and prevent disease. Of children receiving non-prescribed supplements, only 20% had discussed the use with their child's health care provider. Parents sited books and magazine as their primary source of supplement information.
6. Totadri *et al.* 2017	The study sought to evaluate the level of knowledge regarding nutrition and prevalent misconceptions about diet in families with children undergoing therapy for cancer.	A descriptive, cross-sectional questionnaire-based study was used to gather data from parents of children attending a children's cancer ward.	Mothers of 72 children with a median age of 8 years were enrolled for the study	Twenty percent of parents perceived diet to be causally related to cancer. Dietary restriction was practiced by 57% of mothers, with junk foods and chicken the most commonly avoided foods. Certain foods were believed to have health benefits. Disparity existed between the nutritional status of the children and maternal perception.

3.11.2 Quality assessment findings for each study

To provide continuity across the review both quality assessment tools were applied (Hong et al. 2018; CEBM, 2017) to each study (see section 3.10.1). Due to its purpose built design the MMAT proved more useful in probing the key components of the MM study, whilst concurrently addressing the quantitative and qualitative component separately (Hong et al. 2018) (Appendix 2). As argued by Hong et al. (2018), despite the variability in methodological quality, all of the studies should be included in the review. However, alongside the overall score from the quality assessment criterion, an evaluation of the weight and strength of the evidence from each study, for each theme, provides an overall assessment of the quality of the synthesis. An example is provided in Appendix 5 of the calculation of methodological quality for the Coleman et al. (2010) study. Refer to Table 22 for the amalgamated quality assessment data.

Table 22. Methodological quality assessment for each included study

Author pub year	MMAT (Hong et al. 2018) quality rating total	CEBMa (2017) quality rating total
Coleman et al. (2010)	5/11 (45%) LOW-MEDIUM	7/12 (58%) MEDIUM
Shroff et al. (2012)	7/7 (100%) HIGH	10/12 (83%) HIGH
Stapleton et al. (2001)	5/7 (71%) MEDIUM-HIGH	7/12 (58%) MEDIUM
Niggemann et al. (2001)	4/7 (57%) MEDIUM	8/12 (66%) MEDIUM-HIGH
Ball et al. (2005)	2/7 (28%) LOW	4/12 (33%) LOW
Totadri et al. (2017)	3/7 (43%) LOW-MEDIUM	3/12 (25%) LOW

In order to assess the 'thick' and 'thin' findings (Lincoln and Guba, 1985) Table 23, p. 71 provides a matrix of the evidence extracted from each study to support each theme.

Table 23. Matrix providing evidence from the studies to support each theme

Study	Coleman et al. (2010)	Shroff et al. (2012)	Stapleton et al. (2001)	Niggemann et al. (2001)	Ball et al. (2005)	Totadri et al. (2017)
Theme 1 - Role of education in early dietary management	Evidence of improved parental knowledge regarding healthy lifestyle, diabetes and chronic disease prevention from pre to post intervention supported this theme. Parents believed education gave them increased control over their child and family's food choices, improving chronic disease awareness, reducing fear associated with the disease and imparted strength to empower friends and family to support disease management.	The provision of education to deliver the early dietary intervention achieved and maintained a delay in the progression of CKD.	Dietary education and behavioural change strategies for the children increased knowledge in all areas (enzymes, fat, energy and salt), but the impact on children's self-management and communication with carers was short-term, evidencing the need for ongoing education. Learning and behaviour change required dedicated input from HCPs, using varied, age-appropriate resources.	Education increased parent's awareness of the need to make early dietary changes.	There was 'thin' evidence in the study of the role of education, rather the sources of education were detailed, namely, books and magazines, retail outlets, personal research and HCPs.	Most parents reported having received some dietary education, which motivated a change in diet. Dietary education was an important weekly timetabled event and played a role in helping to clear dietary misunderstandings.
Theme 2 - Parents/caregivers role in the early dietary management of chronic disease	Parents and carers played a key role in early dietary self-management. Women were identified as the main leaders of diet/lifestyle change; because of their primary involvement in cooking, the practical cookery workshop proved successful in engaging the women.	Parents were instrumental in ensuring the child attended regular review appointments, completed dietary records and administered the preparation as prescribed.	Parents played a crucial role in the programme. The flexible home-based learning package enabled the program to be tailored to individual parental/family circumstances. Parents and carers perceived that they and their children's knowledge increased because of completing the program.	Parents played the leading role as the primary caregivers in administering the dietary intervention and monitoring tolerance. Regular food records were completed and parents were responsible for bringing their child to the routine review appointments.	Parents were found to initiate or discontinue the administration of dietary supplements, particularly in the management of a chronic disease, and if they were already taking the supplement. Transparency with HCP regarding the use of dietary supplementation was affected by parental perception of its importance, perception of the physician's response, or not being asked.	Mothers, the primary adult participants and the main carers, held powerful views about the role of diet in the development of cancer and were influential in changing their child's diet once cancer treatment had commenced started.

Theme 3 - Early dietary SM of chronic disease	Three key impacts of early dietary self-management were found: a majority lost weight, 50% increased their engagement in leisure-time physical activities, and families better understood the need for regular reviews by the HCP.	Early dietary self-management of chronic disease Early dietary intervention was strongly found to promote a normal 25-hydroxyvitamin D levels in all early stage CKD patients, and 80% of the overall patient cohort receiving the dietary supplementation.	Early dietary intervention was found to be heavily dependent on knowledge acquisition. Self-management skills and communication improved in the short but not long-term. The program was found to have the potential to delay disease progression.	Early dietary self-management was found to improve eczema related symptoms in all of the children; achieving a statistically significant increase in length which was suggested to overcome any growth impairment.	Dietary supplements (both prescribed as a component of conventional medical treatment and non-prescribed products) were provided in the management of chronic disease. Children with cancer were the primary recipients of both prescribed and non-prescribed dietary supplements. The amount (1-9) and variety (198 different products) of dietary supplements given was vast. The monthly cost of dietary supplementation ranged from USD10 and >USD100.	There was some evidence of HCP promoting early dietary self-management, encouraging more home-prepared foods as part of a healthy diet, rather than the using 'special' commercial prepared products, and dispelling myths regarding diet and the aetiology of chronic disease.
Theme 4 - Barriers and enablers to early dietary change	Retention of parents and children at classes was difficult, however this was improved by reminder phone calls. Recognising and rewarding participant's achievements increased satisfaction and confidence. Husbands and extended family were described as the hardest barriers to change.	Compliance with the prescribed preparation was good, but some families found them to be a potential 'burden of care'. The effectiveness of the intervention was measured by the taking of several blood tests; recognised as an invasive procedure.	The program's effectiveness was based on HCPs regular input to support long-term knowledge acquisition and retention of self-management skills. Including a home-based course allowed individual family circumstances to be considered. A dietitian and clinical psychologist developed the program, which enabled it to be tailored to all socioeconomic groups. The program lacked dietary records and proposed age-appropriate resources as an instrument to enhance engagement and interest.	The dietary intervention required parents to keep detailed dietary records, and attend routine clinic appointments to monitor their child's weight and length. The amino acid infant formula was expensive.	A small number of side effects to dietary supplementation were found. Cost was recognised as a factor that influenced the use of dietary supplements. HCPs recognised the need to be more aware of the use of dietary supplements by patients due to limited safety and efficacy data in relation to drug interaction, growth and development. Parental motivations for giving dietary supplementation, included, ethnic or cultural background, socioeconomic status, and access to medical and alternative resources.	There was little focus on the barriers and facilitators of early dietary self-management. HCP were key facilitators providing dietary support and dispelled dietary myths.
Levels of 'thick' and 'thin' evidence to support each theme	Low level of 'thick' evidence	High level of 'thick' evidence	Moderately 'thick' evidence	Moderately 'thick' evidence	'Thin' evidence for theme 1 and 'thick' evidence to support the other themes	'Thin' evidence for theme 4 and moderately 'thick' evidence to support the other themes

In the next section, each of the four themes is discussed in turn, with a concept map highlighting the key factors discussed throughout the studies, bringing together steps 2-4 of the narrative synthesis,:

Step 2: developing a preliminary synthesis
Step 3: exploring relationships within and between studies
Step 4: assessing the robustness of the synthesis

3.11.3 Structured thematic analysis summary and narrative synthesis themes

Through a cyclical interweaving of the narrative synthesis steps 2 and 3, descriptive characteristics were tabulated and thematic analysis allowed terms/phrases to emerge to support and affirm a similar narrative across the studies. Table 24 illustrates the emergence of 'education' as a common theme across the studies.

Table 24. The development of Theme 1 – the role of education in early dietary management

Studies / Theme	Niggemann *et al.* 2001	Stapleton *et al.* 2001	Coleman *et al.* 2010	Shroff *et al.* 2012	Totadri *et al.* 2017	Ball *et al.* 2005
Education		Education can increase parent's and the child's awareness of the importance of regular check-ups to prevent disease			Majority (89%) received dietary education	Books and magazines were the primary sources of education about dietary supplement (78.4%)
		Children respond to education with a short-term change in knowledge	Women said that they no longer feared diabetes and felt more empowered to help their family and friends cope with the disease			
		Education increased children's engagement and communication with carers regarding SM				
	Management of CD was positively impacted by education					
		Dietary and lifestyle education programmes use a variety of methods to engage children and parents				
		Dietary education can increase confidence in the management of a CD	Education increased women's awareness that contact with the HCP did not only have to be at a crisis or routine check-up.			

Analysis of the six studies led to the development of four interrelated themes:

Theme 1 - the role of education in early dietary management
Theme 2 - parents/caregivers role in the early dietary management of chronic disease
Theme 3 - early dietary intervention in the management of chronic disease
Theme 4 - barriers and enablers to early dietary change.

Table 25, p. 74 displays these four themes and their relationship across the studies

Table 25. Four themes and their relationship across the studies

Author, citation	Study design and broad classification	Sample and population group	Theme 1: Role of education in early dietary management	Theme 2: Parent/caregivers role in the early dietary management of chronic disease	Theme 3: Early dietary self-management of chronic disease	Theme 4: Barriers and enablers to early dietary change
Mixed method study						
Coleman et al. 2010	1. Mixed methods study with self-reported questionnaires and/or structured interviews 2. Tailored diabetes prevention program	1. Purposive random sample of eligible parents and children 2. Both African American and Hispanic parents showed interest in the program, but all families that enrolled were Hispanic, with a preference to speak Spanish (classes delivered in Spanish)	1. Parent's knowledge regarding healthy lifestyle, diabetes and chronic disease prevention increased from pre to post intervention 2. Education gave parents more control over what their children were consuming and felt empowered to change their family's health 3. Knowledge reduced fear of diabetes and felt more empowered to help their family and friends cope with the disease 4. Education improved chronic disease awareness	1. 98% of the parents engaging with the program were women 2. Parents had a mean age of 37.5 years 3. Women were the main leaders of diet/lifestyle change in the family 4. Women enjoyed hands on cooking workshops, and the social support from other parents who shared their stories	1. 61% of parents lost some weight, with just < 10% moving to a lower obesity category 2. 50% increase in engagement in leisure-time physical activities as a result of participation in the program 3. Families better understood the importance of regular check-ups, and contact with healthcare professionals did not only have to be when there was a crisis or acute illness	1. Most parents attended on average 4/10 classes 2. Reminder phone calls were very helpful and encouraged parents/children to attend classes 3. Women enjoyed receiving a diploma for completing the program. 4. Children felt special when they received their own certificate 5. Women believed the hardest barriers to changing their lifestyles involved their husband and extended family's resistance 6. Access to workshops and classes was limited by domestic responsibilities

Quantitative study						
Author, citation	**Study design and broad classification**	**Sample and population group**	**Theme 1: Role of education in early dietary management**	**Theme 2: Parent/caregivers role in the early dietary management of chronic disease**	**Theme 3: Early dietary self-management of chronic disease**	**Theme 4: Barriers and enablers to early dietary change**
Stapleton et al. 2001	1. A randomised controlled trial with a dietary education and behavioural change intervention program, followed by 3 knowledge and self-management skills questionnaires. 2. The impact of a 'Go and Grow with CF' intervention program	1. A randomised purposive sample of children with cystic fibrosis 2. Australian	1. The dietary education and strategies for behavioural change intervention program for the children increased knowledge in all areas (enzymes, fat, energy and salt) 2. The intervention increased children's knowledge and self-management skills and communication with carer's in the short-term but not long-term 3. Regular ongoing education and counselling is required, which may support knowledge retention long-term 4. The program nurtured desirable behaviour change through a six-step program each week, which allowed the children to apply their knowledge. 5. To retain interest, education should use a variety of age-appropriate materials and programs on a rotational basis.	1. Parents found learning in the home environment to be a flexible feature of the program 2. Parents and carers perceived that they and their children's knowledge increased because of completing the program.	1. An individual's ability to perform certain behaviours was dependant on the acquisition of adequate knowledge. 2. The intervention increased the number of respondents demonstrating appropriate self-management skills/knowledge in the short but not long-term 3. The improvement in communication as a result of the program may improve clinical outcomes 4. The Go and Grow with CF material could be used as a preventative program to delay disease progression	1. Regular input from healthcare professionals is required to support long-term knowledge and self-management skills 2. The program involved a home-based course which was well received 3. The program was developed by a dietitian in consultation with a clinical psychologist 4. The program was delivered to families across the socioeconomic index 5. A detailed diet dietary would have supported the effectiveness of the intervention, but this was not undertaken. 6. The use of age-appropriate materials might help maintain engagement and interest in the program.

Quantitative study						
Author, citation	**Study design and broad classification**	**Sample and population group**	**Theme 1: Role of education in early dietary management**	**Theme 2: Parent/caregivers role in the early dietary management of chronic disease**	**Theme 3: Early dietary self-management of chronic disease**	**Theme 4: Barriers and enablers to early dietary change**
Niggemann et al. 2001	1. A prospective, controlled multi-centre experimental intervention trial 2. The effectiveness of an amino-acid formula on clinical symptoms, dietary intake and growth.	1. A randomised purposive sample of infants with cow's milk allergy/intolerance 2. Germans	1. Education can increase parent's recognition of the need for dietary change	1. Parents were required to complete a 3-day food diary and attend the hospital for growth measurements regularly over a 6-month period. 2. Parents reported the amino-acid formula to be tolerated and well-accepted by the infants	1. In response to a change in their diet all children experienced an improvement in their eczema related symptoms 2. Dietary change achieved a statistically significantly increase in length. 3. Commencing the AA-formula early in the chronic disease may outgrow their growth impairment	1. Dietary intake using a 3-day food diary, and weight and length were monitored over a 6-month period 2. The special AA-formula is expensive.

Quantitative study						
Author, citation	**Study design and broad classification**	**Sample and population group**	**Theme 1: Role of education in early dietary management**	**Theme 2: Parent/caregivers role in the early dietary management of chronic disease**	**Theme 3: Early dietary self-management of chronic disease**	**Theme 4: Barriers and enablers to early dietary change**
Ball et al. 2005	1. A quantitative descriptive design using a self-reported questionnaire consisting of open and closed-ended questions 2. Assessment of the prevalence and nature of dietary supplement use among chronically ill children	1. A convenience sample of parents of children and adolescents with a variety of chronic diseases. 2. European-American, Hispanic, biracial, American Indian, Asian, African American or Pacific Islander	1. Books and magazines were the primary source of supplementary information about dietary products for chronic disease. 2. Other sources of education and information included shops, personal research, pharmacist, dietitian and other healthcare professionals or facilities.	1. Parents main reason for giving dietary supplements was to improve general health, supplement the diet and prevent disease 2. Parents were more likely to have given or currently be giving their child a dietary supplement if their child had a chronic illness. 3. Parents giving their child a dietary supplement were more likely to use or have used one themselves. 4. Parents reported the reason for discontinuing with dietary supplement use was: (a) the supplement was no longer required; (b) child's physician advised against its use; (c) there was a lack of perceived benefit; (d) the child did not like or tolerate the supplement, and (e) the supplement was too expensive 5. Some parents discussed the use of non-prescribable dietary products with the child's physician. Other who had not discussed their use, did not do so as they did not feel it was important, or perceived the physician would react negatively, or just because they weren't asked about the use of dietary products. 6. Mean age of parents was 37.2 and 40.1 years for mothers and fathers, respectively.	1. Diet supplements were used for children with a range of chronic diseases. 2. Dietary supplements consisted of both prescribed as a component of conventional medical treatment and non-prescribed products 3. Children with cancer used the most non-prescribed supplements 4. The highest use of non-prescribed supplements was amongst children with cancer. 5. 1-9 dietary products were used regularly 6. Dietary products ranged from dietary supplements, vitamins and minerals, botanical preparations to amino acids and other products. 7. The most commonly used products were disease-specific herbal or vitamin preparations 8. Dietary supplement users spent between $10 and >$100 per month on products 9. The study identified the use of 198 different dietary products	1. Very few side effects were noted. Side effects recorded included loose stools, nausea and vomiting. 2. Cost was recognised as a factor influence the use of non-prescribed dietary supplements 3. The need to be aware of patients' possible use of prescribed and non-prescribed dietary supplements. 4. Dietary supplements use may raise concerns due to limited information about safety and efficacy in relation to drug interaction, growth and development. 5. Many factors influence a parent's belief in or desire to use dietary supplements, including ethnic or cultural background, social class, socioeconomic status and the availability of medical and alternative resources.

Quantitative study						
Author, citation	**Study design and broad classification**	**Sample and population group**	**Theme 1: Role of education in early dietary management**	**Theme 2: Parent/caregivers role in the early dietary management of chronic disease**	**Theme 3: Early dietary self-management of chronic disease**	**Theme 4: Barriers and enablers to early dietary change**
Totadri et al. 2017	1. A descriptive, cross-sectional structured questionnaire-based study. 2. Evaluate the level of knowledge regarding nutrition and misconceptions about diet in the treatment of children undergoing therapy for cancer.	1. A purposive sample of parents of children attending a children's cancer ward for treatment. 2. Indian	1. Majority of parents had received some dietary education. 2. Education on diet and hygiene was provided weekly by the paediatric oncologist, dietitian, psycho- oncologist and nursing staff. 3.Education helped to clear misconceptions related to diet 4. Education focused on encouraging a healthy diet now rather than being concerned about previous dietary intake.	1. Parents (20%) believed diet to have contributed to the development of the cancer. 2. Mothers had made significant changes in their child's diet after starting treatment for cancer. 3. The main foods parents related issues attributed to be possible triggers for cancer included large amounts of milk products and junk food, and inadequate feeding. 4. Other foods that parents avoided during therapy included chicken, mutton, street food, oily foods, fish, raw foods, nuts and meat of any kind. 5. Foods parents believed to be beneficial and actively encouraged during therapy included, vegetables, eggs, fish, meat, finger millet, commercially available protein supplement powders, pulses, nuts, milk, pomegranate and mushrooms 6. Parents believed childhood cancer and nutrition are directly related. 7. Mothers were the main carers who participated in the study.	1. Healthcare professional must emphasise intake of commonly available foods and more home-prepared foods as part of a healthy diet, rather than the addition of 'special' foods to the diet. 2. Healthcare professional needs to dispel the myths regarding diet and chronic disease development and focus on the multifactorial aetiology of chronic disease.	1. Regular interaction with healthcare professionals can support the intake of a healthy diet and help to dispel misconceptions. 2. Commercially available protein powders were often believed to provide special therapeutic benefits

3.11.4 🔵 Theme 1: The role of education in early dietary management

Research studies included in this systematic review all recognised the vital role of education in enhancing the knowledge and awareness of early dietary management in the treatment of chronic disease. Coleman et al. (2010) and Stapleton et al. (2001) showed within their mixed-methods and quantitative study, respectively, that dietary education and lifestyle change strategies had increased the knowledge and SM skills of parents and children. Adapting a program to the individual needs of the population supported engagement. Coleman et al. (2010) delivered a tailored diabetes prevention program to families in their native language (Spanish), at their child's elementary school. The educational classes gave parents more control over what their children ate, enabling them to improve the health of their families (Coleman et al. 2010). Parents also reported that their newly found dietary knowledge reduced fear and increased awareness of and management of chronic disease (Coleman et al. 2010).

The Australian study 'Go and Grow with CF' by Stapleton et al. (2010) provided children and their parents with a nutrition and pancreatic enzyme knowledge and SM skills education program at home. A six-step program enabled children with cystic fibrosis (CF) and their caregivers to apply their knowledge using the constructs of social learning theory. Education improved children's understanding and knowledge base, which improved their SM involvement and communication with their parents and caregivers on disease management (Stapleton et al. 2001). Long-term retention of knowledge was poor, recognising the need for regular, ongoing education and advice, using a variety of age-appropriate materials to support better long-term results (Stapleton et al. 2001). The tailoring of the program to support families across the socioeconomic index also helped to maintain engagement and interest (Coleman et al. 2010). Providing a weekly education forum with the opportunity to receive advice on diet and hygiene for families of children with cancer encouraged positive commitment to education (Totadri et al. 2005).

The retention of participants can be a challenge and it is important to find ways to keep families involved. Adults attending the diabetes prevention classes (Coleman et

al. 2010) received incentives such as insulated lunch bags, hats, hand lotion, sunscreen and toys to encourage class participation, reward attendance and for bringing a healthy snack. Healthy snacks such as fruit, cheese sticks and yogurt were provided at every session.

The delivery of prescribed nutritional products requires training and advice on purpose, dose and follow up plans (Shroff et al. 2012; Niggemann et al. 2001). An interventional study with early stage chronic kidney disease patients required that children take a daily dose of an ergocalciferol (vitamin D) supplement with regular blood tests to evaluate its efficacy. Comprehension of 25-hydroxyvitamin D [25(OH) D] supplement supported concordance and the delayed development of secondary hyperparathyroidism[1] (Shroff et al. 2012). The German interventional study (Niggemann et al. 2001) showed how infants with severe atopic dermatitis and proven cow's milk allergy/intolerance, who followed a prescribed amino-acid based formula diet, experienced an improvement in their symptoms associated with their eczema.

Access to credible education and advice can help to eliminate misunderstandings and myths. The survey of parents of 505 chronically ill children and adolescents in Salt Lake City, Utah (Ball et al. 2005), found that books and magazine were the primary source of dietary supplementation information for chronic disease management. Parents sought expert advice from HCPs such as the dietitian, pharmacist and clinician, but often after personal research. In the Indian study (Totadri et al. 2017), many parents of children with cancer based their dietary beliefs on misconceptions about foods with "special" properties to increase the number of white blood cells. By providing evidence–based advice, HCP can dispel myths about diet and chronic disease, focus on the multifactorial aetiology of chronic disease (Totadri et al. 2017), and discourage the use of large quantities of expensive, often non-prescribable dietary products, as detailed in the US study (Ball et al. 2005). Knowledge gave

[1] Secondary hyperthyroidism occurs in CKD when an increase in parathyroid hormone secretion occurs because of a reduction in serum calcium because of various factors that include reduced renal excretion of phosphate, vitamin D deficiency and impaired ability of the kidney to hydroxylate that vitamin D.

children the confidence to be more persuasive with grandparents about their health problems and longstanding beliefs (Coleman et al. 2010).

The development and delivery of dietary education by experts in the field was demonstrated in both the Australian CF study (Stapleton et al. 2001) and the Indian parental survey (Totadri et al. 2017) of children undergoing cancer treatment, in which dietitians, psychologists and other specialist HCPs played a key role in the education strategies. This can contribute to keeping a program relevant and realistic once the class is over. It is important to build on current knowledge and life experience, and develop educational programs that enable children and parents to replicate the advice easily in their own home/life environment. During the cooking workshops in the diabetes study (Coleman et al. 2010) hands-on practical advice was given about preparation, accessibility and affordability. In the Indian study (Totadri et al. 2017) parents were encouraged to use everyday home based ingredients to promote a healthy diet and avoid costly, commercially prepared dietary products or unnecessary dietary restrictions. Providing culturally appropriate recipes for parents sought to motivate ongoing habits (Coleman et al. 2010). Education can be affirmed by giving children and parents' opportunity to support each other through social networking and the sharing of stories/experiences. This was demonstrated in the diabetes prevention program (Coleman et al. 2010).

The different coloured units indicate a relationship across the themes. See Table 26 for the colouring coding used in the concept maps.

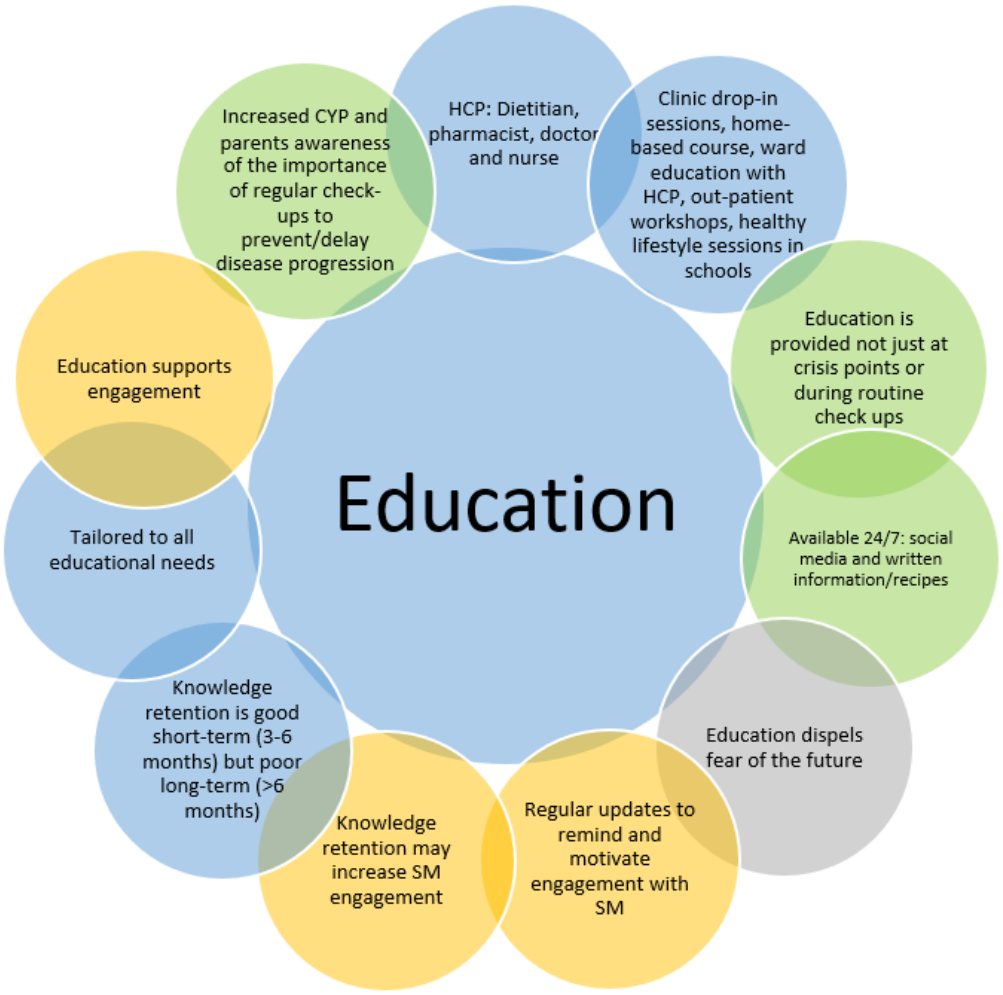
Table 26. Themes and colour coding in the concept maps

Theme 1	Education	blue
Theme 2	Parents/caregivers	green
Theme 3	Early dietary self-management of chronic disease	orange
Theme 4	Barriers and enablers to early dietary change	grey

The following concept map (see Figure 6) captures some of the key sources and facets of education, and the different coloured units indicate a relationship across the themes. Education, a central component of a dietary self-management strategy,

requires working in partnership with the parents/caregivers, to utilise the enablers, and overcome the challenges to effect long-term change. However, to have lasting impact it needs to be accessible, frequently updated, and tailored to the evolving and changing needs of the CYP and their family. Education can increase awareness, motivating action, which can positively impact self-management.

Figure 6. Key sources and facets of education and their relationship with other themes



3.11.5 Theme 2: Parents/caregivers role in early dietary SM

This theme focused on the role and profile of parents and caregivers in the early dietary SM of children with chronic diseases. Although the education sessions and dietary intervention strategies were mostly tailored to appeal to all family members, the studies reported limited attendance or reference to male caregivers. Only 2% (n=2) of the parents who participated in the diabetes prevention program were men (Coleman et al. 2010). This may reflect work commitments in this Californian, Hispanic population, and the perception that changes in diet and lifestyle change should be directed towards women because of their culturally dominant involvement in childcare and meal preparation. This was shown again in the purposive sample for the Indian study, in which only mothers were recruited (Totadri et al. 2017). These observations suggest that mothers are often the leaders of a family's dietary change. Interestingly, in two of the studies in which the mean age of enrolling parents was indicated, 37.5 years (Coleman et al. 2010) and 37.2-40.1 years (Ball et al. 2001) were comparable.

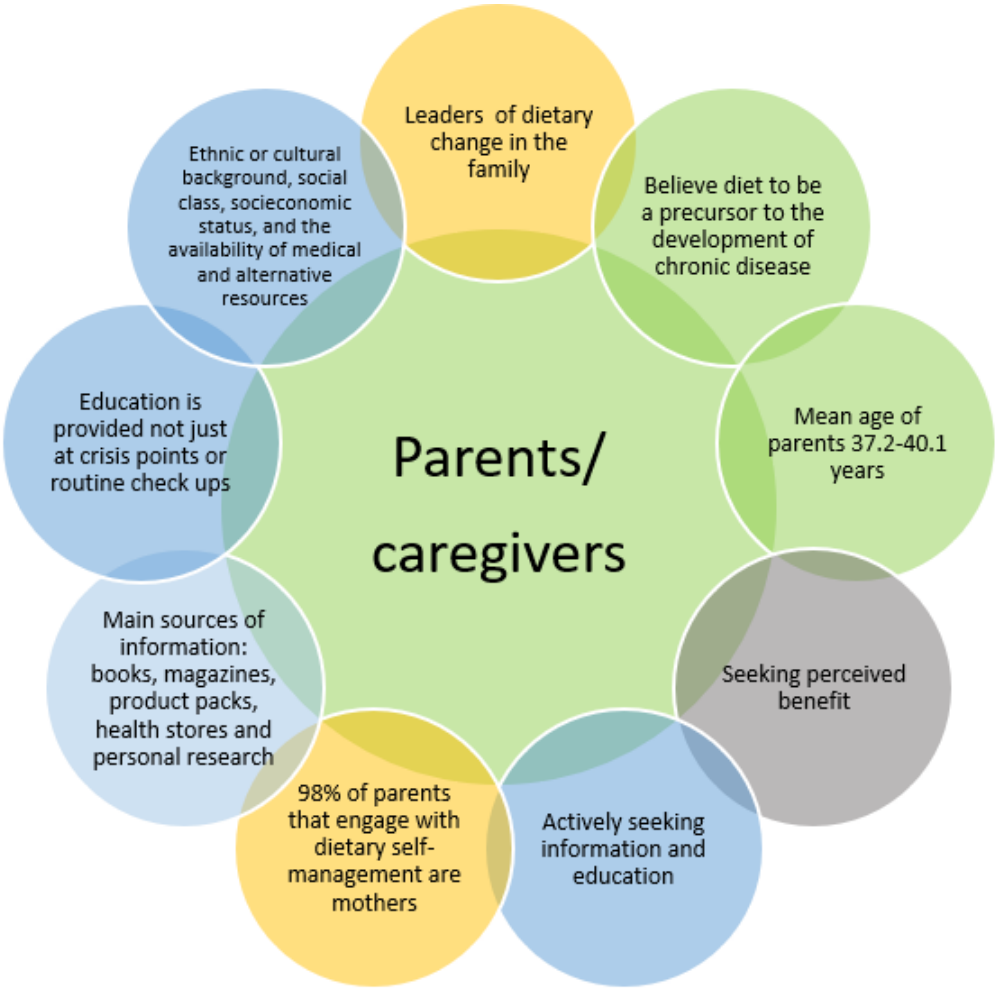
To monitor the efficacy of dietary interventions parents and children were often required to keep daily records of the child's intake for future analysis. In both the UK (Shroff et al. 2012) and German (Niggemann et al. 2001) study of CKD and infant atopic dermatitis sufferers, respectively, completion of a 3-day food diary was carried out to assess the child's intake of calcium and vitamin D, and overall nutritional intake to support growth. Tolerance and palatability of the amino-acid formula required descriptive feedback from parents to ensure that infants (Niggemann et al. 2001) took prescribed dietary preparation appropriately. The efficacy of treatment with CKD children depended on the parents' concordance with the prescribed ergocalciferol dose, confirmed by parental feedback. The understanding of the dietary product by caregivers will influence their level of engagement. Other factors that influenced the parents' belief or desire to provide their child with a dietary supplement included ethnic or cultural background, social class, socioeconomic status, and the availability of medical and alternative resources (Ball et al. 2005).

Engaging with the whole family enables them to learn together. It can encourage and motivate long-term change by sharing experiences and learning from other families in the same situation as them. The flexible feature of the CF intervention (Stapleton et al. 2001) meant that children and parents took part in a home-based course, sandwiched between an introductory and concluding group workshop with other families. The home-based course allowed the teaching to be tailored to the bespoke needs of the family. Parents felt that their children's knowledge of nutrition and enzymes increased following completion of the program. This was confirmed by a statistically significant increase in their short-term pre-and post-intervention knowledge scores *(p=0.001)*. Engagement with the whole family proved to be a positive step in the diabetes prevention program, where 61% (n=23) of the parents with pre and post program height and weight data (n=38) achieved a weight loss, with 18% (n=7) moving to a lower obesity bracket (Coleman et al. 2010). Leisure-time physical activity increased by 36% in children and 50% in parents because of the program. Families better understood the importance of regular check-ups, and contact with the HCP did not have to be when there was a crisis or acute illness.

Building a good working relationship with the HCP can influence caregivers' concordance and honest communication. In the US survey (Ball et al. 2005) some parents had talked to their child's doctor about the use of non-prescribed dietary products. Others did not discuss their use, either because they did not feel it was important, because they perceived that the doctor would respond negatively or simply because they were not asked.

The following concept map (see Figure 7, p. 85) captures the key role and profile of parents and caregivers in the early dietary SM of children with chronic disease, and the colours indicate their relationship to the other themes. Parents and caregivers roles are affected by their varying perceptions, motivations and unique individual and family needs, each of which needs to be considered when planning a dietary self-management strategy.

Figure 7. Key role and profile of parents and caregivers in the early dietary self-management of children with chronic diseases and the relationship with other themes.



3.11.6 Theme 3: Early dietary intervention in the management of chronic disease

In each of the studies reviewed dietary intervention is recognised by parents and/or HCPs as an important aspect of chronic disease management. In the US survey (Ball et al. 2005) on the use of dietary supplements in chronically ill children, caregivers gave three main reasons for dietary supplements: improving health, supplementing diet, and preventing disease. Dietary supplementation can take many forms, with 198 different preparations reported in the US survey across a range of chronic diseases including cancer, CF, neurobehavioural disorders, renal and liver disease (Ball et al.

2005). Dietary products included vitamins, minerals, botanicals, amino acids, and dietary supplements such as n-3 fatty acids, pre/probiotics, concentrates, metabolites, extracts or a combination of these preparations (Ball et al. 2005). The most commonly used products were disease-specific vitamin or herbal preparations (Ball et al. 2005). Children with cancer were reported to use the most non-prescribed dietary products (Ball et al. 2001).

Parental belief that diet can play an important role in management of chronic diseases was demonstrated in the US study (Ball et al. 2005), with parents reporting a greater use of dietary products since their child had been diagnosed with a chronic illness. Mothers in the Indian study (Totadri et al. 2017) had made a significant change in their child's diet since commencing treatment for cancer. Furthermore, if the child's past diet was perceived by parents to have contributed to the development of the chronic disease, parents were motivated to change (Totadri et al. 2017). Foods identified as possible cancer triggers included large amounts of milk products and junk foods (Totadri et al. 2017). Other foods that were avoided during active cancer therapy included all forms of meat (although the population were mainly meat eaters), fish, raw foods and street food.

In addition, where parents themselves took certain foods or dietary products, they were more likely to have given or be giving this to their child (Ball et al. 2005). This strengthens the benefit of providing a dietary strategy that the whole family can follow rather than identifying the child with a chronic disease illness as having different needs. Since diet is a practical way for carers to contribute to the management of chronic diseases, it is important that education and information is up to date, and advice made practically relevant to their home/life environment (Coleman et al. 2010), discouraging the purchase of large quantities of special dietary products.

Ergocalciferol dietary treatment achieved a normal 25-hydroxyvitamin D level in 80% (n=16) of children with CKD and 100% (n=8) of those with early stage CKD (Shroff et al. 2012), demonstrating increased efficacy in early stage CKD patients. By nature of the population (median age 5.7 months [range 1.6-9 months of age]), infants with atopic dermatitis randomised to the intervention group in the German study

(Niggemann et al. 2001) received an amino-acid dietary intervention early in their treatment. In response to a change in their diet, all infants experienced an improvement in their eczema related symptoms (Niggemann et al. 2001). Early dietary intervention significantly improved growth results ($p<0.04$), a known complication of chronic eczema disease progression (Niggemann et al. 2001). Although anthropometric measurements were not recorded in the Australian CF intervention (Stapleton et al. 2001), increased knowledge of nutrition and enzyme control has the potential to have a positive effect on weight gain, which in turn can prevent decline in pulmonary function and disease progression (McDonald et al. 2013; Levy et al. 1985). As Stapleton et al. reported, the 'Go and Grow with CF' program could be used as a strategy for disease prevention.

The following concept map (see Figure 8) captures the many factors, such as individual motivations and perceptions, culture, cost and choice, all of which influence early dietary SM; and the colours indicate their relationship to the other themes.

Figure 8. Factors influencing early dietary self-management and their relationship with other themes



Some of the key findings of early dietary intervention reported in the quantitative studies are detailed in Table 27.

Table 27. Quantitative findings regarding early dietary intervention

Author	Change in dietary intervention	Test result
Shroff et al. (2012)	Time to develop hyperparathyroidism was significantly longer in children receiving ergocalciferol treatment compared with placebo	Ergocalciferol treated group – no significant rise in PTH levels between baseline and final follow-up (median= -8.0%, range=+74 to -88%, *p*=0.30) Placebo treated group – a significant rise in PTH was noted (median= +22.4%, range=+267 to -13%, *p*=0.046) (Hazard ratio, 0.30; 95% CI, 0.09-0.93)
Stapleton et al. (2001)	Mean difference in children's knowledge and self-management skills score between pre-intervention and immediately post-intervention (short-term)	Pre-intervention group mean score 23.3±1.1 Post-intervention mean score change 4.2±0.6 (*p*=0.001) Control group mean score 23.7 ±1.1 Control group mean score change 0.7±0.7
Niggemann et al. (2001)	Increase in length standard deviation (SD) scores (SDS) were statistically significant in children receiving the amino-acid based formula	Amino acid based (AA) formula group length SDS at the start of the study =median - 0.5 (range 1.0 to -4.1) AA group at study end (6-months) SDS= median 0 (range 2.8 to -2.3) (*p*=<0.04) Extensively hydrolysed formula (eHF) group at start of study = median SDS -0.9 (range 0.2 to -4.4) eHF group at study end SDS =-0.9 (range 0.5 to -2.6)
Ball et al. (2005)	Amongst children with chronic disease the greatest use of dietary supplements was reported in children with cystic fibrosis	Cystic fibrosis mean number of dietary supplements used ±SDS = 2.7±2.2 (range 0-9, *p*=0.001)
Totadri et al. (2017)	Mothers believing certain "special" foods needed to be added in the diet during cancer therapy, as they perceived them to increase white blood cell count	Questionnaire administered to n=72 mothers Mothers believing that certain "special" foods needs to be adjusted in the diet n=27 (37%)

3.11.7 Theme 4: Challenges and enablers to dietary change

Almost all of the selected studies identified challenges and enablers in the provision of dietary SM to children with a chronic disease. For clarity, these factors will be discussed separately in this theme.

Challenges

Callenges included the limited access or an inability to attend education classes/workshops, extended family's resistance to change (Coleman et al. 2010); disease burden, blood testing, child's refusal (Shroff et al. 2012); cost, frequent monitoring (Niggemann et al. 2001); loose stools, nausea and vomiting (Ball et al. 2005); and commercially available protein powders and other products claiming special therapeutic benefits (Totadri et al. 2017).

Hispanic women in the Californian study (Coleman et al. 2010) described many life circumstances that hindered their ability to attend all the classes, such as personal illness, and spousal demands to prepare meals during class times. Additionally, lack of transport, work commitments and the need to prepare children for the next school day were reasons given for poor class attendance. Although many of the women were unable to attend classes, they reported a change in their families' eating habits and exercise patterns as a result of talking with women who had attended. The most difficult barrier to change concerned their husbands' and extended families' lifestyles and eating habits, in particular portion sizes. Despite grandparents having health problems of their own, women described how difficult it was to change their habits. However, grandparents were often more receptive and willing to listen to their grandchildren who had attended the classes.

In the UK study of CKD patients, Shroff described the burden of care and disease (Shroff et al. 2012). Although parents are willing to adhere to the prescribed dietary treatment, there is a 'burden' of care in carrying out the treatment every day, in addition to the stress, uncertainty and many other responsibilities and pressures associated with the management of a child with a chronic disease. Despite meeting the criteria for inclusion, some children refused to take ergocalciferol, this refusal to

treat may be attributed to its colour, odour, taste, dose, blood-testing frequency to evaluate its efficacy or a combination of these, which are all a burden of care. Shroff also described the increasing awareness of evidence to suggest that a deficiency of 25-hydroxyvitamin D [25(OH) D] may carry its own disease burden that requires further research.

Whether the healthcare provider or caregiver pays for the products, amino acid specialised formulas for infants are more expensive than standard infant formulas (Taylor et al. 2012). The authors of the German study (Niggemann et al. 2001) recognised the importance of providing a product that is palatable and accepted by the infants, stating that many specialised formulae have a strong and bitter taste that can be hard to disguise. Amino-acid preparations are costly and the test product cost 25% more than the control product. The cost of non-prescribable supplements was noted in the Utah survey (Ball et al. 2005). To assess the effectiveness of the special amino acid formula in the German intervention, the parents had to attend the hospital for frequent review appointments. This could have been a barrier for families living a distance from the centre.

Parents surveyed in the Utah study (Ball et al. 2005) reported a number of challenges or side effects to the use of certain dietary products, including loose stools, nausea and vomiting. The limited information available on safety and efficacy in relation to drug interaction, growth and development was a concern, especially in view of the large number of non-prescribable dietary products on the market.

As mentioned earlier in the theme on the role of education in early dietary management, commercially available protein powders which are believed to offer special therapeutic benefits, can be misleading and very expensive (Totadri et al. 2017). Access to credible information through education and support from HCPs, or signposting to written or online evidence-based advice is important in eliminating misconceptions and promote appropriate advice.

Enablers

Enablers that support the provision of dietary SM for children, included the use of reminder techniques, strategies that incorporate incentives and a reward system that recognises achievements. Additionally, a venue that provides optimal access to the

target audience, the use of everyday activities/equipment and using the spoken language of the majority (Coleman et al. 2010). Also, limited imposition on foreign travel (Shroff et al. 2012); flexible home-based program and the use of age-appropriate materials (Stapleton et al. 2001).

The delivery of the diabetes prevention program (Coleman et al. 2010) at an elementary school provided a venue accessible and familiar to participants. The program was advertised through mail shots sent home with eligible children, meeting with staff, returning to school and attending school events and table top displays in the playground to catch parents during school drop-offs. As all the recruited families were Spanish speaking, the program was delivered in Spanish through an interactive bilingual-bicultural presentation. Parents received a telephone call to encourage attendance. The classes began with an all-age family exercise session for 30-minutes. Age-appropriate education workshops were provided for 0 to 5 years, 6 to 13 years, and parents. The class finished with all the families coming back together for 30-minute recap and planning session. During the exercise class, everyday household utensils were used, such as pots and pans, boxes, sheets and balls. This sought to make it inexpensive and transferable to their everyday lives. The two all-age group sessions encouraged social interaction between families, to share experiences, build confidence and accountability.

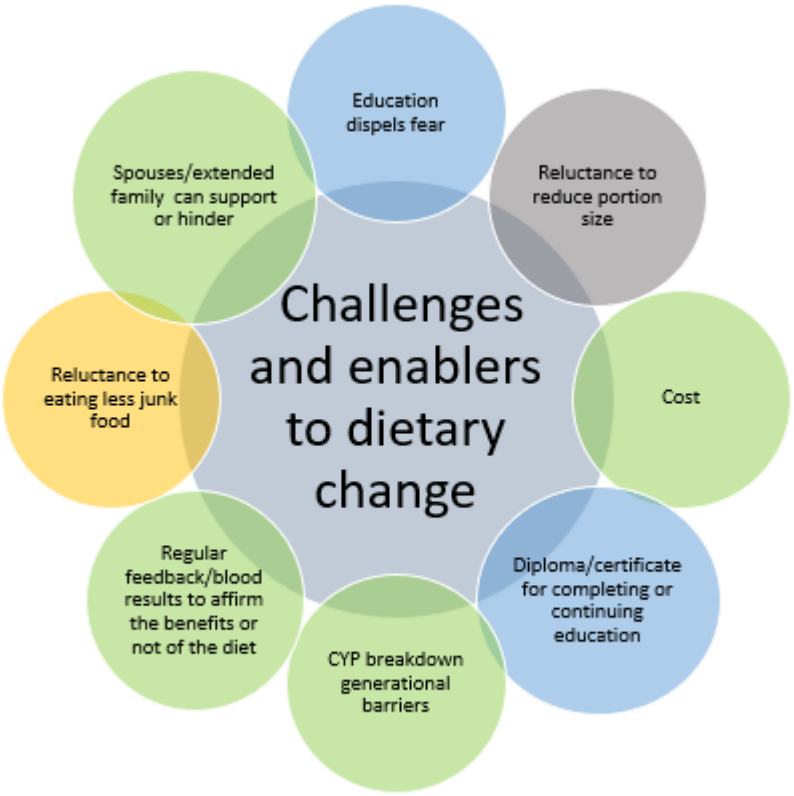
Healthy snacks such as fruit, cheese sticks and yogurt were provided as an incentive to attend the sessions. Lunch bags, hats, hand lotion, sunscreen, and toys were also rewards for attendance and engagement. Parents were invited to participate in two cookery workshops to demonstrate how affordable, healthy food is prepared in 10-20 minutes. Recipes were culturally relevant, and were provided to parents on request. Parents were taught the benefits of healthy food, budgeting and shopping during the workshop. To recognise their completion of the classes, family members each received diploma certificates.

The safe, stable and well tolerated nature of the 25-hydroxyvitamin D preparation was a factor that promoted concordance with dietary treatment in the ergocalciferol trial (Shroff et al. 2012). In fact, during the trial period, eight children travelled abroad, which did not interfere with the study compliance.

As already mentioned in the parent/caregiver theme, location was a key facilitator in the CF trial (Stapleton et al. 2001). The educational intervention involved a home-based course between an introductory and final group workshop with other families at the hospital. This offered the opportunity to adapt the program to the unique needs of each family, while promoting social and peer interaction during the hospital workshops. The use of age-appropriate materials on a rotational basis sought to keep the course fresh and engaging for everyone.

The following concept map (see Figure 9) captures some of the many challenges and enablers to dietary change, and their relationship to the other themes. Apathy or reluctance, cost, and established habits can all act as a barrier to dietary change. Conversely, education, reward schemes, inter-generational influences, regular feedback and blood results can be powerful motivators.

Figure 9. Challenges and enablers to dietary change and their relationship with other themes



Bringing together the four themes, the following section presents a discussion of the thematic analysis, data interaction and integration of the findings.

3.12 Discussion of the thematic analysis, data interaction and integration

This knowledge synthesis of children's, parents' and other stakeholders' perspectives on early dietary SM to delay the progression of chronic diseases in children provides a coherent evidence base for understanding the importance of education as a baseline tool to improve knowledge. It also highlights a need to provide ongoing education and support throughout the child and families' chronic disease journey. The parents' and caregivers' perceptions towards dietary intervention, their beliefs about food, and desire to do what they can to make a practical difference to their child's health and disease course was recognised. The important role that dietary intervention can play in delaying the progression of chronic disease, and the many challenges and enablers to implementing and supporting early dietary SM were highlighted.

This narrative synthesis served as a tool for drawing lessons from empirical research together to guide service development and future research in practical ways. For example parents and children value practical education/cookery workshops, and healthy snacks serve as useful incentives to encouragement engagement. This narrative synthesis identified key messages (see Box 3, p. 94) that informed the qualitative interview guide with CYP, parents and HCPs in Phase 2 of this doctoral study.

The heterogeneity of the methodological approaches of the studies reviewed – mixed-methods, quantitative and qualitative – and the synthesis of the narrative meant that neither a meta-synthesis nor a meta-data analysis was possible. Despite this wider search of the literature, this mixed studies systematic review with narrative synthesis clearly confirmed the research gap that no study was found on stakeholders' perspectives towards the development of an EPSM strategy. However, it has highlighted that the use of dietary intervention in the early management of chronic disease should focus not only on the real and perceived factors that influence involvement and implementation, but also on contextual factors.

Box 3 details some of the key messages that were carried forward to inform the development of the interview guide for children, parents and HCPs in Phase 2 of this study.

Box 3. Key messages informing the interview guide development in Phase 2

- To encourage better long-term retention of knowledge, HCPs with expertise in nutrition need to provide regular evidence-based education and support for the child with a chronic disease and their family.
- Programs should target the main leaders of diet and lifestyle change in a family, encouraging buy-in from the whole family (including extended family and grandparents), and tailored to their evolving and changing needs.
- Dietary interventions should be considered as part of the early management of chronic disease and not just an adjunct later on.
- More communication should be encouraged between HCPs and families to ensure they have access to up to date evidence-based information.
- Dietary intervention needs to include practical hands-on workshops that use every day, culturally relevant recipes and ingredients that are affordable and accessible.
- Opportunity should be given for families to share experiences/stories, for children's peer engagement to learn and motivate change long-term.
- Although parents and HCPs generally recognise the importance of diet in chronic disease management, families face barriers to accessing and implementing change. These barriers include transport, family members' resistance to change, domestic duties and work commitments, and the disease and dietary burden of care.

3.13 Summary

This narrative synthesis has provided a better understanding and recognition of the use of dietary intervention as a tool for SM in early chronic disease management in children. Although the methodological quality and robustness of the evidence reviewed was varied, it has facilitated a platform to learn from empirical research. It has made it possible to better understand the importance of contextual differences

between the studies and the need to adapt the dietary education strategy to the age, race, cultural and socio-economic status of the target audience. The perceptions, misunderstandings and expectations of parents, caregivers and other stakeholder groups and the importance of providing ongoing evidence-based education and support to limit inappropriate, potentially harmful practices, have been recognised. Delaying disease progression is recognised not only as a motivator for parents' early dietary intervention, but also a key driver for improving clinical outcomes. The effectiveness of any dietary intervention, however, is multifactorial, with many challenges and enablers that warrant consideration. Building upon the outcome of the scoping search, this mixed studies systematic review with a narrative synthesis further confirmed the originality of this doctoral study. Chapter 4 introduces the qualitative research method of semi-structured interviews used in this doctoral MM study during Phase 2.

Chapter 4: Phase 2 – Methods for the qualitative dimension

4.1 Introduction

Following a first phase that comprised of a systematic review with narrative synthesis, the second phase explores the views of CYP, parents and key HCPs utilising a semi-structured interview (Figure 2, p. 21). In this chapter the method is outlined. As each phase informed the next, the satisfactory completion of phase one was necessary prior to the commencement of the next phase.

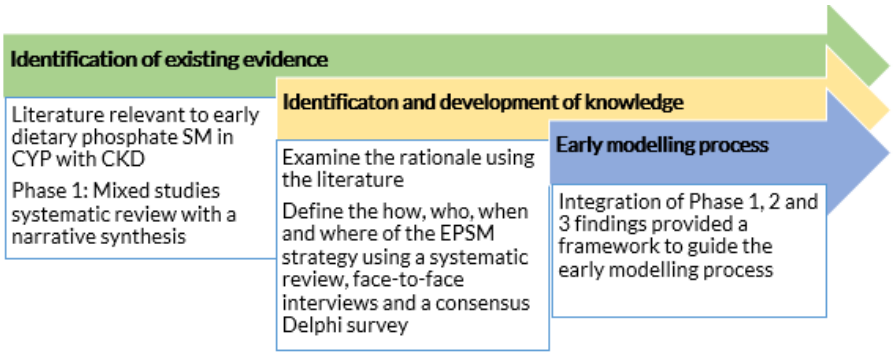
Chapter 6 begins by placing the study within the context of the Medical Research Council (MRC) Framework for intervention development. Next, the Phase 2 study population, sample size and strategy, inclusion and exclusion criteria, and the research setting are presented. The specific data collection and analysis techniques employed are discussed. Last, the ethical considerations of undertaking the research, with a focus on the involvement of CYP are presented.

4.1.1 MRC Framework - Intervention development

To appropriately address the research aim and objectives (see section 2.2), the study must be placed within the context of intervention development. An early dietary phosphate self-management strategy in CYP with early stage CKD can be considered a complex intervention as defined by the Medical Research Council (MRC). As with any complex intervention, multiple interacting components (Craig et al. 2008) are at work, influencing the outcome of the intervention. In this study where early dietary phosphate education would be the key 'active ingredient' (Hoffman et al. 2014) seeking to motivate and empower individuals to change and maintain diet and lifestyle factors, a significant active component would be the unforeseen dynamic nature of human relationships. Additionally, service structure, delivery, support and resource provision would all serve as components that either facilitate or hinder the implementation and effectiveness of the intervention.

According to the MRC's Complex Intervention framework (2008), this study is situated in the "Intervention Development" stage. The MRC Framework outlines three intervention development steps (Figure 10). Elements of the EPSM study which address each of these steps are detailed (Figure 10).

Figure 10. MRC Framework intervention development



As discussed in the literature review section (3.1.3), the excessive intake of inorganic phosphate by CYP with early stage CKD is identified as the problem, influencing CKD progression. With dietary education and support recognised as tools to support the early management of chronic disease, developing an early dietary phosphate education strategy to educate CYP and their parents is proposed as a way of addressing this problem. This MMR study sought to undertake this vital early exploratory intervention development phase, preparing the ground for future feasibility testing of an EPSM strategy (see section 9.8.3). Hoddinott (2015) recognised that the rationale and decision-making processes undertaken during the early phase of intervention development are seldom reported. Craig and colleagues highlighted the necessity for researchers to,

> *"…be prepared to explain to decision-makers the need for adequate development work…to challenge decision makers…"*
>
> *(Craig et al. 2008)*

In response, this study established the evidence base, and rigorously explored through direct face-to-face interviews with CYP, parents and HCPs, their perspectives towards the development of an EPSM strategy. This study explores whether the idea of an EPSM is warranted, and if so, what needs to be in place to support its delivery

and effectiveness. By taking a multi-stakeholder perspective, it was possible to gain understanding of what the intervention could and should look like from these different viewpoints. By considering the 'what, why, how, who and when' (Hoddinott, 2015) of the intervention, individuals contextually explored the idea of an EPSM, which informed the rationale and decision-making process. Then, through the Delphi survey, consensus was established on the issues that the expert panel felt were most important when planning the development of an EPSM. As a result, essential groundwork has started to take shape, with the emergence of some early modelling for the EPSM strategy (see response to objectives section 8.2) and directions for future research (see section 9.8.3)

4.2 Study population, sample size and sampling strategy

4.2.1 Study population

The population, or 'universe of units' from which the sample is selected reflects the research question with its aims and objectives (Bryman, 2016). In this study, the research aim: 'the exploration of the perspective of children and young people, parents and key health care professionals, towards the development of an early dietary phosphate SM strategy,' clearly identified the study population with three participant groups. Current or recent direct involvement in the CKD paediatric service rendered this sample ideally suited for the exploration of perspectives by the researcher.

In order to capture a diversity of perspectives within a purposive sampling framework (see section 4.2.2), the participants varied in age, gender and occupational status. Participants were male and female CYP and adults. CYP included had varying stages of CKD. Some were in the early stages (CKD 1-3); others had more advanced CKD, including those who were receiving dialysis or had been transplanted. This sought to provide insights from those who are/have journeyed through the early stages of CKD. During the interviews the participants often reflect across their CKD journey, for this reason, the unique identifier (see section 5.1.1) does not include the CYP's current CKD level. However, as appropriate, the quotation often makes explicit the CKD level. The second group included 'expert parents' (Kirk and Glendinning, 2002) with

recognised CKD management expertise, who had a child (0-16 years) with CKD (any stage). The third group included HCPs with expertise in the management of CYP with CKD 1-5. These individuals included paediatric renal consultants, specialist nephrology nurses and specialist paediatric renal dietitians.

4.2.2 Sample size and sampling strategy

Phase 2 of the study aimed to recruit a total of 10-15 CYP, parents and HCPs for in-depth qualitative interviews, with a balanced sample of at least 5 participants from each category (see Figure 11, p. 102). A balance of gender and geographical locations were taken into account. Thorne (2008) contests the defining of a specific sample size to achieve data saturation, rather participants should be recruited until the depth and richness of the data is sufficiently developed to warrant reporting on the subject and answer the research question.

As demonstrated in the qualitative studies by Wennman-Larsen (2002) and Ralston et al. (2004), 11 and 9 participants provided rich data on caregivers' expectations of end of life care, and diabetic patient's experience in a web-based disease management programme, respectively.

A purposeful sample of CYP, parents and HCPs was reasoned to be acceptable in qualitative interviews using ID, where the aim was to provide rich, in-depth information from individuals by virtue of their experience or specialist understanding of the topic rather than scope the breadth of a participant's knowledge (Snape and Spencer, 2003; Thorne, 2008). In practice the recruitment of HCPs took on a chain referral or snowball sampling approach (Biernacki and Waldorf, 1981), whereby a previously purposefully selected participant was asked to mention the study to their renal colleagues, these in turn discussed the research with other individuals.

4.2.3 Inclusion criteria

Children and young people had to meet the following criteria to be recruited to the study:

- Aged 10-16 years. At this age, children can effectively communicate personal views, and have the skills to self-evaluate and reflect on their experience (Owen et al. 2004).
- Diagnosed with any stage of CKD more than 6 months before recruitment. This accounted for the adjustment required when coming to terms with a chronic diagnosis.
- Must attend the tertiary hospital for treatment.
- Parental/legal guardian consent for 10-15 year olds to take part in the study (though not mandatory, for 16 years olds additional parental consent was received).

Although the study was exploring the development of an EPSM strategy for CYP with CKD 1-3, the perspective of CYP with all stages of CKD was sought. Children and young people with early stage CKD (1-3) had a view from the perspective of not currently receiving dietary phosphate support. Children and young people with more advanced CKD (4-5) provided a retrospective view on what they know now, with suggestions for change.

Parents had to meet the following criteria for recruitment to the study:

- An adult was considered from 18 years and above
- Should have recognised expertise to manage CKD as judged by the consultant paediatric nephrologist in the supervisory team, and/or an EMEESY clinical staff member involved in their child's care.
- Have a child (0-16 years) diagnosed with any stage of CKD more than 6 months prior to recruitment.
- Their child must attend the tertiary hospital for treatment.
- A parent could be recruited without their CYP being recruited.

HCPs had to meet the following criteria to be recruited to the study:

- Must work or previously worked within the EMEESY children's kidney network.
- Have recognised expertise (as recognised by their role) in the management of CYP with any stage of CKD 1-5.

4.2.4 Exclusion criteria

- All participants who did not meet the above criteria were excluded.
- All participants who were not fluent in the English language. This reflected a lack of funding for interpreters
- If the participant could not be interviewed at the tertiary centre and the geographical distance to their home was too far for the researcher to travel within the working day, they could not be included in the study.

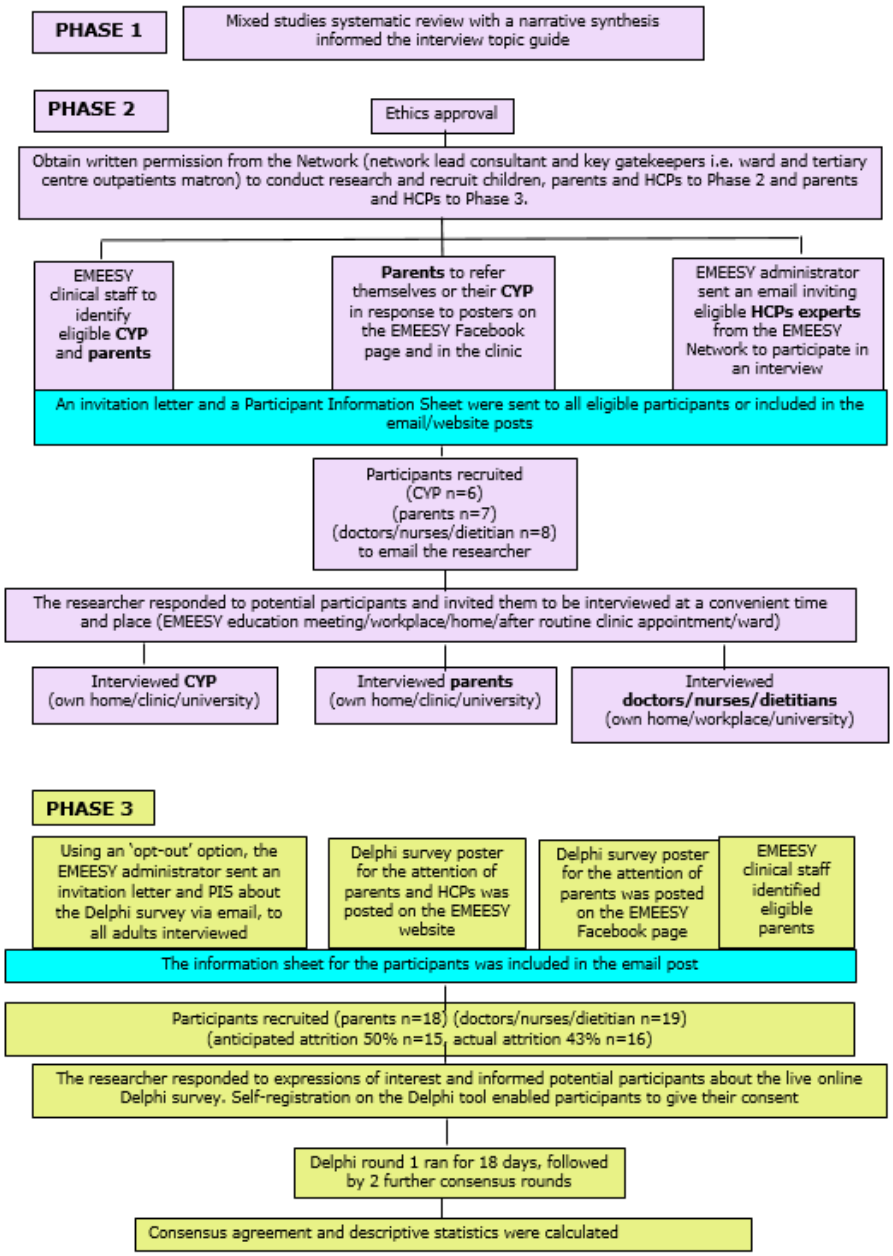
4.2.5 Research sites

As described in section 1.4.2, the children's kidney network for the East Midlands, East of England, and South Yorkshire (EMEESY) operating from Nottingham Children's Hospital at the Queen's Medical Centre, Nottingham, UK, was used to recruit for this MMR study. The EMEESY network comprises shared-care clinics in 20 local hospitals across the region.

4.2.6 Access and recruitment

Recruitment and data collection commenced once ethical approval had been granted by Health Research Authority (HRA)/Research and Innovation (R & I) (see Appendix 16) (see section 4.7). Figure 11, p. 102 details the research process, including gaining access and recruiting participants from the EMEESY network to the study. Permission was sought from key gatekeepers before the study began (Greig et al. 2017), namely the EMEESY network lead paediatric nephrologist, children's renal ward and out-patient matron. Suitable eligible participants were recommended by EMEESY clinical staff or self-referral in response to recruitment email/website/clinic posts. The number of participants recruited for phase two and three, and location of interviews are detailed in Figure 11.

Figure 11. Research process, access and recruitment



Due to the busy schedule of family's and HCPs, and to optimise recruitment, interviews were conducted in no particular order.

Recruitment posters

Posters advertising the study were placed in the children's renal ward and out-patient clinic (see Appendix 6). An advertisement was placed on the patient and parent EMEESY Facebook page and on the EMEESY network website for EMEESY HCPs. These

platforms were used, as prospective participant groups had easy or frequent access, offering optimum reach and consideration (Grove et al. 2013).

During a lunchtime paediatric renal meeting, attended by the tertiary multi-professional team members, the researcher presented an outline of this doctoral MMR study, highlighting the inclusion and exclusion criteria, with encouragement to identify eligible CYP, parents and HCP (Whitebird et al. 2010). Clinicians identified eligible participants to protect personal identifiable data and keep within the Trust. Also, clinicians were instrumental in judging CYP and parents experience and specialist CKD understanding, a necessary part of purposive sampling (see section 4.2.2).

The EMEESY administrator sent out an email invitation and participant information sheet (PIS) to all prospective EMEESY delegates to coincide with a forthcoming EMEESY education meeting. The lead consultant for the EMEESY network signed the invitation letter (see Appendix 7).

Children, young people, and parents

During routine tertiary and EMEESY shared-care renal clinics, clinicians identified eligible parents and CYP, explained the purpose of the study and invited their participation (Grove et al. 2013). An EPSM study pack was offered, containing an invitation letter and PIS sheets for parents and an age-appropriate version for CYP (see Appendix 8). This sought to maximise the CYP understanding of what is involved in the study (HRA, 2014). Individuals were given the opportunity to ask questions of the clinician, or call the researcher using the contact details in the pack (HRA, 2014). During routine tertiary renal clinics, the clinician had the option to contact the researcher by mobile phone, requesting she join the consultation to answer any questions about the study. Parents and CYP who self-referred using the contact details on the online/clinic posters were sent an invitation letter and PIS (email or post), with opportunity to ask questions as required.

Children and young people were encouraged to take the information home to read and talk to their parents about the study. This involvement of the family in the

decision-making process is encouraged by the HRA as good practice (2014). If the CYP had questions that the parent could not answer, parents were encouraged to contact the researcher.

The researcher sought permission to contact the family once they had been given 'reasonable time' to consider participating, to arrange a convenient time and place for the interview. There are no fixed guidelines for an appropriate amount of time to give potential participants to consider what involvement means, before making a decision to participate (HRA, 2014).

Although the researcher facilitated the recruitment process with the support of the EMEESY clinicians and administrator, the researcher had no contact with the CYP or the parent until an EMEESY clinician had made initial contact or the parent made direct contact with the researcher.

Healthcare Professionals

HCPs responding to the EMEESY administrator's email or EMEESY website poster (see Appendix 6) were encouraged to contact the researcher directly. Further letters of invitation and PIS were sent to HCPs, as requested, and any questions were answered before a date and time was agreed for the interview. As the researcher worked part-time at the tertiary centre, in response to recruitment posts, her HCP colleagues in the EMEESY network made direct contact, expressing interest in being recruited for the study. Section 9.6 discusses the insider/outsider considerations when recruiting colleagues for research (Kersen, 2016). After receiving an emailed letter of invitation and PIS, and asking any questions, a convenient date and time was set for the interviews. As colleagues shared their interview experience with others, a snowball effect occurred as word of mouth led to other HCPs requesting interviews in short succession (see section 4.2.2).

The informed consent and assent (refer to section 4.7.2) process was discussed with each participant, and informed consent was given before each interview began (HRA, 2014) and regularly reviewed during the Phase 2 interviews, aided by coloured cards (see section 4.7.3).

4.3. Research setting

4.3.1 Interview location and context

Participants had the opportunity to select a venue and time of their choice for the interview, all of which needed to be convenient for the researcher (Grove et al. 2013). This created a balanced dynamic of power from the beginning, encouraging the participant to contribute to a favourable setting (Grove et al. 2013). To minimise inconvenience to CYP and parents, interviews were offered at a time and venue that coincided with any scheduled clinic visits, this included booking a private room at the university or on the children's renal ward (Grove et al. 2013). Where this was not possible due to the timing of the data collection period, in accordance with the School of Health Science's lone working policy, an interview was offered in the family home. Additionally, this offered convenience for participants with time-related childcare or school run commitments, and the familiarity, contributed to a balance of power, supporting honest contribution (Shaw et al. 2011). A safeguarding plan was put in place to protect the CYP and parent, HCPs and researcher during home visits (refer to point 5, Safeguarding, section 4.7.3) (Shaw et al. 2011). As CYP living in the East of England can travel up to 160 miles, taking 3.5 hours to get to the tertiary centre for routine appointments, geographical distance influenced participation in Phase 2 of the study (refer to section 4.2.4).

To ensure the safety of the participants and researcher, all interviews with children <16-years were conducted with a parent close at hand (Manning et al. 2014, ESOMAR, 2009). Once the CYP felt at ease, a rapport had been established and the parent had developed trust in the researcher, the parent was given the option to allow the CYP to talk to the researcher alone while remaining in the vicinity (Tekola et al. 2009). To elevate the agency of the CYP (Bandura, 2001) sufficient distance was encouraged to prevent parents from overhearing what was being said (Shaw et al. 2011). Crucially, interviewing CYP at home mitigated some of the power imbalance, reducing feelings of intimidation, which can support open and honest responses (Shaw et al. 2011). Despite being private, there were some interruptions from mobile phones and a talking parrot! If the parent requested to remain for the whole

interview with their CYP, a dyad interview was offered (see section 4.5.3). Alternatively, parents were offered an interview separately.

The main factor determining the location and time of the interviews with HCPs was their busy work schedules. Additionally, the researchers pre-existing relationship with the HCP influenced the flexibility, timing and location of the interviews (McDermid et al. 2014) (refer to Reflexivity in section 9.6 for a further discussion about collegial relationships). Most HCP asked for an interview in the multi-purpose room on the renal ward. This enabled them to be visible to other colleagues if they had an urgent matter to attend. Mindful this would disclose their involvement in the study, the researcher was guided by professionals and common sense (McDermid et al. 2014). Others chose to remove themselves from the busy work environment, requesting a private interview room at the university, or in the university café. Despite the minimal privacy and noise in the café, it didn't affect the quality of the audio recording. One HCP was interviewed in his own home. The venue was very private and quiet, making it ideal for interviewing.

All interviews were conducted during working hours (9:00am-5:00pm) on a weekday, and lasted between 24:56 and 1:20:06 minutes (see Table 30, p. 128). CYP home interviews were scheduled when they returned from school. The interview with 03CYP established a rapport very quickly, lasting over an hour, and yielding some in depth data on her experience and views.

4.4 Data collection tools

4.4.1 Tools for data collection

To support the flow of interview information, a semi-structured interview guide with non-directive probes was developed (Bryman, 2016) for parents and HCP, and an age-appropriate CYP version (Koller et al. 2010). An interview guide provides a framework to the interview, ensuring that similar types of data are collected, however it is not followed strictly and the sequencing of questions is not always the same, allowing the researcher to respond to the participant's lead (Holloway and Galvin, 2017). The tool consisted of broad open-ended questions with prompts to promote

an open and detailed discussion to emerge from the participants (Bryman, 2016). The outline of the interview guides, which can be found in Appendix 9, was driven by the research aims and objectives, and the Phase 1 systematic review. Before submitting the age-appropriate interview guide to the Research Ethics Committee (REC), it was piloted with a young renal patient for content validity, suitability and appropriateness (Grove et al. 2013). Changes were made in light of any remarks. In addition, the flexibility of the ID methodology (Thorne, 2008) made it possible to modify the questions in the interview guide as new insights emerged from the systematic review and as data evolved from each interview. This allowed the researcher to be flexible and sensitive to the specific dynamics of each interaction. At times allowing the participant to 'go off-piste' gave insight into what they see as relevant and important (Bryman, 2016), or interjections gave focus and direction to the interview.

In order to promote a balance of power and a child and young person-centred approach (see section 1.5.4), an art-based approach was developed to engage with CYP (refer to 4.7.3) (Manning et al. 2013). This consisted of coloured modelling dough, felt tip pens, a scrap book, and a sheet with boxes to makes lists and notes.

4.5 Data collection

4.5.1 Data collection process

Qualitative data from semi-structured interviews were collected for a period of 5 months, from February 2018 to June 2018. This included preliminary data analysis, which was undertaken concurrently with data collection (Grove et al. 2013).

4.5.2 Data collection method

Silverman (2010, p. 10) states, the choice of data collection methods should not be 'predetermined'; rather, they should be strongly influenced by the nature of the inquiry, the central objective, and the specific question (Bryman, 2016). As discussed previously, the dominant focus of this MMR (see section 2.4.2) determined by the research question, aim and objectives (see section 2.2) was the inductive exploration and analysis (using interpretive description) of stakeholders' perspectives (Thorne, 2008).

Using a mixed-methods approach, it was necessary to identify a method that would allow the main caregivers (HCPS) and service users (parents and HCPs) to drive the agenda and decide on the issues they considered important when developing an EPSM strategy. The in-depth qualitative semi-structured interview was identified as a valuable and powerful tool (Silverman, 2013), giving the researcher privileged access (Silverman, 2013) to the respondent's perspective on the development of an EPSM strategy. For this reason, the semi-structured interview was chosen as a data collection tool for this doctoral study.

Why separate focus groups were not appropriate

Separate heterogeneous focus groups for CYP, parents and HCPs were considered as the main data collection tool in this study to elicit a variety of views in relation to the development of an EPSM strategy (Bryman, 2016; Acocella, 2012). However, for the following reasons, this method was thought not to be suitable for this population. Owing to the plurality of perspectives, focus groups can push participants to express more socially desirable responses, rather than the depth and richness of individual views that were sought in this study (Acocella, 2012). Focus groups are more suited to groups with a similar professional background, or social status (Grove et al. 2013), and not the varied professions, ages and social strata represented in this doctoral study. In order to encourage active participation, focus groups ideally require 4-10 participants, who can be available at an agreed time and venue (Wiles, 2014). Due to the geographical landscape of the managed paediatric renal network, it would have been difficult to marry the diaries of the participants with the minimum inconvenience. Also, as CYP, parents and HCPs were asked to share their views on dietary phosphate management in CKD stage 1-3 and future disease progression, sensitive issues were raised. Focus groups pose an ethical dilemma in this respect, since they offer no opportunity for anonymity or confidentiality (Ross, 2012).

For clarity, the research method used in Phase 2 (semi-structured interviews) and Phase 3 (Delphi survey) (see graphic representation in Figure 2, p. 21) are discussed separately, here in chapter 4, and 6, respectively.

4.5.3 Semi-structured interviews

Unlike structured interviews, which are often organised with narrow questions in a specific order, with little scope to follow-up questions, the semi-structured interview remains open to how the participant responds, enabling bespoke follow-up questions and allowing the participant's *emic* perspective to emerge (Grove et al. 2013). The open or unstructured interview, by contrast, provides the participant free rein to explore a topic in whichever direction they choose (Bryman, 2016); not a helpful approach when seeking to explore a number of issues related to the development of an EPSM strategy.

Semi-structured interviews are useful for exploring individuals' perspectives on a topic, particularly when the topic of interest is personal and sensitive, and when it is unlikely that participants would speak openly about the subject in a group setting (Holloway and Galvin, 2017). CYP, parents and HCPs are very familiar with the technique of interviewing, as it can be viewed as similar to the consultation experience they engage at each clinic review (Thorne, 2008). However, in the therapeutic interview the clinician likely dominates the conversation, expressing more empathy as they support the patient needs. By contrast, the researcher would be conscious of their position and how their participation affects the participant's reaction, seeking neutrality and not dominating the interview with their voice (Bryman, 2016).

The many advantages to the one-to-one semi-structured interview, over the structured or unstructured interview, include the opportunity for the participant to respond spontaneously and honestly to the researcher's questions, with time to reflect and slowly articulate their views and ideas (Holloway and Galvin, 2017). The researcher can immediately follow up an exchange to clarify the meaning of words or phrases, all the time establishing a rapport and building trust (McDermid et al. 2014). Semi-structured interviews can facilitate an open discussion that may involve ethically sensitive issues (Grove et al. 2013), such as the progression of CKD in this study. These discussions facilitated the collection of thick data (Polit and Beck, 2010) to inform the

research question, and the development of statements for the Phase 3 online Delphi survey.

Dyad interviews

Dyad semi-structured interviews were undertaken with some CYP and their parents. This approach recognises the interdependent nature of human agency (Bandura, 2001) and allow the child or young person to remain with their parent, embracing this phenomenon as a rich source of data rather than trying to control for it (Caldwell, 2014). This interview structure allows two participants to interact in response to researchers' questions (Morgan et al. 2013). One participant's comments can stimulate ideas, prompting or drawing out answers from another that might not have been recognised or remembered (Morgan et al. 2013). This approach provided a forum for the parent and CYP to learn from each other's perspective (Ungar et al. 2006), while recognising the protective role of the parent and their desire to be present.

However, mindful that parents can often influence a CYP's responses, and that different results can emerge when CYP are interviewed alone (Tekola et al. 2009; Brannen and O'Brien, 1995), through respectful explanation parents were encouraged to allow the CYP to be interviewed alone, while remaining in a nearby room (refer to section 4.7.3 on privacy). Giving CYP/parents the option gave them choice, control and self-determination, this in particular enabled the CYP to maintain some control over a decision that directly affected them (Caldwell, 2014). Whether they chose a one-to-one or a dyad interview, it further elevated the child and young person-centred approach (Koller et al. 2010); giving importance to the CYP views and a desire to see them as an expert in their own lives (Tekola et al. 2009), whilst generating rich data (Morgan et al. 2013).

Triadic interview

A triadic interview (Aldridge, 2010) was undertaken with one family. The approach was chosen in order to provide the young person who had experienced difficult circumstances to remain with the people who had 'travelled' with him and were able

to support him in speaking openly and frankly about sensitive issues (Morgan et al. 2013). The young person was enabled to share his views and experiences in a safe space with the support, encouragement and prompting of his parents, at a convenient time and place. Despite being private and making every effort to moderate the flow of the discussion, the family's enthusiasm and eagerness to talk meant that they often spoke over each other, making it difficult to hear what each participant was saying, adversely impacting the quality of the data collected. A recognised challenge when more than one person is interviewed (Bryman, 2016).

4.6 Data analysis using Interpretive Description

Interpretive description (Thorne, 2008), as described in section 2.5, follows an eight-stage data analysis process (see Box 4).

Box 4. The eight-stages of interpretive description (Thorne, 2008)

The eight-stage ID process involves the following:

Descriptive primary data analysis

1. Making accurate records of the data.
2. Familiarisation with the data through 'immersion'. This involves allowing your thinking to repeatedly move to-and-fro between the individual transcripts, listening to the recordings and the data set as a whole, all the time becoming more connected to the words and sounds, rather than simply the storyline.
3. Draw out 'group signifiers' with apparent thematic similarities by making marginal memos or using highlighters. These should capture the nature of the collection, without prematurely allotting meaning.
4. 'Flag and harvest' dominant quotes early, and place them in a "useful quotes" file. This will prevent them holding dominance over evolving analytic frameworks.

Interpretive secondary data analysis

5. Moving from patterns to relationships through contextual meaning is an iterative reasoning process, which involves drawing meanings between the group signifiers. Contextual meaning is draw between the group signifiers. The researcher comes to know the data at a new depth, beyond the self-evident and superficial and inductively build clarity and coherent meaning across the whole data set. This may involve deconstructing initial 'group signifiers' in favour of improved and more comprehensive understandings.
6. Documenting analytic thinking in order to iteratively refine what it all means. This may involve using a blank notebook to regularly brainstorm to capture collections of thematic ideas, questions and emerging patterns in memos to track evolving thoughts of inquiry.
7. Regrouping and connecting earlier disjointed items will allow the data to take shape as the researcher uses their skill of critical reflection and interpretation.
8. The formulation of analytic conclusions comes as the researcher documents the complex stages through which their thinking evolves, bringing them closer to a rich understanding of the eventual structure of the findings.

Since ID has many phases with countless subtle, often unconscious analytical facets (Thorne, 2008) the eight stages are not designed to be adhered to in sequence, rather to progress iteratively during the data analysis journey. For example, the researcher started to 'flag and harvest' (point 4) dominant quotations early in the analysis process as they were overshadowing the analysis process. Key stages of the ID data analysis process will now be discussed, starting with the descriptive and moving into the interpretive data analysis phase.

4.6.1 Descriptive data analysis

Making accurate records of the data (stage 1 in the descriptive primary data analysis ID process).

With permission from the participants, the interviews were audio recorded to capture the participant's own language (Shaw et al. 2011; Grove et al. 2013), then uploaded to a secure drive on the password protected university server, anonymised and transcribed verbatim (Bryman, 2016). Silverman's (2015) simplified transcription symbols supported the transcribing process. In order to build confidence in the transcription, a transcription kit was used, allowing the researcher to adjust the speed of the audio recording, with the option of repeatedly listening to a difficult section by pressing a foot pedal. As soon as the interviews were transcribed, the primary coding began. The analysis of qualitative data was an extremely time-consuming and labour-intensive task (Bryman, 2016). To systematically and rigorously prepare this large qualitative data set for analysis, a Computer-Assisted Qualitative Data Analysis Software (CAQDAS) was used, NVivo 12™ (Rodik and Primorac, 2015).

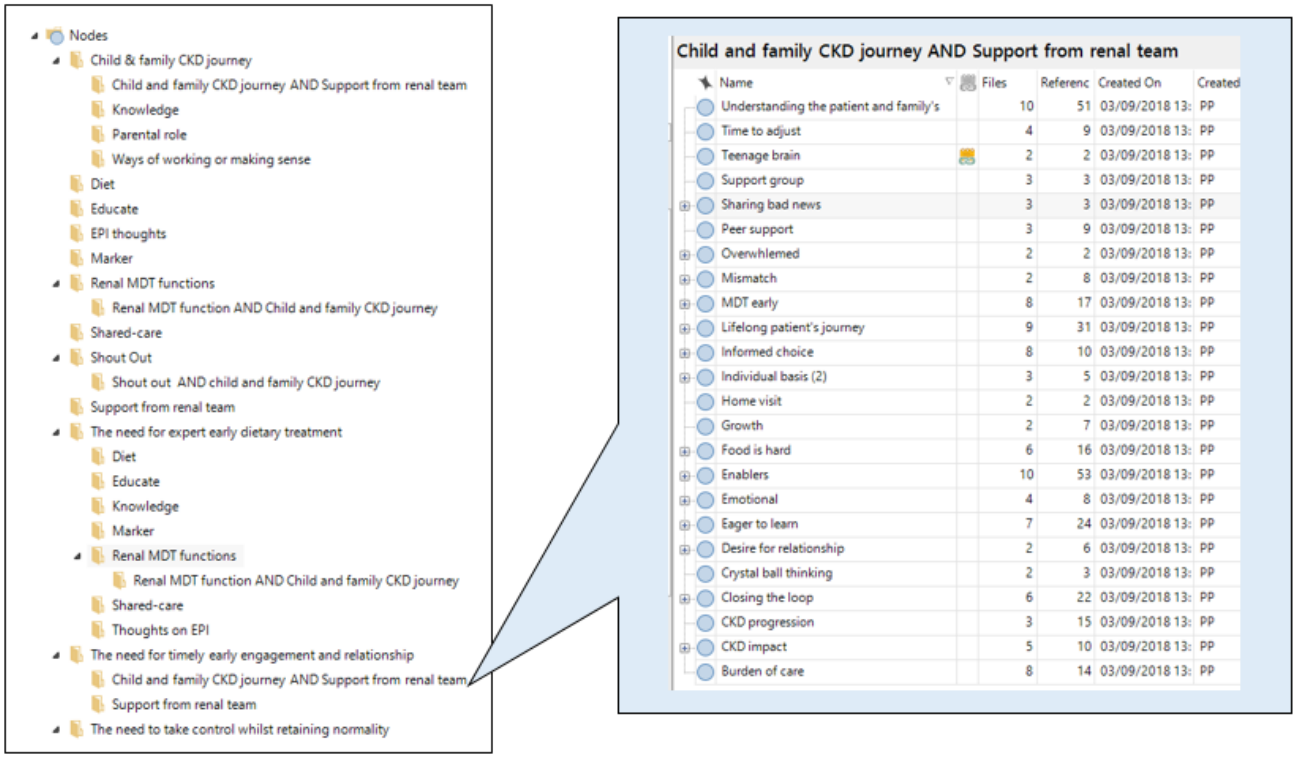
NVivo 12 is a software package produced by QSR International, an Australian based software developer. The usefulness of CAQDAS is an ongoing debate in the management of large amounts of textual data (Rodik and Primorac, 2015; Zamawe, 2015). When analysing qualitative data, the primary objective is to 'see' things from the perspective of the respondents. Rodik and Primorac (2015) argue that allowing a computer program to facilitate the organisation and coding of the words can interfere with this knowledge formation, resulting in the 'loss of shades of meaning and interpretation'. In its defence, NVivo was chosen as it proved to be an expedient data management package that gave the researcher complete control of the data, improving the accuracy and speed of the analysis process and supporting the creation of codes (referred to as nodes in NVivo 12) and the discovery of themes (Zamawe, 2015). Refer to Box 5, p. 114 for an example of nodes developed in NVivo 12 during the descriptive data analysis. Once the audio-recordings were transcribed, they were uploaded to NVivo 12. The data analysis process for this study is discussed in detail below.

Familiarisation with the data through 'immersion' (stage 2 in the descriptive primary data analysis ID process)

Immersion involved spending a lot of time reading and listening to the transcripts and audio recordings, allowing thinking to repeatedly move back and forth between each recording and the data set as a whole (Thorne, 2008; Polit and Beck, 2018). As the researcher listened to the nuances, pronunciations, expressions, explanatory gestures and inflexions in the participant's voice, notes were jotted down to capture their added meaning (Bryman, 2016). All the time the researcher was becoming more connected to the words and sounds, listening to the "deeper" message, rather than simply the storyline. During this early stage of data analysis, prominent quotations were 'flagged and harvested' (Thorne, 2008) and placed in an electronic file labelled "useful quotations" to avoid losing quotations that seem to contain some important insights and prevent them from prematurely dominating the evolving analysis.

Words, phrases and segments of data within and across the transcript began to relate. These were assigned an initial node. This early coding was conducted simultaneously using NVivo 12 and a Word document. Using a line-by-line process, the researcher was able to delve into the minutiae of each transcript and tease out all possible connections, highlighting and drawing words and phrases together with similar meanings (Bryman, 2016). Caution was taken not to be carried away by the process, reacting impulsively to similarities, and mechanically and prematurely sorting and coding the data. Instead, the researcher tried to allow her reason and inductive thinking to guide the analysis process. Initially, a broad-based coding approach with many sub-codes was used (Bryman, 2016). This enabled a collection of words and phrases that had common meaning but were contextually disparate.

Box 5. Example of nodes developed in NVivo 12 during description data analysis



For example, the broad-node "relationship and coping" was coined to cover the multiple facets of the CKD journey, where participants described or implied a desire for or recognition of a relationship with others that seemed closely linked to the action of coping with the disease. The recognition of "isolation", for example, spoke of a desire for connection and support.

When each new node was assigned, the researcher developed a working definition in the Word document (example shown in Table 28, p. 116). This helped her to constantly reflect on the nodes, and repeatedly look back over the data (Bryman, 2016).

As the researcher immersed herself in the data, the early broad-based nodes allowed her to conceptually capture the actions or processes that spoke. Thorne (2008) argues that by taking a broad-based approach, it allows a wide inclusion and collectively assigns the data a place, while distinguishing them from other types of data. By keeping a loose attachment to the nodes, Thorne (2008) argues that the angle of vision can change, and their remains a constant state of revision and fluidity (Bryman, 2016), allowing experimentation with the complexity of the data set. This helped the researcher to appreciate the many options available to handle and reconstruct patterns within the data. According to Thorne (2008), this early coding phase should be carried out cautiously, with repeated critical evaluation, in order to gradually uncover the "meaning that lies hidden within [*the*] apparent chaos." This intense period of connection with the data allowed the researcher to maintain an interactive relationship with the research participants. All the time she was able to gain a more pragmatic understanding of the actions, processes and meanings that participants gave to the development of an EPSM strategy. An example of a broad-node "relationship and coping" and its multiple sub-nodes documented in Word, each defined, is shown in Table 28.

Table 28. The initial (step 1) broad-node "relationship and coping" with its sub-nodes and definitions

Relationship/coping	
Relationship building	Opportunity to build relationship with the child/family
Desire for relationship	An expressed desire to build relationship with HCP or others in a similar situation
Isolation	Acknowledged isolation
Sharing bad news	Examples of sharing bad news
Feelings of guilt	Parents feelings of guilt towards their actions or thoughts
Self-blame	Feeling you are the cause of the problem
Not wanting to offend others	Aware that your conversation or opinions may offend others
Trust and respect	The importance of building trust and respect in order to make lifestyle changes
Lifelong patient's journey	Recognition of the long term patient journey
Understanding the patient/family	Recognition of trying to understand what they are going through and what they need
Crystal ball thinking	Wanting to know what the future will hold
Family affair	When the whole family need to be involved
Timely connection	Choosing when they want to engage and know more about the condition
Burden of care	The patient/carers take on a lot of responsibility in order to manage the CKD
Normalisation	Making the renal diet 'normal' or 'no big deal'
Desire to give back control	Parents desire to give control of the diet to the CYP
Releasing the reins	Letting go of some control
Below the standard	Feeling that what is happening is below the expected standard
Holding on	Holding on to every bit of information
Hope	Clinging onto hope

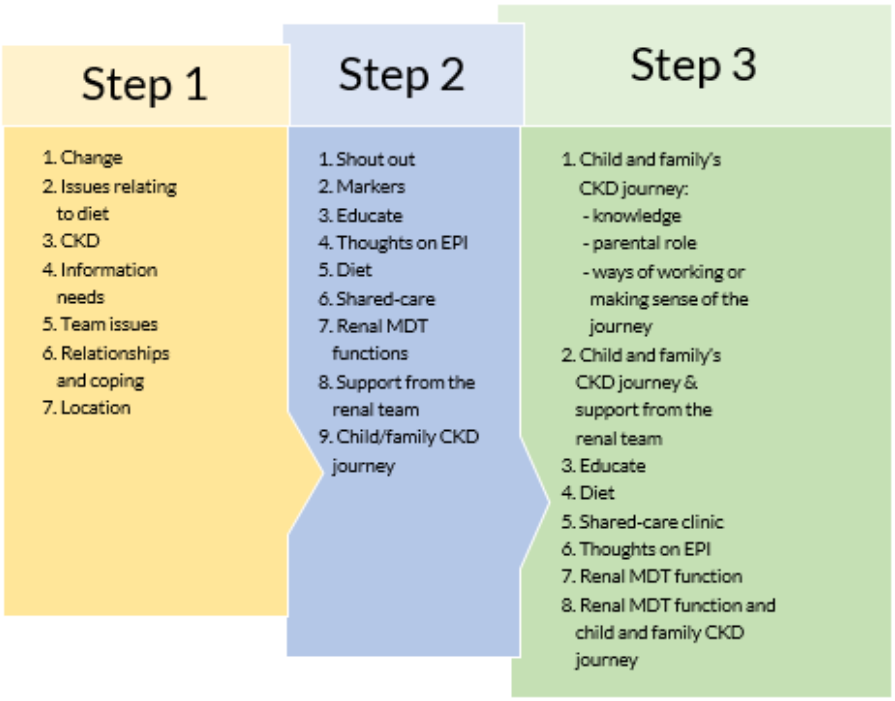
Drawing out 'group signifiers' with apparent thematic similarities (stage 3 in the descriptive primary data analysis ID process)

Each repeated immersion in the data revealed that the broad-based nodes uncovered collective groups, each with significant facets (Bryman, 2016). As Thorne (2008) described, the grouping of nodes using 'group signifiers' allows the researcher to consider these elements collectively. For example, the broad-node "relationship and coping" revealed various contact modes and phases of the chronic disease journey. At many different levels, participants expressed a desire for connection. In the face of a chronic diagnosis, CYP desired authentic relationships to build stability and trust. Contact assumed a more supportive and educational role as their journey progressed, which they requested through a more virtual social media platform. However, as the

journey brought periods of crisis, a text hotline was suggested to ask questions in a measured way, "without having to share anything you don't want."

Primary data analysis, Step 1 (see Figure 12), assigned 246 nodes and 7 group signifiers. To facilitate the departure from the broad-nodes and sub-nodes, and the transition to the formation of group signifiers, in Step 2 the 246 nodes and their definitions were printed, cut out and arranged in order on A1 sheets, allowing the researcher to tangibly explore patterns across the data. In turn, each node was reviewed with duplicates removed and similar/related nodes combined. The analysis progressed through an iterative process of reasoning, with the recognition of patterns and associated relationships (Thorne, 2008). As group signifiers developed notes were written on the A1 sheet to capture the developing impressions. These new clusters of nodes with their group signifiers were photographed before further analysis was carried out (see Appendix 10). Figure 12 shows the evolving group signifiers as the data analysis process progressed.

Figure 12. Evolving group signifiers in Step 1, 2 and 3



4.6.2 Interpretive data analysis

The methodology of ID does not advocate a set of prescriptive data analysis procedures. However, aligned to pragmatism and mixed-methods, it releases the researcher to identify and apply data analysis methods that best serve the research question (Morse and Niehaus, 2009; Thorne 2008). As informed by Thorne (2008), alternative inductive analytical approaches such as thematic analysis and conventional content analysis were considered. As content analysis applies a systematic rule-based procedure, often guided by theory (Hsieh and Shannon, 2005), it was felt not to be appropriate in this study. However, thematic analysis, valued for its highly inductive and flexible approach, was applied in the analysis of the Delphi free-text comments (Clarke and Braun, 2013; Braun and Clarke, 2006) (see section 6.8.2). Using a colour-coding system, patterns and themes were compared and contrasted across survey data sets (Polit and Beck, 2010), informing statement refinement, revision and development.

Move from patterns to relationships through contextual meaning (stage 5 in the interpretive secondary data analysis ID process)

After transitioning from the broad-nodes with their multiple sub-nodes, to group signifiers, that captured collective groups with common ground, the analysis drew on the relationships within and across the data, recognising their contextual meaning. As described by Miles and Huberman (1994), this iterative reasoning process involved inductively building parallels across the groups. In an attempt to not attach superficial links and patterns in the data, Thorne (2008) urges the researcher to step back and create distance from the intensity of this process by shifting attention from individual cases to the entire data set; from parallels in groups of cases to manifestations of difference in other cases. This involved setting aside many of the group signifiers developed so far.

When listening to each recording, the researcher made fresh notes as she allowed her thinking to move between the individual transcripts and audio recordings. Each new group was further examined in relation to the participant groups, namely, CYP, parents, and HCPs. Different but similar nuanced patterns began to emerge within

and across the participating groups as she began to view the data beyond the self-evident and superficial, at a new level (Thorne, 2008). Clarity was inductively built, and coherent meaning began to emerge throughout the whole data set. Initial 'group signifiers' were deconstructed in favour of improved and more comprehensive understanding (Thorne, 2008).

Document analytic thinking, regrouping and formulation of analytic conclusions (stage 6 in the interpretive secondary data analysis ID process)

In addition to the NVivo 12 software files, a detailed Word matrix was developed (see Appendix 11 for a section of the matrix). The researcher captured new thoughts and ideas as her analytical thinking refined and tracked each new thought of inquiry.
As suggested by Thorne (2008), in order to gain further understanding, the researcher must repeatedly ask questions about the data. Table 29 provides an example of analytic notes and questions on one of the substantive topics.

Table 29. An example of analytic notes and questions

Substantive topic	Analytic notes and questions
The need for support through timely early engagement and relationship-building	• **Healthcare professional**: HCPs recognise the importance of first building a relationship with the individual and their family, before dietary lifestyle change can be encouraged. o Will this give understanding of the family's priorities? o How much can the family handle? o How should this support be given? o How much information should be given? o Will this make them feel valued and informed, sufficient to make an informed choice? o Can this be supported by telephone reviews to encourage, and motivate? o Support groups, renal community role models and buddying networks, would these build early engagement?
	• **Parents**: Parents desire and recognise the need for early support and advice. o Is this support and advice only effective if a relationship is established first? o It takes time for the HCP to get to know the unique, evolving and changing needs of each family, and then tailor the support and advice to their needs. How long does this take? o But does this time and effort needs to be put in if advice is to be well received and followed? o Parents want to meet face–to-face with other parents. How can this be achieved?
	• **Children and young people**: CYP feel that EPI should be given early. o Should this be after a period of breathing space, allowing the family to come to terms with the chronic diagnosis? o Should education and support be face-to-face? o Do CYP really like face-to-face contact? o Do they really want to get to know the HCP? o Do they want contact on their own terms? E.g. via social media, at a time and place they choose to interact?

Data were grouped together, connecting earlier disjointed items. Using critical reflection and interpretation new relationships began to emerge from the data. After several drafts to reflect and examine how best to capture the evolving topics, fours themes were developed from the data. Each theme had multiple sub-themes, as detailed in Chapter 5 – Phase 2: results of the qualitative dimension.

During the interpretive data analysis period, diagrams were also prepared to visually capture the developing themes, showing relationships across the themes and their place within the CKD journey. These diagrams are presented before each theme in s The linkage of themes enabled the researcher to make further sense of the data, contributing to the analytical process.

4.7 Ethics application

Ethical approval was obtained from HRA by application through the Integrated Research Application System (IRAS) and Research and Innovation department at the tertiary centre (see Appendix 17). All supporting documents were submitted (see Appendix 12). Figure 13 provides a timeline of the ethics application process with detail of the events involved. A sponsor letter of approval and public liability insurance was obtained from the UoN. The researcher attended a meeting of the Research Ethics Committee with a supervisor to answer any questions about the study.

Figure 13. Ethics application timeline



4.7.1 Ethical considerations

Since data collection involved CYP and their parents sharing sensitive personal medical information, ethical considerations played an important role in the research (Greig et al. 2017). This mainly concerned ensuring the informed consent/assent and anonymity of the participants; specific NHS REC requirements directed and determined how these were best met. The NHS REC guidelines specify that informed consent/assent requires potential research participants be given time to consider what involvement means, before making a decision to participate (HRA, 2017). Potential participants were encouraged to contact the researcher with any questions or concerns about their involvement in the study. The first contact was made by the clinician and not the primary researcher. In order to comply with these guidelines, posters providing information on the study were posted on the patient and parent EMEESY Facebook page, in the children's renal out-patient clinic, and on the renal ward. This was followed by an invitation from the lead consultant for EMEESY sent by email from the EMEESY coordinator to eligible HCPs throughout the EMEESY network. Participant information sheets (PIS) were made available to all eligible participants. The next section discusses the ethical consideration of undertaking research with children.

4.7.2 Ethical consideration when conducting research with children

Health science research is fundamental to the management of chronic disease, and childhood is the most changeable period in the human life cycle (Beazley, 2009). In order to better understand the changing needs of children during chronic diseases and provide services that are better value for money, better designed and fit for purpose (Davis, 2009), it is crucial that researchers contribute to the body of knowledge about children by conducting research with children (Code of Human Research Ethics, 2014; RCPCH, 2016). Child rights-based research gives credence to their agency (James et al. 1990), and the UNCRC (article 12) (UNCRC, 2009, No. 12) states that those who can contribute their views should be given opportunity, and these views should be considered in respect to their age and maturity. Therefore, in

order to effectively explore the views of stakeholders on the development of an EPSM strategy for CYP with CKD, it was clear that this study needed to understand and include the views of the main service users, CYP.

Consent and assent

Written parental (Children Act, 1989) consent and the CYP assent had to be given before the CYP could be involved in the research. As a representative of the CYP, parents of CYP ≤ 16 years could not give their child's consent but could give consent on behalf of their CYP (Lambert and Glacken, 2011, Family Law Reform Act, 1987). Assent given by the child, who had been assessed as having the capacity (Mental Capacity Act, 2005), is the non-legally binding counterpart to consent, which exists to decrease the risk of exploitation (UNCRC article 19, 1989; Miller et al. 2004). The balancing of the child's rights with the researcher's responsibility to keep them safe, requires the researcher to consider whether the child is 'Gillick competent' or whether they meet the 'Fraser guidelines' (1986).

Time and planning, with age-appropriate resources (refer to Appendix 8, 9 and 12) helped the researcher to gauge the CYP's understanding of participation in the study (Kumpunen et al. 2012). Recognising the CYP's unique and differing competencies, giving them the opportunity to express their concerns or expectations, and responding to cues where they sought reassurance, helped to ensure they were making an informed decision. CYP were informed that all personal information would be anonymised, and data collected during the research process would be held on a password protected secure drive. The only time confidential information would be shared was if there was reason to believe a safeguarding concern had arisen that put the safety of the CYP, parent or researcher at risk (UNCRC article 3, 1989). In this situation, advice would be sought from the safeguarding representative, Dr Farida Hussain, and a formal referral would be made to the safeguarding team as appropriate.

Before giving informed consent or assent, participants were assured they could withdraw from the study without giving a reason at any time. Parents and CYP were

reassured that withdrawal from the study would not in any way affect the treatment or care of the child.

Clear and effective communication

Clear, effective communication was used to prevent misleading expectations. Using services available to support the effective design of children's literature, the information provided in this doctoral study was in a format that could be clearly understood by children (National Research Ethics Service, NRES, (2011); UNCRC article 13 and 17c, 1989). The PIS and interview combined written and colourful pictorial elements that were culturally sensitive and age appropriate, thus encouraging a more meaningful contribution from the CYP. To ensure the reading material was suited to the 10-16 year olds recruited to this doctoral study, the Flesch-Kincaid grade was used to achieve an 8-year plus readability score.

4.7.3 Interviewing children and young people

Throughout the interviews the researcher drew on her experience of over 18 years working with children with CKD and their parents. While being sensitive to the unique and differing competencies of the child and parent, the researcher faced some methodological and ethical issues when interviewing CYP: (1) unequal power relationships; (2) access; (3) privacy and (4) safeguarding.

1. **Unequal power relationship:** in order to address the asymmetry in power between the adult and CYP, a child and young person-centred approach (refer to section 1.5.4) was taken. To put the CYP at ease the researcher dressed informally and sat at the same height (Lobe et al. 2008). A CYP toolkit was prepared to empower them to take some control of the interview (Manning et al. 2013). They were encouraged to select the type of material and activity that enabled them to best express their views. Some CYP chose to doodle in the scrap book, be creative with modelling dough, or write notes as they went through the interview. One young person, delighted to play with the dough, quickly established a rapport with the researcher, and openly shared his views while skilfully sculpting. By inviting a flexible interview style, the CYP were encouraged to freely construct their responses (Manning et al. 2014).

During a child/parent dyad interview, the child engaged with the colouring pens early in the interview. Despite the researcher adapting the language and syntax to the cognitive and developmental ability of the child, she shied away from speaking, looking to her mother for instruction, preferring to colour. Green and red 'start and stop' cards enabled CYP to take control during the interview (Shaw et al. 2011); they could be used to request a break, communicate that they'd had enough and wished to stop the interview, or if they thought it would help them to ask a question. At the end of the interview, one young person interviewed with his mother enjoyed using the cards to express his boredom and desire to leave, despite his mother's eagerness to continue. The researcher sought to elevate the CYP's perspective, using language that acknowledged the importance of their distinctive CKD experience and views, recognizing their 'expert role' (INVOLVE, 2016). For example,

> *'…think back to your earliest memory of having chronic kidney disease, what can you remember?'*
>
> *'Tell me more, what do you think about that?'*
>
> *'And how do you feel the information should be delivered…?'*
> ***(INT_02CYP_M)***

2. Access: Permission to access and recruit CYP (parents and HCPs) was obtained from Health Research Authority (HRA)/R and I and the key gatekeepers (see section 4.2.6) (Greig et al. 2017). Refer to Figure 11, p. 102 for a flow diagram of the access and recruitment procedure.

3. Privacy: Negotiating interview privacy can be a delicate issue when interviewing CYP (Lambert and Glacken, 2011; Mauthner, 1997). Recognising and respecting the parent's 'protecting' role was important, as naturally they want to be present (Mauthner, 1997). However, mindful that they may often exercise control over the answers of a CYP, and that different outcomes may arise when CYP are interviewed alone (Mauthner, 1997), as outlined above (see Dyad interviews in section 4.5.3), over time and through respectful explanation parents were encouraged to allow the CYP to be interviewed alone while staying in a neighbouring room. This further elevated the importance of the CYP's views and the desire to see the CYP as an 'expert' in their own lives (Tekola et al. 2009; Brannen and O'Brien, 1995). During

two interviews with CYP in their own home, the parent stayed in the adjacent room with the door ajar. In the other CYP interview at the university, the mother asked for directions to the university café, requesting that we come to find her when the interview was over.

4. **Safeguarding:** Throughout the study the well-being of the CYP (parent, HCPs and researcher) took precedence over the research and data collection. A safeguarding plan was put in place (East Midlands Safeguarding Children Network, 2012). The researcher carried the contact details for the doctoral study and tertiary centre safeguarding representative. If, at any point during the research, safeguarding issues arose, the researcher planned to contact the appropriate representative at the early possible opportunity, discuss the matter and decide on the appropriate course of action.

4.7.4 Data management

In line with the Data Protection Act (2018), confidentiality and anonymity were maintained throughout the research process. Confidentiality involved securing all paper copies of research data in a locked cupboard on the UoN premises, and archiving for seven years. Audio recorded interview data was uploaded to a secure server and deleted as soon as possible after the interviews had been completed. All electronic personal data will be kept for 3-6 months after the end of the study to allow participants to be contacted about the study findings (unless they advise they do not want to be contacted) and then securely destroyed. Participant files were given a unique identifier, and password protected. All information collected during the course of the study has been kept strictly confidential, stored in a secure and locked office in a locked cabinet, on a password protected database.

4.8 Summary

This chapter has discussed the research method of Phase 2 of this MM study. Phase 2 utilised semi-structured interviews to access the in-depth qualitative perspective of CYP, parents and HCPs. Inductively analysing the data using interpretive description (Thorne, 2008) gave access to a pragmatic understanding of the actions, processes and meanings that participants gave to the development of an EPSM strategy. Ethical

considerations play a major role in the study, supporting the sensitive navigation of CKD management and empowering the active involvement of CYP. Chapter 5 reports the findings of the three qualitative themes that inductively developed from the semi-structured interviews.

Chapter 5: Phase 2 – Results of the qualitative dimension

5.1 Results

This chapter begins with an overview of the participants interviewed during Phase 2 of this study. Next, the three themes that developed from the inductive interpretive description (Thorne, 2008) qualitative analysis of the semi-structured interviews with CYP, parents and HCP are presented.

5.1.1 Participants interviewed

This doctoral study included 21 participants, comprising six CYP (4 males), seven parents (1 male) and eight HCP (4 males), over 17 interviews using a non-random purposeful selection approach. Two interviews were dyads (parent and CYP) and one triad (parents and a CYP).

Participants were recruited from each of the three regions within the EMEESY network, with the majority from the East Midlands. Table 30, p. 128 provides details of the participants, their profession/primary role, and the location and length of the interviews. The uniquie identifier included the research method (INT for interview), participant number (01-08), group (e.g. CYP, PAR or HCP) and gender (M or F).

Table 30. Participants interviewed

Identifier	Length of interview	Location	Group	Occupation
HCP				
INT_01HCP_F	24:56	University cafe	HCP	Consultant paediatric nephrologist
INT_02HCP_F	29:41	Multi-purpose room (MPR) on the renal ward	HCP	Nephrology nurse
INT_03HCP_F	27:05	MPR on renal ward	HCP	Paediatric renal dietitian
INT_04HCP_M	35:35	University	HCP	Consultant paediatric nephrologist
INT_05HCP_F	31:56	MPR on renal ward	HCP	Renal nurse
INT_06HCP_M	46:50	MPR on renal ward	HCP	Renal nurse practitioner
INT_07HCP_M	63:31	Home	HCP	Retired consultant paediatric nephrologist
INT_08HCP_M	53:46	University	HCP	Consultant paediatric nephrologist
Parents				
INT_01PAR_01CYP_F_F	44:50	Hospital interview room	Parent	Parent and CYP with CKD
INT_02PAR_F	35:03	MPR on renal ward	Parent	Parent of child with CKD
INT_03PAR_F	1:20:06	MPR on renal ward	Parent	Parent of child with CKD
INT_04PAR_F	47:48	Shared care EMEESY clinic	Parent	Parent of child with CKD
INT_05PAR_05CYP_F_M	53:29	University	Parent	Parent and CYP with CKD
INT_06/07PAR_06CYP_M_F_M	44:07	Home	Parent(s)	Parents and CYP with CKD
CYP				
INT_02CYP_M	32:58	University	CYP	CYP with CKD
INT_03CYP_F	1:02	Home	CYP	CYP with CKD
INT_04CYP_M	40:25	Home	CYP	CYP with CKD

Each of the three themes, namely:

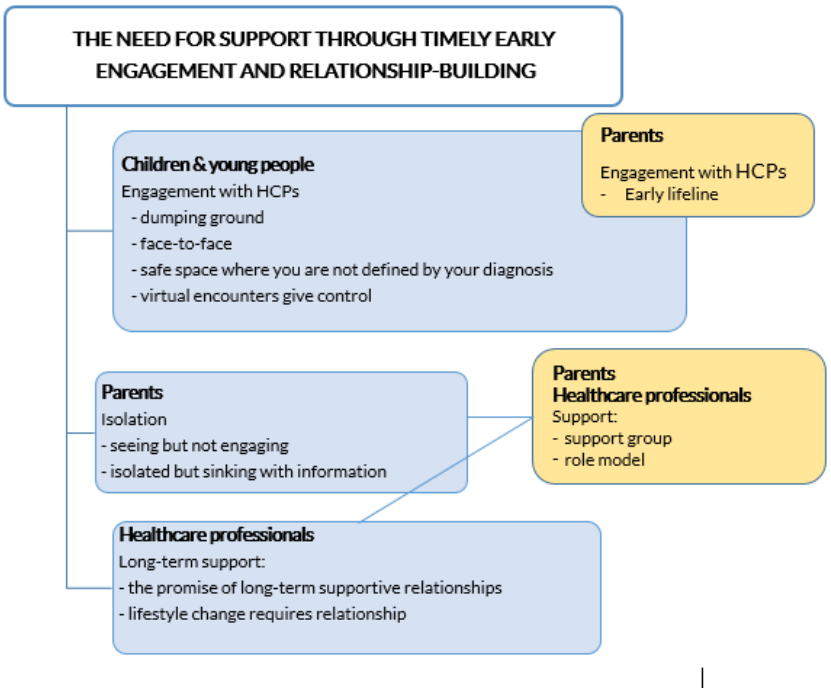
- the need for support through timely early engagement and relationship-building
- the need to take control whilst establishing a new normal
- the need for early expert dietary phosphate education

are separated into the three key response groups: CYP, parents, and HCP, with each sub-theme discussed in turn. Where a sub-theme is shared across response groups, this is depicted in the yellow shaded box (see an example in Figure 14, p. 129), with lines identifying the relevant stakeholder groups. Unique identifiers are used for participants, and all other individuals have been given pseudonyms to anonymise their identity.

5.1.2 Theme 1: The need for support through timely early engagement and relationship-building

This theme identified CYP and parents' need to receive support, and HCPs recognition of a need to provide support through timely engagement and relationship-building. The findings of this theme are summarised in Figure 14.

Figure 14. Summary of theme 1 findings



In order to explore stakeholder views on the development of an EPSM strategy, the researcher invited the CYP and parents to reflect on their own/child's diagnosis period and the information, advice and support they received at this time. HCPs were invited to reflect on their involvement with CYP and parents during the diagnosis period, considering the type of information and support they offered.

Currently, a child's CKD staging, their underlying diagnosis and social circumstances determine which members of the multi-professional team meet the family during the early stages, providing education and support to navigate the road ahead. Usually the primary care practitioners, emergency department clinicians, renal nurse and consultant nephrologist are the first to meet the child and their family, breaking the news of a chronic diagnosis. Children diagnosed with early stage CKD, often have no

input from the multi-professional team, unless social issues or medical procedures require involvement from the children's renal social worker or play specialist. Dietetic involvement is not routinely enlisted until CKD stage 3-4 when biochemistry become deranged (see section 1.3.2).

5.1.2.1 Engagement with HCPs (CYP)

Diagnosing CKD is a life-changing experience requiring timely support that recognises the need for individuals to reconcile with the difficult event. The diagnosis brings feelings of shock, disbelief, fear and uncertainty. It is a time for many CYP to seek comfort from their parents/carers and trusted others, to alleviate anxious feelings of uncertainty. For many CYP, their initial engagement with HCPs can be daunting, involving new faces and a lot of information and advice.

Dumping ground - Reflecting on this period, 02CYP, diagnosed with chronic nephritis[2] at 4 years of age, described the value of spending quality time with people he understood were going to be involved in his care. Before receiving the information, he remembered the impact of those who took time to get to know him and understand his needs. For 02CYP, when a consultation seemed hurried, with a primary agenda to deliver 'a load of information', he discerned minimal interest in him; leaving him feeling like a dumping ground. One encounter during the early days with Peter (pseudonym), a youth worker, made a lasting impact. It was a 2-way communication process as Peter spent time with him; they 'got to know each other'. The exchange imparted trust and confidence, with a desire to follow the advice received. The significance of this encounter shaped his values on the importance of authentic early engagement before the information barrage. For 02CYP, an initial encounter should involve no hidden agenda. Advice should not subtly be introduced, rushed, or slipped in as a written prescribed treatment strategy. Encounters didn't need to be lengthy, just an introduction, and if possible, a sincere interest in him as a person.

> ***CYP:*** *…they'd give you a load of information. And it's like, 'oh, I've never met all of you'…Yeah, I think a more personal level of understanding of the team would be beneficial …when I joined the youth team, when I first interacted with them, Peter came*

[2] Nephritis refers to inflammation of the kidneys

> *down from the renal youth team, and he sat down with me for 20 minutes, and we had a chat, we got to know each other and that was really beneficial. Then when I did things with the youth team, that felt like that was one of the first times I had really met a member of staff well before receiving a load of information about the disease and stuff…and then it helps the relationship interest in the future.*
>
> ***(INT_02CYP)***

Face-to-face - For many CYP dealing with a chronic illness, their impact circle includes face-to-face engagement with family, friends, and guiding adults. Friendships offer strength and encouragement, providing key networks of support. For 03CYP, a 13-year-old traversing the stresses of GCSEs and peer pressures, friendships were 'literally the backbone' to her world. During the early days of facing her CKD diagnosis she felt vulnerable and anxious, reaching out to friends as a sounding board for reassurance and direction. No matter what lay ahead, there was always someone there for her, someone who cared, and took the time to listen. These stalwart pillars of strength brought solace at this vital time in her life.

> ***CYP:*** *… usually a lot of support that I get comes from my friends… and they're literally the backbone. If I ever I've had a bad day or anything, they come over and they support me… I think if people [family and friends] weren't here to support me, I probably wouldn't have gotten through it.*
>
> ***(INT_03CYP)***

Safe space where you are not defined by your diagnosis - As we explored the idea of an EPSM strategy, 03CYP began to share what she perceived support from the renal team would have looked like during that early period. The diagnosis of CKD brought a lot of uncertainty, generating feelings of anxiety for many CYP. 03CYP suggested that the EPSM strategy should be linked to an 'outreach program', offering opportunities for young people to connect with others; a safe space where you are not defined by your diagnosis. She wanted to hang out with other young people just like her, in an informal, non-judgemental environment, with easy-going access to professional support. There would be no rush or agenda, HCP would have time, and a genuine interest in getting to know and support you.

> ***CYP:*** *You know, maybe somewhere where there's trained professionals, people like psychiatrists, and whatever, and carers, and you've got loads of little activities. You've got like a separate little room. So, if people don't want to be in a large crowded room, they can go into a little separate room. They can have a one-on-one with somebody if they need to; and then sometimes have events, but it's always optional. So, you don't have to go if you don't want to, but the offers there if you need to.*
>
> ***(INT_03CYP)***

Virtual encounters give control – since the advent of mobile phones and social media, virtual engagements have dominated social encounters for many young people, decreasing the frequency of face-to-face contacts (O'Keeffe and Clarke-Pearson, 2011). For 03CYP virtual and physical encounters were intertwined. Meeting someone face-to-face has its time and place, providing necessary steadfast support; other times they are a source of pressure producing anxiety and a craving to retreat to her virtual world to gain instant access to friends. These virtual encounters gave her control, creating a levelling effect in her world.

> ***CYP:*** *Usually its real life [face-to-face] sources that make me upset, and then the online sources actually stable it out.*
>
> ***(INT_03CYP)***

Observing and engaging with Snapchat or Instagram, for example, allowed bite-size, impact junctures to punctuate her day. Sometimes these encounters were chatty and light-hearted; other times they were intense and unhelpful, both were important to her and other CYP.

Alongside support from friends, 03CYP described her need for support from HCPs. As a curious thinker, unanswered questions created anxiety and a need for control. She desired answers and an explanation; but she felt telephone conversations could be 'awkward', placing expectation on her to speak when she didn't want to. While remaining anonymous, she wanted to have a voice. Accessing a text hotline, she suggested, would allow her to post a question or comment, giving her control of the interaction, with no pressure to talk about something she didn't want.

> ***CYP:*** *I think I would like a texting service … I don't really like calling, I'm not the type of person to sit there on the phone and lay my voice to other people. I like it when, you know, it's kind of anonymous…Because then, you don't have to share anything you don't want to.*
>
> ***(INT_03CYP)***

Early lifeline (parents) - Prematurely giving birth to twins, then tragically losing one in utero, followed by the surviving daughter being diagnosed with CKD, 04PAR was left bereft with a desperate need to hold on to what remained. Fraught by the crisis of events, she struggled to process what was happening. The torrent of activities in the early days instinctively caused her to withdraw, unable to take any more, 'I didn't, I didn't want to know '. Yet, everything within her cried out for a lifeline - a HCP that would provide reassurance and direction, informing her of what she needed to know and how to process her grief. Driven by the intensity of her circumstances, she was thrown from overwhelming grief to desperation to survive. Despite wanting to shut down internally, her survival instinct surged into overdrive causing her to hang on to every morsel of information and advice she could access.

> ***Mother:*** *But I didn't, I didn't want to know. Because I'd already lost one baby, which was horrible, I didn't…and they said that she might die, and I didn't, I couldn't, I couldn't process what was happening in my brain. So, I was just like, I don't want to know, 'just do what you need to do, and just tell me what I need, I need to know.*
>
> ***(INT_04PAR)***

Worries about diet added to the mounting anxieties experienced by 04PAR. Reflecting on the early diagnosis period she described how valuable it would have been to meet the dietitian, to 'have that face to a name' and establish a meaningful connection. This early engagement would have provided reliable, trusted dietary guidance and support, 'a bit like a crutch'. She desperately wanted to connect with an expert who was able to provide the knowledge and reassurance she needed to best manage the nutritional needs of her treasured daughter. Doing her best, she was horrified to be told by the doctor that [3]breast-feeding may better serve as a

[3] Breast feeding may be discouraged when a child has CKD, as it is difficult to quantify the volume consumed and the electrolyte composition (Nelms, 2018).

supplement and not her daughter's primary feed. Living a distance from the tertiary centre, a meaningful connection was made with the children's doctor in her local hospital; he acted as a sounding board, answering her many questions about food and eating.

> ***Mother:*** *I think diagnosis would be good to have an informal chat [with the dietitian]…Because hearing it from a doctor is different to hearing it from a dietitian … And it's a bit like a crutch, like I know you're there. Whereas when it happened, I was like* ***whoa****! I've never been faced with this in my life before…it's very, like bowls you over a bit. …But those days, those early days when she wouldn't eat, I'm so glad that Dr R [consultant at the local hospital] was around the corner. Because I used to ring him all the time … To have him there was so good, because it was like, right she's not very well, let's ring …. Because it is quite scary when, you know, crikey me, particularly as a breast-feeding mum as well. It was, when you're told like your milks like hurting your baby, it's like, what!*
>
> ***(INT_04PAR)***

5.1.2.2 Isolation

Many parents described the diagnosis period as intense, with lots of appointments to schedule, people to meet, and information to process. Whether parents were entering the renal arena for the first time or had been attending the tertiary centre for their child's treatment for many years, they recalled feelings of loneliness and isolation. Attending the haemodialysis unit at the hospital thrice weekly, gave many parents the opportunity to build relationships and establish familiarity with key HCPs and parents.

03PAR had a traumatic experience when her daughter was diagnosed with CKD after an acute illness, requiring dialysis at 6 years of age. Desperate to gain answers and understand more about her daughter's condition 03PAR expressed an ardent desire to connect with others at a meaningful level. In her search for family and friends who understood her situation, feelings of loneliness and isolation emerged. The poor public image of CKD, often portrayed in the media by distorted storylines, all added to her lonely frustration and drive for authentic connection.

> ***Mother:*** *It was a shock, complete shock. I wasn't expecting it, and even, I just kept thinking, 'I'm sure it's just probably a really nasty infection that took hold, and she's going to get better very quickly.' You know they said, 'she has to have dialysis,' and almost, within sort of twenty-four hours of being here the swelling got worse, she looked just really poorly, and it just kind of happened all very quickly…*

> **After her daughter had been attending dialysis for 12 months**
>
> *I suppose this is coming from sort of spending the last twelve months here really [attending haemodialysis 3 times each week] is that kidney disease is very low profile. So when you talk to people in your family or your friendship groups, there is not people…that know a lot about it, unless they've experienced it themselves in their own families… and I don't really know who to talk to about it… There is no cure; this is for life … and you get really rubbish storylines on Coronation Street where somebody has been transplanted in about a week, and they're up and about… that doesn't help [daughter's name] …I can't watch it…*
>
> ***(INT_03PAR)***

Although for some parents CKD was viewed as just 'another thing' to deal with (INT_05PAR), building a meaningful supportive relationship with a knowledgeable HCP was invaluable at this difficult time. For 05PAR, complications in utero led to her son being diagnosed with CKD in his first week of life, needing urgent and ongoing regular renal appointments for the first year.

Seeing but not engaging (parents) - Despite frequent hospital visits, the season that followed the diagnosis left 05PAR feeling vulnerable with uncertainty 'Will anyone be in touch?' and 'Does anyone really care?' She longed for someone to call to ask, 'How are you doing?' Much like 04PAR, she sought a genuine lifeline to provide the much desired connection and support. She didn't just want information or be reminded of the impending changes; she wanted someone to understand, to express an interest in her world, sowing the seeds of an early relationship. The relationship established with her son's consultant was esteemed as a great source of support. Indeed, it was singled out as a chance encounter, 'we've been lucky in that'. Despite 'seeing' many HCPs at clinic appointments, the opportunity to engage and get to know them was intangible. She was surrounded by potential connections, but limited time in clinic and no invitation to engage, meant the longing remained and the isolation increased. At the time, with so much going on, 'I wasn't bothered', she remarked, but clearly, she was. Her lack of time was mirrored by the absence of time or recognition of her need by the HCPs. Meaningful encounters with HCPs were desired; 05PAR emotionally described her experience during those early months:

> ***Mother:*** *Once the initial advice had gone I didn't really have much input, like 'How are things going?', 'How are you?' I mean, I suppose it was different for me, because I was really busy, because I had two at the same age … for me it was just another thing. It was stressful anyway… No-one really kind of called me, and said actually, 'How are you getting on?' …'Do you need any more advice?' That probably would have, just on reflection, at the time I probably didn't really, I wasn't bothered, yes, but I think that might have just…contact 'how are you?' sort of…maybe build up a relationship, because you don't really know who. You come to the clinic and you see different people. Like, we've always had Dr C, we've been lucky in that; we've always had him, since birth. But other than that, we've seen lots of different people.*
>
> ***(INT_05PAR)***

Isolated with information overload (parents) – Similar to 02CYP (refer to 'dumping ground'), there was no lack of information and advice from HCP; on the contrary, 03PAR felt 'overwhelmed', sinking under the weight of it all. Comparable to 05PAR (refer to 'seeing but not engaging'), even when attending regular clinic appointments, and surrounded by HCP undertaking invasive procedures, 'you don't really see' anyone, then you are 'left to it', going back to your own little world where no-one really understands. 'Seeing' for 03PAR was not just a passive contact but meant connecting on a meaningful level.

> ***Mother: …*** *it was like hitting a brick wall, and having all this information and thinking, 'oh my goodness' just completely overwhelmed… If you've got children that are perhaps, either not on dialysis, but are coming to clinic, or even the children that are on PD, you're a bit isolated really, because you don't really see [anyone], you come to clinic you have bloods…When [daughter's name] was on PD [peritoneal dialysis] I just felt very kind of out of the loop really, and OK you can ring and ask for advice but it was very much kind of we were left to it.*
>
> ***(INT_03PAR)***

The mother of twins (05PAR), who earlier described her longing for real engagement, spoke of her son's isolation and loneliness. She described how difficult it was for him to get to grips with CKD, particularly not knowing any other young people with CKD.

> ***Mother:*** *That's something that [he] definitely struggles with…and he thinks that he's on his own.*
>
> ***(INT_05PAR)***

5.1.2.3 Long-term support

The promise of long-term supportive relationships (HCP) – An experienced consultant paediatric nephrologist reflected on the countless difficult conversations with parents. Delivering a diagnosis of CKD is life changing, and parents frequently race ahead, rehearsing anxieties about their child's future aspirations. What they do not recognise at the time, is their need for the promise of long-term supportive relationships. HCPs shouldn't just go through the motions distributing information and advice. Rather, each family is unique, and it takes time to get to know them and support their long-term needs.

> ***Consultant paediatric nephrologist:*** *If you sit across the table from parents and say, 'Your child has chronic kidney disease', that changes the family prescription forever! The first thing the parents are thinking about is 'My God, they're gonna' need a kidney transplant' or 'Are they ever going to have kids?', 'Will they go to school normally', you know, and 'What's the long term future?' They jump to those things straight away. It happens on every chronic illness. So, then you have to kind of get over that kind of shock and support them with the advice. But you are in for the long haul. You know, you have to know how each child is likely to progress... it's all about building relationship and sharing that information.*
>
> ***(INT_07HCP)***

Many early stage CKD patients seen in the EMEESY network peripheral shared-care clinics, are not offered a paediatric renal dietetic service. Speaking of the shared-care clinic setting, 08HCP a consultant paediatric nephrologist, explained 'it might just be the consultant and it probably needs to be a more multi-disciplinary approach'.

In 08HCP's appreciation of the importance of early nutritional advice, particularly in promoting lifestyle change (as mentioned below), he thought that asking a tertiary dietitian to call a parent from a shared-care clinic without prior face-to-face participation or insight into family dynamics, their needs and management style, could make a telephone consultation hard. In addition, the efficacy of the advice would possibly be limited.

> ***Consultant paediatric nephrologist:*** *And because a lot of it is on the therapeutic relationship you build with someone and it's a lot harder to do that if you're calling out of the blue and giving that advice over the telephone.*
>
> ***(INT_08HCP)***

Lifestyle change requires relationship (HCP) – Participants described how they adapted their style, gauging how to best support each CYP/family. For example, this dialysis nurse described how he 'can chat to someone that's a road sweeper or … a professor' (INT_06HCP). Reflecting on the complexity of the dietary prescription, these HCPs (08HCP and 06HCP) recognised that dietary advice involves more than a prescriptive line of information, it demands lifestyle change that takes time, affecting the dynamics of the entire family. To be effective the dietitian needs to build a trusted and respected relationship, inspiring motivation over time. Advice needs to be tailored to the nuanced, changing and evolving needs of each CYP and their family.

> ***Consultant paediatric nephrologist:*** *I think it's, you know, diet changes are lifestyle changes… That is hard to give if you don't have that relationship that you've built with someone, where they know you, and you have that element of trust and respect for you. Then you can get to know them and find out what works for them, give them specific advice and support in the areas they find difficult, or you know the areas that are new to them.*
>
> ***(INT_08HCP)***
>
> ***Specialist dialysis nurse:*** *It needs to be tailored to…and that's getting to know your patients. And that, I think is probably difficult with the early intervention patients…Because some I know just won't, just won't retain it, so I have to tailor just how I do it. But that's knowing them.*
>
> ***(INT_06HCP)***

5.1.2.4 Support

Connecting with others (parents) - Meeting other families that have experienced similar situations can help parents to better process information. These mothers of twins, 05PAR and 04PAR, spoke of their desire to link with other families, and for 05PAR's son to meet other young people, to realise he's not the only one experiencing this diagnosis. Feeling frustrated, and longing to tear her son away from the vices of social media, 05PAR described the need for a support group - meeting face-to-face with other families, spending time connecting with them.

> ***Mother:*** *Do they ever have support groups, not social media ones, but actual support groups where families can come? You know, that could be something that could be done, even for the children to meet other children.*
>
> ***(INT_05PAR)***

> ***Mother:*** *And I remember when I had [daughter's name] you Google straight away…It's good in some ways, but frightens the life out of me in other ways as well…But to have, I think as well like introducing people to other parents is really good as well…It's a lot to take in with the first diagnosis like…but just making it really friendly as well.*
>
> ***(INT_04PAR)***

Follow my lead (HCP) – As a means of strengthening or substituting advice on early dietary phosphate, consultant paediatric nephrologist 08HCP shared a vision for peer support in the paediatric renal community. Parent and CYP EPSM champions - motivated individuals or families who have already made healthy lifestyle adjustments – could be venerated to model early dietary phosphate education, inspiring and motivating others to follow their lead. 08HCP felt this would echo the way society used to operate. Individuals with lived experience, would have more of an impact on their peers.

> ***Consultant paediatric nephrologist:*** *… we did use to have much smaller communities…people learning from others within the communities, how to be healthy how to either cook and develop those life skills. Although it's a healthy-eating message whether it's easier to hear and take on board something that's coming from someone else who is in the same situation, rather than a health professional …*
>
> ***(INT_08HCP)***

Having a peer support model resonated further with 05HCP in terms of using the community voice as a tool for teaching others. She reflected on previous successful initiatives in the renal team where HCPs have organised and facilitated families to support and inspire each other through shared experiences and support groups.

> ***Specialist renal nurse****: When you are introducing new families, and you have already got some families that are already doing it [early dietary phosphate management], then probably a support network, and do it that way through…like we've always done, we link 'em with another family that's going through that.*
>
> ***(INT_05HCP)***

5.1.2.5 Summary of Theme 1

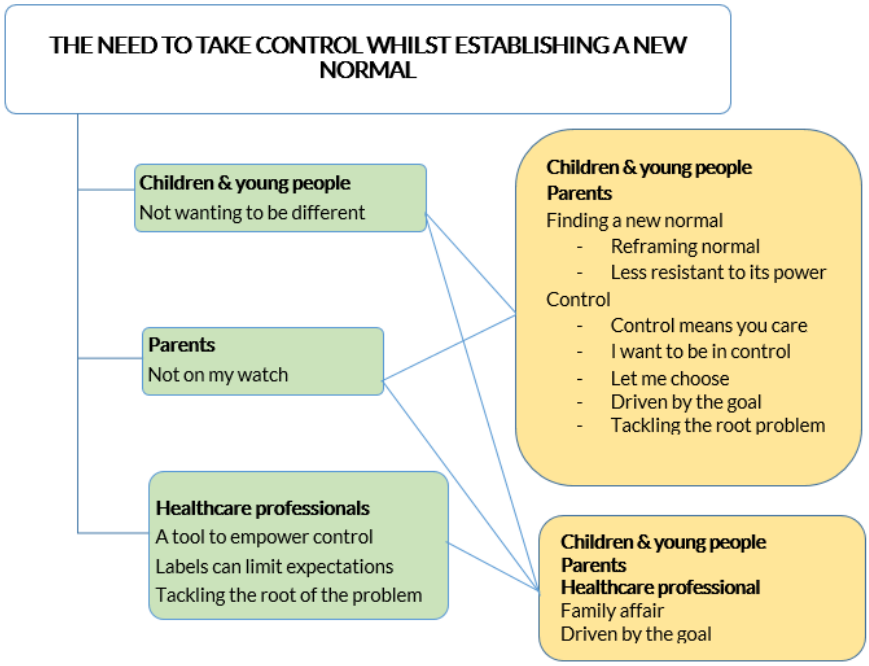
This first theme 'the need for support through timely early engagement and relationship-building', captured the desire of CYP and parents to make meaningful connections with HCP and other families during the early diagnostic period; providing the much needed advice and solace from those knowledgeable in the medical and real-life management of CKD. Such connections would help with the intensity and associated trauma of diagnosis, negating feelings of isolation throughout the CKD journey. While appreciating the need to communicate large amounts of information during the diagnosis period, authentic engagement, leading to long-term support, was sought by all stakeholders. Face-to-face and virtual networks were suggested as possible initiatives to educate and support families long-term.

5.1.3 Theme 2: The need to take control whilst establishing a new normal

Parents described how once they recognised and understood the magnitude of the renal diagnosis and their need for lifestyle change, their initial desire to take control was followed by the navigation of a new level of normality. 'I'm starting to think now that … we're in a routine, and not the nicest one…but this is normal life for us' (INT_03PAR). Participants described many drivers of control. Firstly, while 03CYP wanted to make her mark, she also wanted to conform, not standing out from her peers. Critically, parents exerted a protective role, seeking to do all that they could to make the situation better for their CYP. Control over their CYP's dietary choices often resulted in a struggle for power. However, some CYP recognise this parental control as an expression of their caring role.

The findings of this theme are summarised overleaf in Figure 15, p. 141.

Figure 15. Summary of theme 2 findings



5.1.3.1 Not wanting to be different

Some of the CYP interviewed perceived that compliance with the EPSM diet would mean standing out from their peers. For 03CYP, conformity was important, as being different would attract undue attention. As most of her social engagements involved meeting at McDonald's, this would make her the target of attention, or even worse she may feel socially excluded. Reflecting on her father's veganism, on the occasions when he socialised with her, eating out at McDonald's was awkward, provoking many questions from her friends. If she followed the EPSM diet, she envisaged her friends would similarly view her as 'different'.

> ***CYP:*** *… for example, my dad's a vegan, and we've been out with friends., … and sometimes my dad's come along, and I've gotten asked quite a few questions, cos' obviously my dad can't have anything when we go to McDonalds. He just gets a packet of salad or whatever and some fries. My friends are just like, 'Why does he have to do that?' So, I suppose it's a bit weird knowing that I might be different from everyone else.*
>
> ***(INT_03CYP)***

In addition, 03CYP considered that when visiting friends the EPSM diet could become burdensome, putting extra pressure on them to meet her needs. This inferred another

source of anxiety for her.

> ***CYP: …****, for example, her mum and dad are already really concerned, and they, whenever I come over they literally choose the lowest salt food possible. And it's just the fact that I don't want to make anything, you know, more difficult for them than it already has to be.*
>
> ***(INT_03CYP)***

5.1.3.2 Not on my watch

Young children are often more acquiescent of parents' wishes. As children get older, parents seek to hand over a degree of control, while desperately keeping a grip and fervently looking for ways to influence change through their own example and lifestyle choices. During the interview, the EPSM diet was explained to each participant. Once parents understood the high inorganic phosphate content of processed foods and its potential to accelerate CKD progression, they were keen to understand more. Some were determined to change their child/family diet, implying they would not let this happen on their watch. When a renal dietary change is encouraged, parents adjust to a new set of rules, by navigating control. Concurrently, CYP strive to make sense of the new rules, whether to adopt them or fight back in defiance. Parental role models and family beliefs provide a framework, shaping the young person's response to change. This interview with 05PAR and her son 05CYP captured this tussle to influence dietary change and establish a new normal.

> ***Mother****: Not long ago, did Dr C tell you not to have Coke?*
>
> ***CYP:*** *Yeah, I've not had a Coke. I've never had a Coke. Well, I have had a Coke, but I've not had it since he said…*
>
> ***Mother:*** *He has it now and again, but…*
>
> ***CYP:*** *Yeah, like once every month.*
>
> ***Mother:*** *I mean, I don't drink Coke. I don't like it. And I would prefer him not to have it.*
>
> ***CYP:*** *Mum has Pepsi Max every so often.*
>
> ***Mother:*** *And I don't even really have that, do I?*
>
> ***CYP:*** *She barely has Coke. I only have Coke, like, once every…*
>
> ***Mother:*** *He is allowed it now and again as a treat, but before he might have had it more often.*
>
> ***(INT_05PAR, INT_05CYP)***

05PAR described the reasons for her lifestyle decisions, resulting from her father's death from bowel cancer, which the GP had claimed to be associated with his diet. This loss influenced her abrupt decision to stop eating processed meat; making it her mission to control family food decisions, especially her son's diet. Although she said, the family 'just gave it [*processed meat*] up', in truth they didn't, though she ardently disagreed with their choices, and took action to influence change. As a result, her husband decided to follow a 'plant-based' diet, and she vowed to never serve 'any remotely processed meat' at home as 'I'm against' it. When the EPSM diet was explained to 05PAR, it resonated with her judgement, and she affirmed her decision not to let it happen on her watch.

> ***Mother:*** *So, I'm completely meat free. [Husband's name] probably just this week has decided that he's not having any more chicken. He's given up like red meat … he decided he wanted to be plant-based himself. So we did that research. So he [her son] doesn't have a lot of them things. Like I say, if he goes to McDonalds, he actually doesn't choose meat, he chooses chicken. But that's a treat again.*
>
> ***(INT_05PAR)***

Although 05PAR's strong convictions sought to influence her son's food choices, she ultimately wanted him to own his beliefs. Despite acquiescing to her wishes, 05CYP realised he could occasionally deviate from the rules by asking his father. Navigating his mother's family rules, and the father's inroads, highlighted the need for a unified family approach and his journey to finding a new level of normality.

> ***Mother:*** *He's not allowed to have bacon.*
>
> ***CYP:*** *I barely ever have it. I have it like once or twice a year.*
>
> ***Mother:*** *He wouldn't have bacon…sausage, any remotely processed meat, from me anyway, because I'm against processed meat.*
>
> ***CYP:*** *My dad sometimes lets me have it like, once every now and then.*
>
> ***(INT_05PAR, INT_05CYP)***

5.1.3.3 A tool to empower control

Participants were asked during the interview whether the EPSM diet would be perceived as empowering or imposing. It became apparent that many parents were

eagerly looking for credible information to guide their actions. By teaching CYP and their family the healthy-eating message of early phosphate management, parents could be equipped with the tools to potentially change the trajectory of their CYP's CKD. Where previously they had been at the mercy of the HCP and the natural progression of the disease, this empowering EPSM message would offer them some early control. 04HCP, a consultant paediatric nephrologist, described parent's eagerness to do something to help their loved ones.

> ***Consultant paediatric nephrologist:*** *…in the past patients always say to us, 'Oh I've got kidney problems, what can I do?' and before we say, 'Oh not much, we just have to wait and see when you go on dialysis'. So actually, this is a way of being more positive and framing it in a very positive way…*
>
> ***(INT_04HCP)***

By giving individual's choice, 06HCP a specialist dialysis nurse explained, it imparts power and control. If parents were told the potential impact of following the diet on their child's disease progression, it would serve as a powerful tool, empowering individuals to take some control.

> ***Specialist dialysis nurse…*** *I think if you were saying, '…ok, it's in your hands, if you do this, it might be longer before you meet [the dialysis nurse] in that room.' Do ya' know what I mean? And if you want a picture of me! That's fine. No, you know. I think if you were giving it to them in that way, and saying, '…actually it will be longer before we start seeing you regularly in Nottingham. Travelling there for monthly bloods', then you know, empowering.*
>
> ***(INT_06HCP)***

The consultant paediatric nephrologist, 08HCP, while affirming the empowerment of handing over control to parents and their child with CKD, acknowledged the mismatch that can exist between the clinicians drive to best manage each child's condition, and the individuals who separate 'health…and the rest of their lives'. He perceived some parents would grab this empowering tool with both hands, while others would need more encouragement and support. When attending clinic some parents and CYP are eager and switched on to learn, but once they exit the clinic room, they have the rest of their life to live. Thus, the EPSM tool needs to bridge

managing health and encouraging individuals to find this new level of normal in their everyday lives. It needs to empower change without becoming imposing or burdensome.

> ***Consultant paediatric nephrologist:*** *… There's also the possibility that people sort of come to their clinic appointment and that's the time when they want to think about their health and the rest of the time they want to think about the rest of their lives. Obviously, we [clinicians] spend our whole working lives, and probably some of our home lives thinking about kidney disease and strategies for it. On the other hand, I think some people like not to do that. So it is …helping everybody … to self-manage well and for it to be empowering rather than something that becomes sort of a burden.*
>
> ***(INT_08HCP)***

Another way that parents eagerly seek control is by searching the internet for guidance. The consultant paediatric nephrologist, 04HCP, believed the provision of credible EPSM evidence-based information and resources would empower families, and protect them from quasi SM strategies. He recognised that any new intervention can provoke anxiety, and therefore must be offered in a way that inspires, empowers and is unforced. Individuals want control, they want to retain 'their lifestyles, their rights', this again depicts a striving to hold on to what they view as normal. In their own time they may choose to make changes. This is when the EPSM advice needs to be available to steer them along the right path, to ensure they follow credible, effective counsel.

> ***Consultant paediatric nephrologist:*** *This kind of internet self-management…through Google… actually sometimes, when you introduce an intervention you introduce anxiety… because you have to adapt it them. You don't want to cause extra stress. I think that maybe in the UK we are very mindful about the state; you can't have any enemy state. You can't say, you know, don't smoke, don't eat too much bacon because it causes cancer. But the truth is these things are bad for you, and actually I think it's our responsibility to tell families… In UK I think we have to be mindful people, you know, people like their lifestyles, their rights.*
>
> ***(INT_04HCP)***

These HCPs, the paediatric renal dietitian, 03HCP, and the specialist dialysis nurse, 06HCP argue that all families irrespective of their skills and often limiting social economic status should be supported to embrace the EPSM diet; even small changes

are a positive step forward. Working mums with busy lives, for instance, may depend on processed foods, while others with limited cooking skills may be impeded from embracing the diet. As stated above (see 5.1.3.1 'not wanting to be different'), processed food is strongly linked to the social life of young people, and therefore it is essential to find realistic ways of supporting individuals with the practical, financial and behavioural change aspects of the EPSM diet.

> ***Paediatric renal dietitian:*** *One of the things that worries me about it is we heavily rely on convenience foods in the western diet, and I think a lot of families are working … The older children with sort of going out with friends, if you were trying to get them to cut down on the amount of things that they buy, then that's got a social implication…sometimes I find with patients that they don't have the knowledge to just do the basics, and to try and get somebody to cook a meal on top of that would be hard … With teenagers it's always hard but you can choose where you make the most significant changes. If they're having one less product, it's still an improvement.*
>
> ***(INT_03HCP)***
>
> ***Specialist dialysis nurse:*** *And that doesn't mean you should write off the ones who you don't think will do it. They still need the information. But I think they're the ones who probably will carry on buying the processed food… because, actually the majority of them can't actually afford to change that… and as much as Jamie Oliver says you can eat for a fiver, then, we know you can't, and actually turkey twizzlers do cost less than a joint of beef, its reality, even though one's got more phosphate than the other.*
>
> ***(INT_06HCP)***

5.1.3.4 Labels can limit expectations

In contrast to the empowerment the EPSM diet could give to families, the early introduction of a medically approved nutritional strategy could generate stigma. Most CYP diagnosed with early stage CKD will not feel unwell or experience any adverse symptoms. A primary concern of the consultant paediatric nephrologist, 08HCP, when speaking to parents about the early diagnosis of their child's CKD was the 'sickness or anxiety label' that they imposed upon their child, leading to school absenteeism and psychological concerns. He feared the introduction of EPSM may likewise create a label, which some parents may use inappropriately to leverage attention, limiting their child's potential. He identified the need for the EPSM strategy to be versatile, having a framework that could be adapted to individual's needs.

> ***Consultant paediatric nephrologist:*** *I really want to encourage people … be expecting that its children, you know, they will grow, they will complete their education, they will be physically able. You know not to expect them to feel any symptom at that stage … People with actually relatively mild kidney conditions who label themselves or their child as having a disease … you know school attendance can drop off…and problem with low mood … I think that there are clear benefits in empowering people, and it's having the right structure in place so it's flexible enough to be used in the right way for the right people.*
>
> ***(INT_08HCP)***

5.1.3.5 Tackling the root of the problem

As discussed in section 1.3.2, the NICE hyperphosphataemia guidelines (2013) recognise the need for the specialist paediatric renal dietitian to provide bespoke dietary advice when serum phosphate levels start to rise during CKD 4-5. However, by this stage many detrimental bone changes and early signs of CKD-MBD have already begun to take place. For these consultant nephrologists (04HCP and 07HCP), the EPSM strategy is not only one of prevention, but it would mean really getting to grips with the 'root of the problem'. They argued that dietary intervention needs to start early and continue through the CKD journey. Although medicines can be prescribed to treat the deranged biochemistry, they do not address the real problem.

> ***Consultant paediatric nephrologist:*** *With the stage 3 we know that there is a lot of things really that can affect their CKD mineral bone disease…you can already see that in the biochemistry results. So, your PTH[4] would go up, and you know the only thing that we can do is give them medications, so give them alfacalcidol, maybe treat their vitamin D. But we're really not tackling the root of the problem, which is you know where the phosphate is coming from. What's driving the PTH? …we really do need dietetic input.*
>
> ***(INT_04HCP)***
>
> ***Consultant paediatric nephrologist:*** *If we have evidence of renal impairment then the focus is mainly on… reducing phosphate intake, which is probably one of the major triggers for long-term bone conditions, and its association now with cardiovascular disease… but it's far better to try to ameliorate the diet at an earlier stage. That's a long-term educative process…and that's why I would strongly believe that the dietary involvement in the predialysis stage, at an early stage is really worthwhile.*
>
> ***(INT_07HCP)***

[4] Parathyroid hormone – a type of activated vitamin D given to people to treat secondary hyperparathyroidism associated with CKD

5.1.3.6 Finding a new normal

Reframing normal - Facing a diagnosis of CKD requires adaptation in order to establish a new normal rather than lamenting what could have been. Adaptation takes time and parents instinctively want to protect their children, seeking ways to provide reassurance and rationale that their child's differing needs are in effect uniquely normal, or another way of framing what is good for everyone. At this confusing and anxious time in her life, 03CYP reflected on her mum's reassurance regarding her diet and underlying diagnosis – kidney dysplasia[5].

> ***CYP:*** *Mum said to me… 'Your diet it's not really a specialist kind of diet, it's a diet that everybody should be following.'*
>
> *[The doctor] showed mum on the screen and it was…my left kidney which was you know, a normal size, and right kidney which was tiny and has a scar across it … I broke down into tears, cos' I didn't realise what it meant for me at the time, but I knew that it was definitely not a good thing, you know. I do remember one thing that she said to me was, she tried to reassure me by telling me, you know like some people don't have feet that are the same size…*
>
> ***(INT_03CYP)***

Less resistant to its power - Reconciling to the new normal brings tension - anxiously holding on to the past, while becoming more accepting of the present. 03PAR, the mother of a child receiving home dialysis, described how she has changed by having a child with CKD and it took time to concede to its influence. As the new normal moved in she became less resistant to its power. An even unpleasant routine provided order and gave structure to her new way of life.

> ***Mother:*** *The circumstances change, and actually you change as a person, and what you might have held on to in the past, you know, as you become more accommodating of what's happened, you might then have more willingness to perhaps change things…I'm starting to think now that actually we're in this now, we're in a routine, and not the nicest one to say, a routine but this is normal life for us.*
>
> ***(INT_03PAR)***

[5] Kidney dysplasia is a condition in which the internal structure of one or both kidneys does not develop normally, resulting in a small misshaped kidney.

5.1.3.7 Control

Control means you care – Although CYP reluctantly concede to parental control, this young person recognised, that what she viewed as the obsessive 'crazy' behaviour of her mother, was actually a caring language. Although her behaviour remained annoying, knowing that she cared made her control easier to accept.

> ***CYP:*** *Mum … started going crazy when she first found out [about the CKD diagnosis] … I know she went to quite extreme lengths about my food as well. She was saying like, 'Ok, you've had that bit of cheese, no more cheese for the rest of this week' you know. She bought me those, you know Quorn, those Quorn meats, apparently they had lower salt, so she was buying me all those. You know, she was keeping track of basically everything I ate. I think of course I know it's really good that she started doing that, cos' obviously she cared about me, but it didn't stop getting annoying.*
>
> ***(INT_03CYP)***

Similarly, 05CYP recognised his parent's checks and boundaries as a desire to protect him.

> ***CYP:*** *Now when I get a fizzy drink, I have, dad has to check what's in it.*
>
> ***(INT_05CYP)***

I want to be in control – For 01PAR whose daughter was diagnosed with established renal disease, not satisfied with the practical application of the information she received at the hospital; she searched for online material to inspire confidence to manage her daughter's diet. Desperate to exert control, she undertook some independent web-based studies. On completion she was delighted with the renal recipe book reward.

> ***Mother:*** *Cos' I actually did a course online … yeah 5-day renal course of reading up … cos' I didn't know what I was doing. I hadn't got a clue. Yeah, recipes and stuff though that I could pick for her. You know like snacks for in her lunchbox… New ideas, new things that they can eat…it is tough, it is tough … I had to then like answer questions to see if I was like taking it in. At the end of it I got like a recipe book online, they sent to me. It was dead good, and mean some of the things she'll like no I don't want that, but some of the things she tried.*
>
> ***(INT_01PAR)***

Dietary knowledge gave these mothers the power to alleviate harm. While providing around-the-clock care for her son with established CKD and other complex health needs, food became a central focus for 02PAR, allowing her to correct adverse deviations in his blood levels. For 04PAR, losing a twin daughter at birth made her desperate to understand what was beneficial for her child. They passionately shared their views.

> ***Mother:*** *It's the control, it's the power of being able to make the changes … if you've got the understanding… I'm in control and that is a massive part… I want to be able to control as much of this as I can, and when it's something, I'm not saying as silly as food, but something that could be fixed so easily, just with more understanding …*
>
> ***(INT_02PAR)***
>
> ***Mother:*** *I wanted to be in control …cos' I wanted to make sure that I knew what, what she was eating was gonna', it wasn't gonna' make her kidneys worse.*
>
> ***(INT_04PAR)***

Let me choose (CYP) – When considering the EPSM diet, the researcher asked participants whether it should be prescriptive or optional. For some a prescriptive diet made them feel informed, imparting trust. For others a choice was preferred, making them feel valued and involved, in turn this motivated change. Having choice for 04CYP offered him human agency, making him better informed to decide. If he had been encouraged to change his diet earlier, he would have viewed this positively, seizing the opportunity. By nature, patient choice allows deviation from 'best practice', which can leave HCPs frustrated or exasperated. However, as 04CYP commented, if an individual chooses not to opt in, the education and support can be directed elsewhere, making better use of available resources.

> ***CYP:*** *If I found out that I could have it [EPI diet] earlier, then yeah, I would take that opportunity… A choice would be best, the people that, like you insist it on, might not want to, so then it would be a waste of NHS time like, they [HCP] could be helping someone else. Then if it's more a choice then its people who want to follow can do it.*
>
> ***(INT_04CYP)***

Similarly, this young person asserted strongly that the EPSM diet should definitely be offered, but once offered, it should be a choice. Food choice is multi-faceted,

incorporating conscious, automatic, habitual and subconscious decisions (Furst et al. 1996). People make choices about what they want to eat, and as 03CYP reasoned, 'diet is part of their freedom'. She implied that by imposing change, one is pushing against these deep routed facets of an individual's identity. Until there is definitive evidence, not just an association between reducing early dietary phosphate and delayed CKD progression, imposing dietary change may provoke kick-back, and a reluctance even to consider a different way. Giving a person information allows them to face up to their food habits and decide whether to change.

> ***CYP:*** *Because a person's diet is part of their freedom, isn't it, you can't tell a person exactly what they need to eat… 'You have to have that cereal at breakfast. You want that one, tough luck, you are having that one.' You can't do that.*
>
> ***(INT_03CYP)***

Let me choose (parents) – When considering if the EPSM diet should be prescribed or optional, three models of SM were proposed: the medical model where the HCP direct the plan, giving clear clinical goals; the partnership model where the parents and CYP share the decision with the HCPs, together they come to an agreement regarding the best way forward; and finally the self-agency model where the parents and CYP direct the decisions. The traumatic loss of a twin daughter left 04PAR fearful she may do something wrong. Following prescribed advice meant that 'if it goes wrong it's not my fault'. As her daughter grew, and she gained confidence as a carer she felt more empowered to speak out, working in partnership with HCPs.

> ***Mother:*** *So, I was just like, I don't want to know, 'just do what you need to do, and just tell me what I need, I need to know… I use the first one [medical model]… cos' then if it goes wrong it's not my fault…Do you know what I mean, but then at the same time, now she's getting older, a bit of the second one as well. Cos' I want her to be included…*
>
> ***(INT_04PAR)***

Let me choose (HCP) - For the specialist dialysis nurse, giving individuals' a choice to implement the diet meant giving them power.

> ***Specialist dialysis nurse:*** *If you were giving them the option, it can be empowering for them.*
>
> ***(INT_06HCP)***

5.1.3.8 Family affair

Family affair (CYP) – During the interview participants were asked whether the EPSM diet should solely be directed at the child with CKD. 03CYP believed that by targeting the whole family she would not be singled out as having different needs from others.

> ***CYP:*** *It would be better to encourage the family as a whole, because then I'm not eating differently from everyone else. …I think it'd be better that way, at least, because then I wouldn't feel left out.*
>
> ***(INT_03CYP)***

Reflecting on his experience, 02CYP perceived that following the EPSM diet would be a positive family venture, affirming good eating habits and beliefs, promoting an overall increase in family health. Similar to 03CYP, he maintained it would prevent the CYP with CKD from feeling alone. Moreover, compared to the diet he had to follow, it sounded 'not that bad'.

> ***CYP:*** *The changes don't seem as drastic as ones that I've come to terms with. I think home cooking a meal is good for a family in general, gets the whole family together. It's your own work; it's your own pride. So, I think it'd be very good for the family. I mean if one person doesn't eat fast food then it's generally most people don't eat that fast food, and there's a general health increase overall to the family…. I think it should definitely be followed by as many people as possible, and the patient doesn't feel alone in the diet.*
>
> ***(INT_02CYP)***

Family affair (parents) – 04PAR believed that by making the EPSM diet a family affair it would give a clear message that she [her daughter] is important.

> ***Mother:*** *So doing it together would be [doing] her thing. It's her body, it's her life, it's her…it's not mine, it's not, I can help her as much as I can, but…I think as well for them to feel important too, they need to have that.*
>
> ***(INT_04PAR)***

Once certain dietary changes had become the norm for the whole family, this father attested how siblings were more accepting of change.

> ***Father:*** *Even his little brother don't drink it now, cos' I mean they did used to buy bottles of it [Coke]… he just said 'Alright I'll do it cos' [name of brother] can't have it'.*
>
> ***(INT_06PAR)***

For 03PAR, making the EPSM diet a family affair was an opportunity for hands-on, fun food preparation sessions. Involving the whole family also made the diet inclusive, giving everyone ownership. It especially helped siblings to understand why the diet was important for the child with CKD.

> ***Mother:*** *…that's educating them isn't it. So, I suppose it's making them sort of being, or take ownership of what it is that they're doing and understanding why they're doing it.*
>
> ***(INT_03PAR)***

Family affair (HCP) – 08HCP and 01HCP affirmed the importance of making the EPSM diet for everyone; children with CKD would feel less singled out as different, or worse still punished for having CKD by denying them choice foods. Making changes early also allows for the formation of long-term habits, attaining higher sustainability and success.

> ***Consultant paediatric nephrologist:*** *Actually, this is a healthy lifestyle change for everyone, so it becomes a habit for the whole family, and that's much more likely to be imbedded in the long term.*
>
> ***(INT_08HCP)***
>
> ***Consultant paediatric nephrologist:*** *If you make whole families make changes…it's been much more successful, than if you're just picking out one child, almost penalising them …you know that's not fair, is it? That's not sustainable.*
>
> ***(INT_01HCP)***

CYP learn by example. One of the renal dietitians explained, making the EPSM diet a family affair, provides the child with a great opportunity to learn from others, and as they see the diet in action they may follow suit.

> ***Paediatric renal dietitian:*** *I think if you're expecting a child to change their school of thinking, then they mimic the parents and what's going on at home, other siblings. At the end of the day, reducing processed foods is only going to be healthy for all children.*
> ***(INT_03HCP)***

It made sense for this renal nurse; if you are telling people to restrict lots of foods, that's really hard; but the EPSM diet is healthy and ideal for the whole family.

> ***Specialist renal nurse:*** *Because if you are going to say to a family that they can't ever have cheese, or they can't ever have milk or yogurt, that sounds horrendous. Actually, the thought for me…this is a healthy, good quality meat, plant-based diet, actually let's get the whole family on it.*
>
> ***(INT_02HCP)***

5.1.3.9 Driven by the goal

Driven by the goal (CYP) - Change is not easy, but as 02CYP expressed, if there is a 'good purpose' or a promise of a better future, the change may be worth the gain. While reflecting on his experience, 02CYP allotted food a good or bad status, perceiving the EPSM diet to be 'less of a shock' and the food was 'not all bad' compared to the advanced stage renal diet that he had followed.

> ***CYP:*** *I guess that's still got quite a lot of options, and good food that you can eat, and know that you are eating for a good purpose. It's helping you. I think generally not eating as much processed stuff that would be better in a way.*
>
> ***(INT_02CYP)***

For 04CYP, who was diagnosed with CKD in the past year, receiving dietary phosphate advice during the diagnosis period meant being able to 'keep myself safe'. A tool that provided protection against future complications – delaying the progression of the disease - gave him a clear rationale to keep going. Motivated by this knowledge, he made it his responsibility to adhere to the diet.

> ***CYP:*** *… like I couldn't have a lot of potassium and phosphate, and I had to watch what I was eating, otherwise it would make it worse, and transplant and dialysis would come earlier in life.*
>
> ***(INT_04CYP)***

When 06CYP was diagnosed with established stage 5 CKD aged 14 years, time was of the essence, and dietary change was essential. It was a shock to the whole family to learn the significance of diet and its impact on his health. The diet at CKD 5 is prescriptive and imposing. It was not an option to take it or leave it, but essential to

prevent deterioration. The hardest adjustment for 06CYP was breaking his cola-drinking habit. His voice captured an element of fear; shocked by the severity of the diagnosis, he understood food as a tool that could help him make a difference to his future – imparting strength to rebuild. With the support of his family, he was determined to change.

> ***CYP:*** *I think it was more the drink, cos' with… cola, I have been drinking that for like years, and making a sudden change made me feel like…and I really struggled with… like water, like Sprite, 7UP all them drinks…like as a substitute. I thought the only way you're gonna' get better is by eating more healthy, and not eating junk … then you're gonna' build yourself up, make you more stronger…And obviously being with mum and dad they pushed me to go and do it anyway.*
>
> ***(INT_06CYP)***

Earlier, 02CYP assigned food a good or bad status, here, recognising its impact on the acceleration or delay of renal function he attributes food with toxic or healing properties. Allied to this idea, he reflected on the use of fear as a driver of change in other health conditions, believing it would be 'a good incentive' to motivate action in the early stages of CKD.

> ***CYP:*** *Yeah, I think it works in smoking and stuff, show up some picture of horrible lungs and… yeah if you said this Big Mac burger has got this hormone, and this is gonna' do this to your body, and this is what your kidneys gonna' look like, and this is what could happen, but this can prevent it. I think that would be a good incentive as well to get on with it.*
>
> ***(INT_02CYP)***

Driven by the goal (parents) – Based on her experience and her daughter's current reliance on dialysis - a place 'where you don't want to be' - 03PAR shared how crucial it is for parents new to CKD to recognise and comprehend the rationale for the EPSM diet, in particular its potential to delay disease progression. Recounting her daughter's journey, she cautioned against dialysis, understanding something of its impact on a child's growth and development. She implored the need for information to be given early to ensure they understood how it might help them in the future.

> ***Mother:*** *Because I'm aware that obviously having a young child on dialysis it's not good. Whereas if you can get a child into their early twenties and then they have to have dialysis, maybe because all the growing and all the things that happen as a child, and you've got past that…So it's slowly sort of imparting that information, but relating it to how it will help in the future which I think is hard.*
>
> ***(INT_03PAR)***

Driven by the goal (HCP) – For the HCP 07, 03 and 08, it was important to consider the ethical implications of introducing a new dietary intervention strategy and the balance of benefit versus side-effects as well as potential general health benefits.

> ***Consultant paediatric nephrologist:*** *Early [dietary] advice can do no harm and may do good.*
>
> ***(INT_07HCP)***
>
> ***Paediatric renal dietitian:*** *At the end of the day reducing processed foods is only going to be healthy for all children, so it's not going to have consequences*
>
> ***(INT_03HCP)***
>
> ***Consultant paediatric nephrologist:*** *You know a lot of the changes… are healthy changes, and actually there's a lot of benefits for people's general health. Their cardiovascular health, which we know is something that's important with our society on a whole, but even more so if you got chronic kidney disease.*
>
> ***(INT_08HCP)***

Furthermore, as CKD has no cure, 04HCP and 08HCP believe the preventative message of EPSM is the way forward. Indeed, it is their duty to inform and equip individuals to be proactive. As 08HCP recalls, parents and carers frequently seek ways to delay progression, and as the EPSM diet has a clear rationale to benefit, all families should be encouraged to use it as a SM tool.

> ***Consultant paediatric nephrologist:*** *But the truth is these things [processed foods] are bad for you, and actually I think it's our responsibility to tell families.*
>
> ***(INT_04HCP)***

> ***Consultant paediatric nephrologist:*** *… prevention is better than cure … once you're on dialysis there isn't a cure, transplant is just another treatment…There's clear benefit to preventing rather than treating more advanced kidney disease … It's about prevention, it's about healthy messages …empowering young people, parents and carers to be proactive in doing things to help themselves. A lot do say "can we do anything to make a difference?" and having that thing [diet] is really helpful.*
>
> ***(INT_08HCP)***

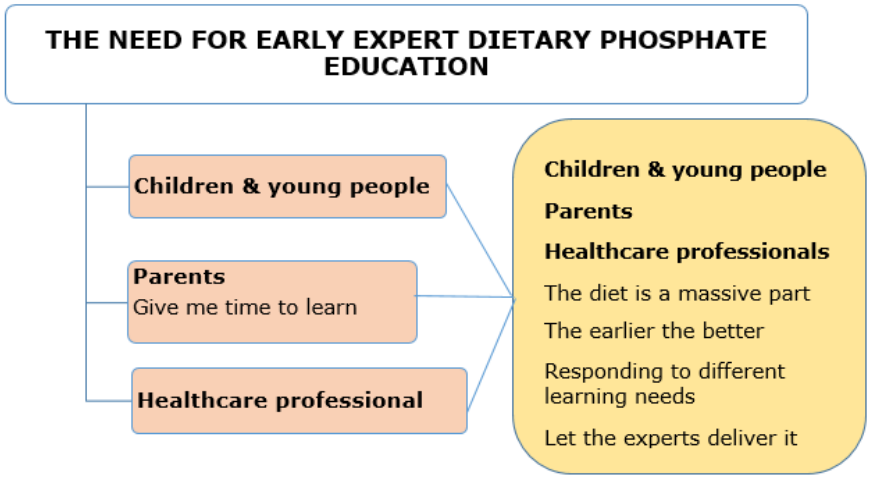
5.1.3.10 Summary of Theme 2

The second theme 'the need to take control whilst establishing a new normal' captured CYPs desire to fit in with their peers, not desiring to be different. Taking control for parents, involved a negotiation of power, through a caring language. The EPSM strategy holds the promise of an empowering tool to ameliorate the trajectory of the disease, enabling HCP to hand over some control, whilst tackling the root of the problem. Recognition of its limitations and possible concerns is essential if the tool is to be accepted and utilised by the family as a whole.

5.1.4 Theme 3: The need for early dietary phosphate education

This third theme recognised stakeholders' desire for an early dietary phosphate education strategy. Namely how, when and who should deliver the EPSM education, and what format would best meets the needs of the people it will serve. The findings of this theme are summarised in Figure 16, p. 158.

Figure 16. Summary of theme 3 findings



5.1.4.1 Give me time to learn

In order to become a lifestyle change new dietary information needs to be provided gradually and in small amounts, with tailored help and encouragement to sustain ongoing compliance. As described by 03PAR, children learn and respond to information better if it include hands-on practical demonstrations, with opportunities to get involved. Over time, as the information is repeated in fresh and engaging ways, the whole family will start to comprehend its significance and embrace its message.

> ***Mother:*** *It needs to be almost like drip feed them [children] a little bit… if they're involved with the actual process and they understand, then they're more likely to probably take on those… If you've got the beauty of having time and I suppose information, and you know sort of supporting them do it, actually having sort of practical kind of input… you can't just educate and expect that to happen just like that. I think people need time and … nurturing maybe.*
>
> ***(INT_03PAR)***

03PAR described, as her daughter's condition changed, the family's needs and views also evolved. HCPs provided a lot of detailed information during the diagnostic period, but with so much going on at the time, such as learning to do dialysis at home, she held on to what she could manage, what she deemed to be important, or felt able to cope with. 03PAR maintained that because information was received,

HCPs should not assume that it is still there. Life pressure fluctuates, and the information she received was often selectively retained or shelved, fading away. She described, how as her perspective changed over time, her willingness to engage with the education increased, so repeated exposure was important to remind her of the message.

> ***Mother:*** *When I think back, I probably missed some of that information about the diet side of things, because there was a lot of other things we were being told…and having to be taught how to do the PD [peritoneal dialysis] at home… Sometimes you almost … filter the information and it gets pushed back… But then I think it's not assuming that all that other information is still there, it needs to be revisited and refreshed and keep revisiting it as well… the circumstances change and actually you change as a person and what you might have held onto in the past, you know, as you become more accepting of what's happened you might then have more willingness to perhaps change things and then you might be a bit more proactive.*
>
> ***(INT_03PAR)***

5.1.4.2 The diet is a massive part

The diet is a massive part (CYP) – When families enter the 'world' of CKD they often do not realise the importance of diet in the management of the disease. Indeed, as discussed in section 1.3.2, children with early CKD do not routinely receive specialist renal dietetic input until their biochemistry suggests the need for intervention, usually at CKD stage 3-4. Crucially, many parents are unaware that specialist paediatric renal dietitians are a key member of the multi-professional renal team.

02CYP, a teenager diagnosed with established CKD when he was 5 years of age, jokingly reflected on the enormity of his diet. 'For the renal diet, I was quite sad', indeed many of my favourite foods were reduced or removed. 'I really liked cheese…chocolate…bananas, and I wasn't allowed much.' However, it was not all bad, 'like not eating so much fruit, which for a 5-year-old is like the greatest thing you can ever hear…and sweets are great!'

The diet is a massive part (parents) – For 03PAR, whose daughter already had complex medical needs requiring tube feeds since birth, the rigour of the renal dietary prescription became quite overwhelming when diagnosed with established CKD stage 4 at 6 years of age. Shocked at the enormity and complexity of the task,

she tried to take stock of what might be involved, but clearly didn't expect the diet to be such a difficult thing; her relief was apparent when she realised that specialist renal dietitians were on hand to support this crucial intervention.

> ***Mother:*** *But that's [diet] probably one of the hardest parts about this whole disease isn't it…I couldn't believe you've got sort of dedicated dietitians and I'm thinking…didn't see that, but obviously… it's such a massive part you've got to have that service. Thank goodness. It's massive and it's complex.*
>
> **(INT_03PAR)**

02PAR had provided around the clock care since her son was born. The diagnosis of established CKD, requiring intravenous protein infusions at home, and haemodialysis to remove the waste products from his body, compounded his already complex needs. As diet was central to his management, she had developed an adept understanding of how dietary change could acutely alter his blood levels. Children receiving haemodialysis have fortnightly blood tests (often more frequently if required), with their diet, medication and dialysis modified accordingly. Waiting for the results of biochemistry can be an anxious time for parents. They worry if they are doing the right thing, and since diet is primarily in their control, there is a sense of duty to get it right. Many parents register online with the PatientView program, which gives them access to their child's blood results from home as soon as they are available. Equipped with dietary knowledge 02PAR had a SM tool that gave her control.

> ***Mother:*** *In the dialysis unit quite often each week or a fortnightly we will get bloods done… the few things that we look for as parents is the phosphate level, and the proteins, the potassium, and they are a massive part of our children. A massive, massive part, yeah…We're lucky because we can access these bloods at home… if you've got an understanding of that, you can change it through food before you go back to have your results… I honestly think dietary is a bigger part as any type of treatment you are gonna' give, it's a massive part.*
>
> ***(INT_02PAR)***

The diet is a massive part (HCP) – One of the four paediatric nephrology consultants interviewed described the dietary prescription as tantamount to the medical and psychosocial prescription. The complexity of CKD means that food,

medication, and the relationships between these facets of life, family, and friends become intertwined, making it difficult to separate one from the other. CKD imposes a high level of change on an already evolving plain, and the CKD journey involves negotiating each of these elements, and many more, in order to navigate each stage of the disease. Problems arise over time and support needs to be put in place to best manage each individual's situation. As an experienced clinician, with a priority to support the growth and development of each child, he elevated dietary advice as paramount in supporting the family.

> ***Consultant paediatric nephrologist:*** *The management of children with chronic kidney disease requires three basic prescriptions…the medical prescription…the dietary prescription and the…psychosocial prescription, and all of them need to be interlinked. One of the most important ways of supporting them is to give them actual dietary advice because children, as opposed to adults, are growing all the time. So, looking at the aspects of growth and development, the dietary aspects are of major importance.*
>
> ***(INT_ 07HCP)***

5.1.4.3 The earlier the better

The earlier the better (CYP) – During the interview participants were asked their views on the best time to deliver the EPSM diet. While recognising the intensity of the diagnosis period, 03CYP and 02CYP defended the need for early dietary advice but felt a measure of 'breathing room' was crucial. Giving too much information too soon was a waste of time, as very little would be taken in.

> ***CYP:*** *I think as soon as they find out they've got CKD… soon after diagnosis. Maybe not straight after diagnosis…you want them to…take in the fact that they've got it, but definitely soon after, they need to be told that 'This diet could help you out…if you want it.'*
>
> ***(INT_03CYP)***
>
> ***CYP:*** *I think it is important to come to terms with the disease…a suitable timeframe should be given before the bombardment of information. Cos' if you get given everything at once you are going to forget something… I think there does have to be some breathing room for the family.*
>
> ***(INT_02CYP)***

The earlier the better (parents) – While confidently believing dietary phosphate advice should be offered as soon as possible after diagnosis, 03PAR gave an honest reflection on the challenges of eating less processed food when you have a sick child. As detailed in section 1.3, reducing inorganic dietary phosphate involves preparing more home-cooked meals. When juggling the demands of life, home-cooking is not always easy or possible. Having the support to change your mind-set and realistically make small changes towards the goal will help you to see the benefits and sustain the change long-term.

> ***Mother:*** *I think it's as early as you are aware that you've got this as a diagnosis. …when you've got a sick child, actually cooking a meal from scratch, and having all that thought, sometimes you just don't have the time… with busy lives…how you can bring it into your normal life without it feeling like a chore, which it actually does, that's the truth.*
>
> ***(INT_03PAR)***

The earlier the better (HCP) – Using an analogy from the emergency services, 07HCP felt dietitians are called upon when the blood levels are high, and the damage has been done. Congruent with the belief that dietetic input needs to be harnessed early, 07HCP emphasised the importance of dietitians getting involved for the long haul. Reactive behaviour addresses a need, but it does not prevent it from happening again.

> ***Consultant paediatric nephrologist:*** *Most dietary advice is given for [CKD] level 5, obviously for those on dialysis, and those approaching dialysis., but in fact we feel that the advice should be given much earlier on…You know, you're calling in a dietitian to firefight…So [the dietitian] isn't really involved in the family long-term, and a firefighting role is no good, you have to be constant…This is a lifelong condition, and that changes, that actually reinforces the need for them to be there [early].*
>
> ***(INT_07HCP)***

As a champion for EPSM, the specialist renal dietitian recognised the importance of journeying with the family from the beginning.

> ***Specialist paediatric renal dietitian:*** *I think if we [dietitians] were … seen as part of it from day one, so if someone presented at birth or … even if they presented at CKD 2 … I think that if you've met the dietitian to start with that's a really good start of working … diet is seen as a key part of things throughout the treatment of kidney disease.*
>
> ***(INT_03HCP)***

For the dialysis nurse, introducing the diet early and getting into that way of thinking would make it easier later on.

> ***Specialist dialysis nurse****: There's a lot of information to get your head around and I think if you start that earlier on … when you get to the latter stages of CKD, that it's…not easier, but it's a more natural way of thinking… And I think the earlier the better.*
>
> ***(INT_06HCP)***

HCPs acknowledged how difficult it was to balance the timing and quantity of information given to parents. This consultant paediatric nephrologist, 01HCP expressed her concerns about giving dietary phosphate advice during the early stages, knowing that it will add to the already immense volume of information. Families need sufficient information to be informed, gradually and in small amounts, but not too much that they feel overwhelmed and alarmed.

> ***Consultant paediatric nephrologist:*** *You've got to gauge it right, because you've got to give parents a lot of information, but not completely scare them, and so it's often drip feeding information over time….It's that balance, of not playing it down, but not frightening them completely, versus giving them all of that information to enable them to have it from the outset. Really, it's a fine line, it's a balance.*
>
> ***(INT_01HCP)***

5.1.4.4 Responding to different learning needs

Responding to different learning needs (CYP) - Educational information needs to be provided in a format that best meets the needs of the people it will serve. As 'everyone's on social media [*and*] got a phone' young people want social media platforms to convey and affirm information. They felt the information should be available 24/7, with a measure of humour, in a snappy and engaging format.

> ***CYP:*** *Having a social media platform for that [EPI information] would be extremely helpful … a YouTube I think would be the most helpful.*
>
> ***(INT_03CYP)***
>
> ***CYP:*** *When I watch…it makes me understand it better…especially …like cartoon videos…they tell ya' the information you need. But it's also a bit funny…you still get the information you need, but you get it in like a better way to catch the attention of the child.*
>
> ***(INT_05CYP)***

Conversely, 06CYP preferred to speak face-to-face with a HCP. This would give chance to really engage with the experts, ask questions, and get to grips with the information.

> ***CYP:*** *I would probably rather have it…from healthcare rather than a booklet, cos' anyone could right get it off the internet…rather get it from a healthcare persons perspective, cos' they know more.*
>
> ***(INT_06CYP)***

Responding to different learning needs (parents) – Busy mums need information to be at hand. For 02PAR that meant a real recipe book with exciting, easy-to-prepare meals that kids would love. Others parents preferred to take an online course and scroll through recipe ideas on their mobile phone. For 01PAR, whose daughter was diagnosed with established CKD aged 6 years, requiring home dialysis, she kept her credit card–sized renal diet guide with her at all times for constant guidance. Dietary information was summarised in a concise, bullet point fashion and the handy format meant that she could take it everywhere with her.

> ***Mother:*** *Even if it's a booklet, an advice booklet, anything, but I genuinely think it does need to be accessible… I don't take stuff off a screen…A recipe book you know for parents, because we all look for like an exciting meal or something to keep our kids going…I think that would be brilliant. Absolutely.*
>
> ***(INT_02PAR)***

> ***Mother:*** *It was a renal course [online]…and at the end of it, I got a recipe book online…. I was dead good. I think the leaflets that were handed to us [at the hospital] were really good… but if I was unsure of anything, I would just refer back to that [credit card traffic light renal diet guide] if I was in the supermarket. That was a quick 'go to' that was on me all of the time, and it's still … on me now, and I'm sure will be forever.*
>
> ***(INT_01PAR)***

Responding to different learning needs (HCP) - Credible, trusted online resources such as the EMEESY network website allowed instant access to multiple resources. However, for 01HCP, to be effective, patient education needs to be regularly followed up, evaluating their understanding and ability to practically apply the information to their everyday lives.

> ***Consultant paediatric nephrologist:*** *They've got to have someone they can contact back, to say '… I didn't quite understand this, can I clarify this'…you need to close that loop and make sure you're following it through … You've got to keep revisiting it.*
>
> ***(INT_01HCP)***

Eager to explore interactive educational ideas, this specialist renal dietitian was teeming with engaging suggestions. Group cooking sessions would provide a plethora of opportunities for families to come together, share experiences, learn from each other, all the while gaining new skills and socialising. Visiting a family in their own home would give insight to the family set-up and cooking facilities, allowing advice to be tailored to their needs. She fervently described how parents want more than just written resource; they want authentic face-to-face input - it instils worth. She described the importance of explaining and translating detailed dietary information into meaningful practical advice.

> ***Specialist paediatric renal dietitian:*** *It would be amazing to be able to do cooking sessions…with a group; because you get people to bounce ideas off each other…but the other way to do it is individual home visits… or supermarket shops. It's hard to envisage what everybody's got available at home for cooking as well … A lot more practical advice would be helpful. I think giving out written information doesn't replace meeting the family, so you could probably get the information on the net… but people don't, because we need it explained…and people need to know that they are valued, and I can't do that by giving them a piece of paper.*
>
> ***(INT_03HCP)***

5.1.4.5 Let the experts deliver it

Let the experts deliver it (CYP) – These young people attested to the role of the specialist paediatric renal dietitian when considering who is best placed to deliver the EPSM education. Appreciating the dietitian's experience and specialist dietary knowledge, 02CYP and 04CYP acknowledged their invaluable role. Additionally, meeting the dietitian and doctor together during the early stage would give a clear message of dietary importance.

> ***CYP:*** *It should be the dietitian…first-hand; people with experience, the experts in the situation should be the ones giving the things.*
>
> ***(INT_02CYP)***
>
> ***CYP:*** *If the dietitian and doctor were in the same room that might be quite helpful. Because then it would tell the people that the dietitian is associated with the doctor… the doctor doesn't really have the sort of insight a dietitian does…They can't obviously go into details like dietitians can.*
>
> ***(INT_04CYP)***

Let the experts deliver it (parents) – Conferring with the young people, 02PAR and 05PAR perceived the dietitian as the best person to provide the EPSM education. Not only because it is their profession and they have a wealth of knowledge on the matter, but the doctor has many other priorities, and diet is not their main focus. Unlike the doctor who is viewed as intense and serious, the dietitian is more approachable, affable and easier to talk to.

> ***Mother:*** *I genuinely think… a dietitian is somebody that parents can approach. A doctor carries the different….do you see what I mean. It should be… that's that person's profession…100%. That's their main… whereas the doctor to me has lots of key areas, diet would not be their main focus.*
>
> ***(INT_02PAR)***
>
> ***Mother:*** *It is better if it came from…a dietitian, because the doctor is very medical, and they'd perhaps don't know as much about it as what you [the dietitian] do…and it might be a bit more relaxed.*
>
> ***(INT_05PAR)***

Let the experts deliver it (HCP) – Affirming this belief, 07HCP referenced the doctor's poor nutritional knowledge, praising the dietitian as unique in their ability to connect with families in a relaxed but practically relevant manner, able to translate the complexities of the dietary message into meaningful everyday language. Speaking of the weight of 'authority' conveyed when advice is delivered by an expert in the field, he elevated the role of the dietitian at this crucial stage in the management of CKD, claiming each family 'deserves' the best.

> ***Consultant paediatric nephrologist:*** *The families deserve it…information always carries much more authority when it comes from people who are experts…In my career in paediatric nephrology, I would say, that the doctors know very little about dietetics, they know some of the principles, but the practical achievement of…the goals is very difficult. You need familial dietary advice…The time is best done by dietitians who know what it's about, and who can assess the families… in a more relaxed way than a nephrologist can, you know in 10-15 minutes in clinic.*
>
> ***(INT_07HCP)***

Upholding their esteemed role, this specialist renal nurse claimed no one could take the place of the paediatric renal dietitian; indeed, that is "the gold standard" for our patients (INT_02HCP). Placing herself in the parent's shoes, she described how receiving advice from the specialist in the field would give confidence and reassurance that the best advice had been given.

> ***Specialist renal nurse:*** *I don't think it will ever replace having a paediatric renal dietitian sitting there…that would be the gold standard…If I was a parent sitting in the clinic and I knew that there was a paediatric renal dietitian there delivering that information and guiding me along the way, then I would be very happy and very confident that I was getting the right specialist professional advice.*
>
> ***(INT_02HCP)***

5.1.4.6 Summary of Theme 3

The third theme 'the need for early dietary phosphate education' captured the CYPs', and parents' desire to receive the EPSM education gradually and in small amounts. From the outset diet needs to be high on the agenda, as it has such an important part to play in the management of CKD. While allowing the family breathing space to come to terms with the chronic diagnosis, the earlier that the diet is introduced, the

better. Information needs to be sufficient to inform, but not too much that it overwhelms, in an accessible format that best meets the needs of the people it will serve. Hands-on cookery workshops, recipe books and online resources were proposed. Finally, if patients are to be given the best possible care, the EPSM diet should be delivered by the expert, the specialist paediatric renal dietitian.

5.2 Summary

This chapter has presented the qualitative findings from Phase 2 of this MM doctoral study. The three stakeholder groups agreed early dietary intervention should be high on the CKD management agenda. Authentic engagement with HCP and other families during the early diagnostic period would provide a lifeline of reassurance and direction; establishing long-term therapeutic relationships, aiding education and support, and averting feelings of isolation. Particularly for CYP, virtual engagements provide control, and instant access to information and support. Multiple platforms should be considered to best meet the needs of the service users i.e. face-to-face and online. Information overload was frequently experienced by parents and CYP during the diagnosis period.

Delivering an EPSM involves HCP empowering CYP and parents to take back some control. CYP and HCPs fear the EPSM could attract undue attention or stigma, and be burdensome. Parents are eagerly seeking credible information to guide their actions. The empowering EPSM strategy needs to bridge managing health, with encouraging individuals to find a new normal in their everyday lives; getting to the root of the problem to change the trajectory of disease. Making a choice, offers power, and targeting the whole family, avoids singling out the CYP with CKD.

Specialist paediatric renal dietitians are a key member of the multi-disciplinary team, carrying a weight of authority in the translation of the complex dietary message into meaningful everyday advice. The dietary prescription, which is tantamount to the medical and psychosocial prescription, needs to be offered during the diagnosis period. Education needs to account for different learning needs, and should be delivered little and often, in a 'drip feed' manner. Hands-on cookery workshops would provide opportunity to share experiences, learn new skills and socialise, with back-up resources such as recipe books and online tools. Too much information can be

overwhelming and cause alarm, but sufficient is required to make an informed choice. Dietary change involves a change in mindset towards a sustainable long-term goal.

Chapter 6 introduces the Delphi quantitative method used in Phase 3 of this MM study.

Chapter 6: Phase 3 – methods for the quantitative dimension

6.1 Introduction

The development of an EPSM strategy was undertaken using a three-phase exploratory, sequential QUALITATIVE-quantitative mixed methods design, graphically represented in Figure 2 (p. 21). This chapter outlines the quantitative method used in Phase 3 – the Delphi Survey (DS).

6.2 The Delphi technique

The name *Delphi* is derived from the Greek Oracle of Delphi who was the mythological seer of the future (Shelton and Creghan, 2015). In order to provide an expert forecast from the viewpoint of the Soviet Union, on the number of A-bombs to drop on US industrial targets to reduce munitions output by a prescribed amount, the Delphi technique was first pioneered in the early 1950s by the RAND Corporation in the United States of America (Dalkey and Helmer, 1963). Its recognition as a tool for reliably gaining consensus from a group of experts has steadily grown in popularity (Keeney, Hasson and McKenna, 2001; Okoli and Pawlowski, 2004); beyond technological forecasting to many areas of research, including education (Phillips et al. 2014), framework planning (Fefer et al. 2016), physical activity (Robinson et al. 2014) and treatment interventions in health care research (Lambert et al. 2019; Jünger et al. 2017). Many important clinical issues such as the development of an EPSM strategy, do not yield to randomised clinical trials, rather they require individuals to draw on their training and personal experience (Balasubramanian and Agarwal, 2012). Delphi provides a formal structured process for facilitating decisions to improve clinical practice (Jünger et al. 2017), thus the Delphi technique was used to attain expert consensus, and explore and expose multiple stakeholder views regarding the development of an EPSM strategy.

6.2.1 Principle features of Delphi

As informed by Mullen (2003), the principle features of a DS involve the research team sending out (postal or online) a structured or unstructured survey to an "expert

panel" (p. 38). In the classical DS, an initial exploratory round seeks to facilitate idea generation to elicit opinion (Keeney et al. 2011). Survey responses are then collated in a qualitative format accompanied by a quantitative rating (e.g. Likert scale) of the survey statements. Considering the comments of the panellist, new, revised or original statements are re-circulated, accompanied by an anonymous summary of participant responses (Keeney et al. 2011). Panellists are invited to confirm or modify their previous responses. This process is repeated for a predetermined number of 'rounds' or 'iterations' until a predetermined criterion such as consensus or level of agreement (LOA) (refer to section 6.4.2) is fulfilled (Keeney et al. 2011).

6.2.2 The Delphi process

Figure 17 provides a flow diagram detailing the EPSM strategy Delphi process.

Figure 17. The Delphi process

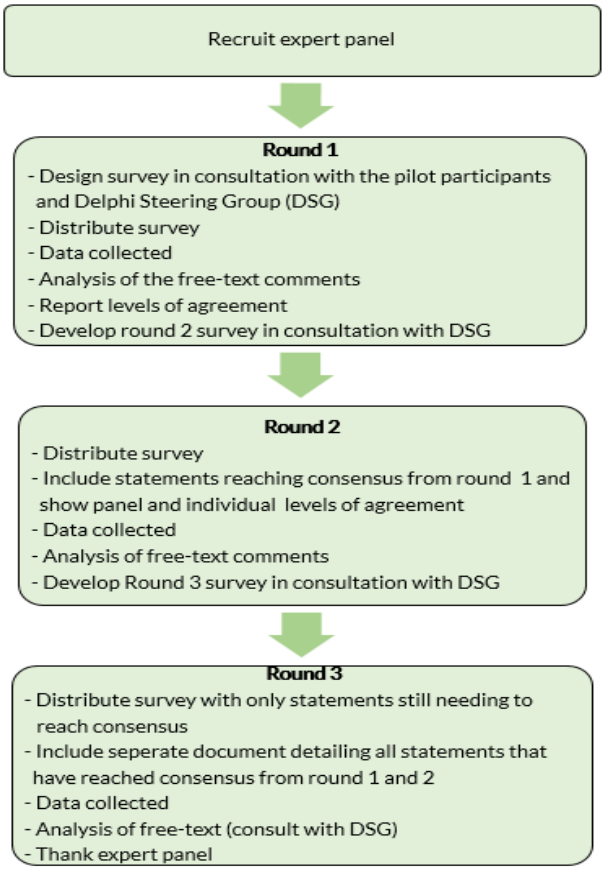


Diagram adapted from Shelton and Creghan (2015)

6.2.3 Defining Delphi

The broad application of Delphi has resulted in its lack of a clear definition. Linstone and Turoff (1975) acknowledged this absence in their seminal text on the Delphi Method, stating many authors who have attempted to define the classical or "true" Delphi have been contradicted by other efficacious users. Embracing the *"many valuable applications of this versatile technique" (*Mullen, 2003, p. 37), Linstone and Turoff (1975, p. 3) claim that, *"in its design and use Delphi is more of an art than a science",* they invite the researcher to creatively 'sculpt' both the DS content and process to meet the requirements of the research question.

Despite this lack of consensus regarding its definition, Linstone and Turoff (1975) offered the following description:

> *"Delphi may be characterised as a method for structuring a group communication process so that the process is effective in allowing a group of individuals, as a whole, to deal with a complex problem."*
>
> *(Linstone and Turoff 1975, p. 3)*

They explained, in order to accomplish this structured communication process:

> *"There is provided some feedback of individual contributions of information and knowledge; some assessment of the group judgment or view; some opportunity for individuals to revise views; and some degree of anonymity for the individual responses."*
>
> *(Linstone and Turoff 1975, p. 3)*

6.3 Conducting and Reporting of Delphi Studies (CREDES)

Guidance on Conducting and Reporting Delphi Studies (CREDES) was originally developed to improve consistency and quality in the conduct and reporting of palliative care studies using the Delphi technique (Jünger et al. 2017). Many disciplines outside of palliative care, such as information systems (Paré et al. 2013), nursing (Hasson et al. 2000) and epidemiology (Diamond et al. 2014; Boulkedid et al. 2011) have called for guidance to enhance the rigour and transparency of Delphi studies' conduct and reporting; the CREDES guidance offers the only tool of this kind

(Jünger et al. 2017). For this reason, CREDES guided the conduct of the Early Phosphate Intervention (EPI) DS, informing for example, the rationale, planning, defining consensus, selection of expert panel and interpretation of results.

6.3.1 Rationale for the choice of the Delphi technique

Justification

Many researchers (Delbecq et al. 1975; Turoff, 1970; Gellatly et al. 2005; Hsu and Sandford, 2007; Jünger et al. 2017) endorse the Delphi technique as a tool to:

- aggregate the judgements of several individuals in order to improve the quality of decision-making on a complex matter to improve clinical practice
- provide group interaction that does not require face-to-face contact, and is therefore not constrained by time, geographical location, finance, affective relationships and professional standing
- provide anonymity of the group respondents
- facilitate equal participation regardless of how assertive or submissive the personalities within the group may be
- gain consensus on a contentious topic from a range of disciplines
- establish the differing positions advocated on a topic, and the principal pro and con arguments for those positions
- exploration and exposure of underlying assumptions or views which leads to different judgements on a topic

In line with this rationale and in order to elevate the expert opinion of parents, placing it on an equal footing as the HCPs without feeling intimidated by them, the Delphi technique was chosen to seek consensus and to establish the different stakeholder positions on the development of an EPSM strategy from a variety of disciplinary perspectives, geographically dispersed across the EMEESY network (see section 1.4.2).

So far, Thorne's inductive analytical ID approach has elicited the CYP's, parents and HCPs' views on the development of an EPSM strategy. Using the co-constructed knowledge produced during Phase 2 as a platform for the round 1 DS, the Delphi

technique's heuristic device facilitated expert parents and HCPs to further explore and negotiate, building consensus in the development of an EPSM strategy, to pragmatically improve clinical practice.

6.4 Planning the Delphi Survey

6.4.1 Modified Delphi

Modification of the 'classical' Delphi (where an initial exploratory round seeks to facilitate idea generation to generate a set of statements) typically involves replacing the exploratory first round with interviews or focus groups (Keeney et al. 2011). The primary advantages of this modification are, (1) it can reduce the overall time demands on busy parents and professionals, (2) which can improve the first round response rate, and (3) provides a solid grounding in previously developed work (Custer et al. 1999). In this MM doctoral study, the first Delphi round was modified, and replaced by mapping the Phase 1 (literature and systematic review findings) and 2 (semi-structured interview with CYP, parents and HCP findings) against the Template for Intervention Description and Replication (TIDieR) checklist (2014) to inform the development of the round 1 statements. The TIDieR checklist (2014), is an official extension of the Consolidated Standards of Reporting Trials (CONSORT) 2010 statement (item 5) and Standard Protocol Items: Recommendations for Interventional Trials (SPIRIT) 2013 statement (item 11), and is promoted by van Vliet et al. (2016) and Yamato et al. (2016) to improve the completeness of reporting and replication of interventions. The mapping process is detailed in section 6.7.7.

6.4.2 Level of agreement

A recognised advantage of the Delphi technique is the achievement of 'level of agreement' or consensus, terms that seek to illustrate the respondents' collective judgments (Keeney et al. 2011; Diamond et al. 2014; Jones and Hunter, 1995). Unlike comparable approaches such as committee meetings or group discussions, Delphi ensures that the opinions of every panellist are represented (Dalkey et al. 1972). Despite its merits, critics have linked the term consensus to a "moveable feast" (Williams and Webb, 1994), with flexibility to adjust the parameters to meet the research design. Goodman (1987, p. 733) argued, in addressing this apparent

adaptability that any agreement on an item, "which may cover a bimodal or flat answer distribution", must support the consistency of the community response over successive rounds. Additionally, Sackman (1974) contended, some 'die hard' panellists with strong outlier views, may force a consensus by conforming to the views of others by the third round in order to avoid another round for everyone. Writing on this dilemma, Linstone stated:

> *"the "tyranny of the majority", sometimes threaten to swamp the single maverick who may actually have a better insight than the rest of the "experts" who all agree with each other".*
>
> *Linstone (1975, p. 567)*

Different criteria can determine consensus, the most common approach involves statistical measures such as a predetermined percentage LOA or the median scores on a rating scale (Keeney, Hasson and McKenna et al. 2011). A reliable method involves assigning a numerical level of consensus *a priori* (Williams and Webb, 1994). The different consensus values offered, ranging from 51% (McKenna, 1994), 75% (Keeney et al. 2006), and up to 100% agreement (Williams and Webb, 1979), requires the researcher to defend the choice.

Although most Delphi surveys seek only to gain consensus on a matter, as Scheibe et al. (1975) contended, in seeking only consensus, many studies fail to make full use of the panellists' contributions. Since exploring and examining the panellists' different views can provide valuable insight to complex issues such as the development of an EPSM strategy, this DS involved a collation of the panellists' comments after each round; informing the development of new, revised or original statements in the subsequent Delphi rounds 2 and 3.

6.5 Study population, sample size and sampling strategy

6.5.1 Study population

Selection of a national panel of experts for the DS reflected the complexity and variety of expertise needed to manage CYP with CKD. In his critique of an 'expert', Sackman (1975) argued against selecting only those who are professionally qualified

or of a high standing, rather an expert defined anyone adequately "informed" on a matter. Cantrill et al. (1996) further clarified this issue, suggesting:

> *"the definition of [an expert] should include any individual with relevant knowledge and experience of a particular topic, including patients and carers".*
>
> *Cantrill* et al. *(1996, p. 69)*

This reinforced the decision to include parents as expert Delphi panellists in this doctoral study. Capturing the CYP's voice in the DS would also have been preferable, but it was not possible due to the study's timeline and the need for preparation to enable CYP to complete the DS (see section 6.5.7 exclusion criteria).

6.5.2 Sample size

Similar to the absence of a clear definition, there is uncertainty about the optimum Delphi panel size. In his review of 100 studies, Diamond (2014) suggested the most common sample size for a representative pool of judgements is 11-25. Delbecq et al. (1975) felt that 10-15 was enough and a group of over 30 generated minimal new information or ideas in their experience - a concept similar to data saturation (Polit and Beck, 2018). Concurring with Diamond (2014) and Delbecq et al. (1975), and accounting for an average attrition rate of 50% (Baruch, 1999), and poor completion due to busy work/home schedules, the sample target size for this doctoral Delphi survey was 30 (Hsu and Sandford, 2007; Goldberg et al. 2016). The goal was to retain a group of 15 panellists throughout the 3 Delphi rounds.

6.5.3 Sampling strategy

Using an 'opt out' strategy, all parents and HCP undertaking a Phase 2 semi-structured interview were invited to participate in the DS through an emailed letter of invitation accompanied by a PIS. A 50% decline rate was expected (Junghans et al. 2005). If insufficient individuals agreed to participate, additional panellists were recruited from the network using a non-randomised purposive sampling method (Keeney, Hasson and McKenna et al. 2011); by virtue of their suitability for the

purpose of the study, thereby meeting the inclusion criteria (see 6.5.6) and the requirements for an expert panellist (see section 6.5.4 below).

6.5.4 Expert panellist

Expert panellists comprised a core group of paediatric nephrologists and paediatricians with an interest in nephrology; renal nurses and paediatric renal dietitians; and parents/carers who manage/self-manage the complex chronic disease CKD. Parent/carer is defined as having legal rights and responsibilities (gov.uk, 2019), or providing a substantial amount of care on a regular basis for a CYP (Carers Act, 1995), respectively.

> This study defined **expert panellists** as any individual identified by the researcher and EMEESY clinicians, as having relevant expert knowledge and experience in managing CYP with CKD.

Focusing the DS around a select group of 'experts' is viewed as the cornerstone of an effective Delphi (Keeney et al. 2011; Stitt-Gohdes and Crew, 2004) and was essential to capture quality 'real world' views and experience on the issues considered to be important to the development and practical delivery of an EPSM strategy (Balasubramanian and Agarwal, 2012).

6.5.5 Participant anonymity

Panellist anonymity from each other, a clear advantage of the Delphi method, offers an equal opportunity for each panellist to contribute and react to ideas unbiased by the identities of other participants (Shelton and Creghan, 2015). It encouraged panellists to be open and honest in their views, psychologically unpressurised by the dominance of others (Keeney et al. 2011), which promised the production of insightful data. By comparison, due to purposive sampling (see 6.5.3), the researcher was not shielded from each participant's response. However, in order to optimise quality in the data analysis process, steps were taken to ensure that each comment carried the same weight and equal importance, reducing the risk of subject bias (see section 9.7) (Keeney et al. 2011).

Reflecting on his own completion of lengthy DS with limited time given to each question, Sackman (1974) offered strong criticism of anonymity. Mindful that respondents are not accountable for their contributions to other panellists, he suggested it could encourage snap judgements and typecast thinking. In defence, Goodman (1987) believed that instantaneous unconsidered responses are less likely to occur when the recruitment of panellists is based on their expert knowledge and experience. There was evidence in this doctoral study of the importance of anonymity among expert panellists, with both parents and HCP regularly contributing free text comments.

6.5.6 Inclusion criteria

The following criteria had to be met by parents/carers and HCP to be recruited to the DS:

- **Parents/carers** had recognised expertise to manage CKD, and must have a child (0-16 years) diagnosed with any stage of CKD >6 months prior to recruitment. The child must attend the tertiary hospital for treatment. The child does not have to have participated in Phase 2 for the parent to be recruited to the DS.
- **HCPs** must have expertise in the management of children with CKD, must work, or previously worked within the EMEESY network.

6.5.7 Exclusion criteria

Parents/carers and HCP were excluded if:

- Not fluent in the English language (both written and verbal). No funding was available to design the DS in different languages
- Not able to meet the above inclusion criteria

The views of the CYP elicited during Phase 2 (semi-structured interviews) informed the design of the round 1 DS (see section 6.7.7). It would have been preferred to include CYP in the DS, however for the following reasons CYP were not recruited to the DS:

- CYP would need a separate email address from parents
- Parental support would be required to assist with understanding and instructions
- Parental support may influence the child's contribution/opinion

Addressing these issues required additional training and time, both of which would have been difficult to facilitate remotely across the EMEESY network geographical spread.

6.5.8 Recruitment

To optimise potential HCP and parental participant reach across the EMEESY network, Delphi adverts were posted on the EMEESY website and parents Facebook page. Alternatively, mailshots were considered, however no funding was available to support this.

To enhance research validity and authenticity, clear selection criteria were devised (refer to inclusion criteria section 6.5.6), supported by the professional backing of the EMEESY clinical staff, to identify potential experts for recruitment (Balasubramanian and Agarwal, 2012). For details on the recruitment process for Phase 3 (Delphi survey), see Figure 11, p. 102. The participants' availability (convenience sample) and word of mouth knowledge of the study (snowballing effect) also factored in the selection process (Polit and Beck, 2018).

Potential participants were encouraged to call or email the researcher with any questions about the study before deciding whether to participate in the DS. Once eligibility status had been checked, the EMEESY Network lead or administrator accessed the medical notes and sent out a letter of invitation and PIS. Interested participants were asked to send the researcher an email to register their interest to be recruited to the DS.

6.6 Research setting

As described in section 1.4.2, the children's kidney network for the East Midlands, East of England, and South Yorkshire (EMEESY) comprises 20 local hospitals across the region, with parents and HCPs spanning a geographical area between 0-150 miles from the tertiary centre. The online Delphi survey collated this disparate group of experts' subjective opinions (Balasubramanian and Agarwal, 2012). Delbecq et al. (1975) favoured a heterogeneous group, such as the varied stakeholder group recruited to this doctoral DS, believing it more likely to produce larger proportions of quality judgements. Table 33, p. 205 details the regions represented by panellists in the DS.

6.7 Tool development

6.7.1 Demographic questionnaire

Demographic variables as used in most surveys (Saris and Gallhofer, 2014) were collected as part of the Delphi round 1. Collection of socio-demographic data allowed the researcher to identify important dimensions of heterogeneity (age, gender, primary role, geographic location of service provision, and years of experience and specialist group membership) (Sauermann and Roach, 2013). The demographic questionnaire (see Appendix 13) development was informed by Keeney et al. (2011).

6.7.2 The level of agreement used

The 'level of agreement' (LOA) used in this DS, set *a priori* at ≥70%, was a pragmatic decision that was felt to be realistically attainable when seeking the opinions of individuals from a variety of disciplines and backgrounds while maintaining 'credibility' in the Delphi literature (Robinson et al. 2014; Diamond et al. 2014; Goldberg et al. 2016). Thus, all statements in round 1 reaching ≥70% LOA were fed back to panellists in round 2 indicating that no further action was required.

6.7.3 Statements that did not reach agreement

After round 1 and 2, the panellist free-text comments were collated and all statements that failed to reach agreement were reviewed alongside these comments.

In light of comments, new, revised or the original statements were re-circulated to the panel. An anonymous summary of the panels overall LOA was provided for each statement, alongside the individual panellist's rating. Panellists were informed that their comments had been consulted, but in order to limit the amount of material circulated and the time required to read it, round 2 and 3 did not disseminate the summary of the free text comments. Refer to section 7.6.7 (Table 49, p. 251) for statement that did not reach consensus.

Upon receipt of round 2 and 3, panellists were invited to indicate their LOA on all new statements and review all original and revised statements that had not reached agreement in the previous round, confirming or modifying their previous responses. For pragmatic reasons, concerning demands on the panellists' time and the timeline of this PhD study, this iterative process agreed at the outset of the study consisted of a maximum of three rounds (see section 6.7.13).

Affirming the belief that achieving the LOA agreed *a priori* does not imply reaching a 'correct ' judgment (Jünger et al. 2017), any statements that did not reach agreement after round 3 provided some informative insights, highlighting different stakeholder perspectives (see section 8.2.2).

6.7.4 Development of a Delphi Steering Group

As informed by Slade et al. (2016) a Delphi Steering Group was formed to provide expert guidance throughout the Delphi process. Steering Group members were approached for their ability to meet one or both of the qualifications detailed in Box 6, p. 182. Correspondence was facilitated by email or telephone calls due to members' location.

The group consisted of members from a number of specialist professional groups (the European Society for Paediatric Nephrology, the British Psychological Society, the Royal College of Nursing, the Paediatric Renal Dietitians' Interest Group, the British Dietetic Association, and the Health and Care Professions Council), providing a varied expert viewpoint, with a balance of perspectives.

Box 6. Delphi Steering Group

Delphi Steering Group
Qualifications • Possess expert experience in the care of CYP with CKD or chronic disease OR • Previous expertise in the design and development of a Delphi survey
Responsibilities • Provide expert guidance regarding: - The rewording of statements - Timing between rounds - Delphi survey design and methodology issues - Review survey data prior to release to the panel
Delphi Steering Group members • Parent of a child with early stage CKD • Global expert in the Delphi Technique (University of Stirling) • Clinical psychologist with expertise in the Delphi Technique (University of Nottingham) • Consultant paediatric nephrologist/supervisor (Nottingham Children's Hospital) • Paediatric renal dietitian (Edinburgh Hospital for Sick Children) and Chair of the Paediatric Renal Dietitians Interest Group (PRiNG) • Chronic diseases nurse with expertise in early intervention (University of Nottingham)

6.7.5 Designing the Delphi survey

Delphi Decision Aid (Armstrong, 2003) and Bristol Online Surveys (BOS) were explored as initial design options, supported by the Health e-Learning and Media (HELM) team at the host university. After sending an initial draft of the Delphi tool to the supervisory team, feedback indicated some challenges with the software: formatting issues, limited functions for creating a Likert scale or extrapolate data from one round to the next. Training/software expertise was sought, but due to financial costs the researcher decided to use Word Developer™ to design the Delphi survey, converting it into a fillable questionnaire by NUANCE pdf converter enterprise 8.0 software™, which allowed the development of a bespoke DS.

6.7.6 Instructions for use

Delphi round 1 included instructions to the expert panel on how to complete the survey (Keeney et al. 2011). The estimated time to complete, aim of the survey and an explanation of the 4 levels of agreement were detailed. A hypothetical example statement was given (see Appendix 13) with stepwise instructions on how to respond, with opportunity for additional comments or suggestions after each statement/group of statements, and at the end of the survey.

6.7.7 Preparing the statements for the round 1 Delphi survey

The modified Delphi method (see section 6.4.1) expedited the investigative process of the classical exploratory first round, and allowed the researcher to take control over the range and scope of the issues to be discussed and refined in the Delphi statements. The TIDieR checklist (see section 6.4.1) provided a framework for designing the round 1 DS, identifying the key domains to address when describing and developing an intervention (see Box 7).

Box 7. Adapted Hoffman's TIDieR (2014) checklist items that informed round 1 DS categories

Item 1. **Brief name:** Provide a definition for the EPI

Item 2. **Why:** Provide a rationale for the development of the EPI

Item 3. **What (materials):** Describe the physical or informational materials needed to deliver the EPI

Item 4. **What (procedures):** Describe the procedures/activities/support needed to effectively deliver the EPI

Item 5. **Who:** Describe who should provide the EPI and the intended users

Item 6: **How:** Describe the modes of delivery for the EPI

Item 7: **Where:** Describe the type of location(s) where the EPI should be delivered

Item 8: **When and how much:** Describe when the EPI should be delivered, and over what period of time

Item 9: **Tailoring:** Decide if the EPI will be tailored to the individuals needs

Additional categories informed by the TIDieR checklist:

Considerations: Describe issues that need to be considered in the development of an EPI

Potential concerns: Describe any potential concerns with the development of an EPI

The Phase 1 and 2 qualitative data were analysed using interpretive description (Thorne, 2008) and mapped against the TIDieR framework (see Appendix 14). Interpretive description highlighted the clinical focus and stakeholder issues as important in improving clinical practice (Thorne, 2007; Cooper and Endacott, 2007).

By mapping the Phase 2 data against the TIDieR framework, low levels of abstraction and verbatim phrasing ensured that the round 1 statements remained true to the comments and views of stakeholder's, allowing Delphi panellists participating in Phase 2 to recognise their contributions (Keeney et al. 2011). Further integration of data throughout this sequential MM study (see section 2.4.2) meant that additional statements were constructed to capture the key findings and recommendations from the Phase 1 systematic review. Informed by Thorne's ID (2008), the following steps were taken to analyse the data and develop the round 1 statements:

1. The interpretive data analysis (see section 4.6.2) records filed in NVivo 12, and tabulated as a Word document were reread, allowing familiarisation with the data through 'immersion' (Thorne, 2008)
2. The broad codes and 'group signifiers' already used to capture thematic similarities provided a focus for mapping the data against the TIDieR checklist items (see Box 7, p. 183).
3. The researcher reread the thematic analysis and narrative synthesis of the systematic review (see section 3.11) to capture key messages from the literature that also mapped against the TIDieR checklist.
4. The researcher reread the data and analysed the similarities across the interview transcripts to develop statements for each checklist item capturing the views of the interviewees.

As argued by Keeney et al. (2011), the development of statements using Phase 1 and 2 data analysis extraction and review posed a threat to the internal validity of the data due to potential researcher bias. To minimise this effect the researcher provided a trail for decision-making by tabulating memos to describe and explain the development process, arguably improving the research procedural auditability, confirmability and trustworthiness (Keeney et al. 2011). All statements were checked by the Delphi Steering Group and supervisory team before use. The number of statements reflects the need to capture the breadth of the topic, while not being too onerous for panellists to complete (Keeney et al. 2011). Delphi surveys vary significantly in the number of included statements, from 46-86 (Goldberg et al. 2016; Robinson et al. 2014; Lambert et al. 2019).

6.7.8 A 4–point Likert scale

The Likert scale, the most frequently used scale in survey research was used in the Delphi to help panellists indicate their LOA with a statement (Keeney et al. 2011). A 4-point Likert scale, using graduations of agreement, with the avoidance of a midpoint, was selected for the Delphi survey (Shelton and Creghan, 2015; Rayens and Hahn, 2000). Offering a midpoint allows panellists to indicate neutrality or ambivalence, often argued as an easy or more 'comfortable' way out, leaving panellists the opportunity to avoid thinking about the issue (Nunnally, 1967). Following this line of reasoning Klopfer and Madden (1980) believe omitting the midpoint can increase data quality. Therefore 5 point and 7 point scales were rejected. The Likert items in this doctoral DS corresponded to the following responses:

4 Very high agreement – I completely agree that the *statement should be used* to support the development of an Early dietary Phosphate Intervention strategy

3 High agreement – I agree, but the statement requires some *small* changes before it should be used to support the development of an Early dietary Phosphate Intervention strategy

2 Moderate agreement – I partly agree, but the statement requires *significant changes* before it should be used to support the development of an Early dietary Phosphate Intervention strategy

1 Low agreement – I feel that this statement *should not* be used to support the development of an Early dietary Phosphate Intervention strategy

Labelling all 4-response categories as opposed to just the endpoints (see Box 8, p. 186) can increase reliability and reduce inherent Likert subjectivity (Weng, 2004). Greenleaf (1992) contends this increase in reliability may be due in part to bias in response style, whereby respondents have a greater desire to show agreement than disagreement (McClendon, 1991). Fully labelling, however, reduces cognitive difficulty and achieves more clarity, which can reduce this response style bias (Weijters et al. 2009). Hutsjo et al. (2011) similarly provided a fully labelled 4-point scale in their DS. This practice was adopted for this study.

Box 8. An example of labelling all 4-response categories on the Likert scale

Domain:	Statement - why design an EPI?		Very high agreement	High agreement	Moderate agreement	Low agreement
Rationale	5.	'Junk foods' and processed foodstuffs are viewed as problem foods to be limited in the management of a chronic disease				
How the panel responded in round 1			46.5%	35.7%	17.8%	
Your round 2 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after round 1.						
Rationale	5.	Processed foods can be high in salt and contain added phosphates; to support the management of many chronic diseases, advice should be given to consume less processed foods, with suggestions regarding nutrient-rich alternatives.	☐	☐	☐	☐

6.7.9 Piloting the round 1 Delphi Survey

To establish content validity (Shelton and Creghan, 2015) the round 1 survey instrument was pilot-tested to ensure that any issues regarding wording, ease of completion and completion time, design and layout, software difficulties, and instruction adequacy were addressed and rectified (Bryman, 2012) before the first round delivery to the panel of experts. The eight individuals who agreed to pilot the round 1 survey comprised: two nurses, a health educator, a paediatric dietitian, three parents of children on dialysis and a dietetic support worker. Feedback collated from the pilot survey participants led to changes in the wording of the demographic questions and the phrasing of certain statements. One respondent did not like the term 'burden'. However, as the intention was to remain true to the words and phrases used by respondents in the Phase 2 interviews (Keeney et al. 2011), it was agreed that the term should remain in the survey. Another respondent commented on the amount of

information provided and the crowded nature of the survey. This led to a change in the formatting and font size - factors recognised to influence panellists' response (Dillman et al. 2009). A further comment was made regarding the need for greater clarity in the instructions. As a result, the instruction section was revised making it clearer and more concise. The Delphi Steering Group also reviewed the round 1 DS and subsequent rounds.

Despite undertaking the pilot survey, new challenges arose during round 1. The NUANCE software generated some formatting issues, such that some panellists accessing the survey using Word's older software editions encountered problems opening, completing and saving the survey. Other panellists experienced access issues owing to firewalls and filters on their work computer, a recognised but challenging problem (Garson, 2014). After many amendments, the survey was reformatted and the document was saved as a Windows 2016 version, making it more compatible with older versions of Word. A version using Word 97-2003 was also prepared for those who continued to experience problems. This version was also Media Access Control (MAC) compatible for those accessing the DS on a Macintosh computer. Although the different DS versions could be opened on a mobile device, a mobile platform could not be used to complete the survey. Time and financial constraints meant that software access was not available to develop a mobile compatible survey. Additional instructions to help the panellists complete and save the different versions of the DS were provided.

6.7.10 Postal survey

Panellists continuing to experience problems accessing the DS and those not technically literate (Garson, 2014) had the option of receiving the surveys by mail in a stamped self-addressed envelope. Five participants requested postal surveys.

6.7.11 Why use an online Delphi survey?

The internet is a successful area of survey research, with readily available technology for creating and conducting an online survey (Shelton and Creghan, 2015). Delphi surveys and correspondence can be delivered to panellists instantly regardless of

their geographical location. One of the biggest advantages is the shorter response times, 5.59 days on average, compared to the 12.21 day average for mail surveys (Sheehan and McMillan, 1999). Other advantages include: cost savings, and if the survey is received at an inconvenient time, completion can be postponed until they have time, with the option to mull over the statements, cerebrating the response of the group, and coming back to the survey several times before returning it (Shelton and Creghan, 2015).

6.7.12 The best day to send out the survey

Despite finding no significant differences in response rates to online surveys across days of the week, Sauermann and Roach (2013) acknowledged that some individuals face high work demands and low motivation on a Monday, while on a Tuesday part-time workers may experience the same demands. Survey researchers can use a "dynamic strategy" to exploit this heterogeneity, which varies the days between the initial contact and subsequent reminders (Sauermann and Roach, 2013, p. 276). Such a dynamic strategy attempts to appeal to different segments of the survey population in each round and exploits the fact that only one response is needed from each person for each round. Sending a reminder after 7 days can increase response rates (Sauermann and Roach, 2013). Therefore, sending the initial invitation on a Monday but the reminder the following Tuesday increases the likelihood that a person will respond positively to at least one of the contacts (Sauermann and Roach, 2013). The EPI DS adopted this dynamic strategy.

6.7.13 Number of Delphi rounds

There is no agreement on the number of rounds that should be undertaken in Delphi surveys. Since the Delphi's aim is to reach consensus on a topic, the process can continue until consensus is determined to have been achieved (Balasubramanian and Agarwal, 2012). In the systematic review by Jünger et al. (2017), the number of rounds ranged from 1-5, with the majority reporting 2-3. Ludwig (1994), Custer et al. (1999) and, Linstone and Turoff (2002) suggest that three rounds are often enough to gather the information necessary and reach consensus. Three rounds have been used in many recent health science Delphi surveys (Goldberg et al. 2016; Lambert et al. 2019;

Mendes and da Silva, 2013). Therefore, a 3-round Delphi survey was planned for this study, with the option to close the survey earlier if consensus was reached on all of the statements.

6.8 Data collection and analyses

6.8.1 Round 1 data collection

On the 3rd October 2018 the Delphi round 1 survey was sent out by email. In order to optimise engagement and minimise attrition the covering email contained one attachment, which comprised the following:

- An outline of the Delphi process, the main objectives and the panel's commitment to participate over 2-3 rounds, with feedback after each round (Keeney et al. 2011).
- An invitation to give written consent by marking an ☒ in the box (Keeney et al. 2011).
- A demographic questionnaire containing six multiple choice questions requesting information regarding gender, age group, primary role with CYP with CKD, location of work/residence, years of experience and membership of any specialist groups (Keeney et al. 2011).
- Instructions on how to complete the Delphi round 1 survey, including an example using fictitious data that did not reflect the panel's responses (Keeney et al. 2011).
- A copy of the Delphi round 1 survey containing 48 statements (see section 7.2), with space for free-text comments after each statement which increased as panellists typed so as not to adversely influence the amount of information provided by respondents (Shelton and Creghan, 2015). A table to capture additional comments/statements was included at the end the survey (see Appendix 13).

Providing opportunity for panellists to comment after every statement sought to harness the 'intellectual apparatus that makes them experts', increasing engagement as they see their comments as part of subsequent rounds (Mitchell, 1991, p. 344).

The covering email thanked the panellists for agreeing to participate, provided a summary of the Delphi survey, an approximation of the time it should take to complete, and the planned two-week closing date for round 1. Panellists were encouraged to contact the researcher regarding any concerns, and Mac users were encouraged to request a Mac-compatible version.

In response to each survey returned by the panellists, an acknowledgement email was sent out. Seeking to provide a 'personal touch' (McKenna, 1994a), the panellists who had not yet responded received a reminder email at 7 and 12 days (Keeney et al. 2011), detailing the response rate to date and the importance of their contribution to the research process (Sandrey and Bulger, 2008). The value of their involvement was affirmed by an encouraging 'personal touch' voice message for two panellists, with a reminder to participate. As it may be considered potentially coercive, the researcher decided to discontinue this approach (Keeney et al. 2011).

As the agreed deadline approached, the panellists' response rate was calculated, and a decision was taken on an extension of the closing date to optimise the survey's take-up. For each round, a response rate of 70% was sought, as this has been suggested to maintain rigor (Keeney et al. 2011; Sumsion, 1998) (see Figure 18, p. 206). Table 31, p. 191 details a schedule of the activities undertaken during round 1.

Table 31. Schedule for Delphi

Activity	Start Date	End Date	Duration (days)
Prepared Delphi round 1 survey	03.08.18	03.10.18	61
Prepared covering email	24.09.18	26.09.18	1
Delphi round 1 emailed to expert panel	03.10.18	20.10.18	18
Email sent to panel regarding saving the survey	05.10.18	05.10.18	1
Reminder email sent	10.10.18	10.10.18	1
Reminder email sent	15.10.18	15.10.18	1
Delphi round 1 extended	17.10.18	20.10.18	3
Delphi round 1 closed online	**03.10.18**	**20.10.18**	**18**
Final postal survey arrived	25.10.18	25.10.18	1
Analysed data from round 1	3.10.18	29.10.18	27
Prepared Delphi round 2 survey	25.10.18	29.10.18	4
Prepared covering email for round 2	29.10.18	29.10.18	1
Delphi round 1 analysis emailed/discussed with Delphi Steering Group. Amended round Survey accordingly	2.11.18	13.11.18	12
Email panelists an update on Delphi Survey progress	2.11.18	2.11.18	1
Delphi round 2 emailed to expert panel reporting consensus statements	13.11.18	1.12.18	19
Reminder email sent	23.11.18	23.11.18	1
Reminder email sent	27.11.18	27.11.18	1
Deadline extended	27.11.18	1.12.18	4
Delphi round 2 closed online	13.11.18	1.12.18	19
Analysed data from Round 2 and met with Delphi Steering Group	13.10.18	2.12.18	21
Prepared Delphi round 3	4.12.18	5.12.18	2
Delphi round 3 emailed to expert panel listing only statements not yet reached consensus	5.12.18	19.12.18	15
Reminder email sent	12.12.18	12.12.18	1
Delphi round 3 closed online	**19.12.18**	**19.12.18**	**1**
Final survey returned in the post	11.01.19	11.01.19	1
Analysed data from round 3 with Delphi Steering group	5.12.18	11.1.19	38
Total days from sending out round 1 to analysing data from the final survey returned by post	03.10.18	11.1.19	101

6.8.2 Round 1 data analyses

The data obtained from the demographic questions were tabulated, uploaded to IBM SPSS™ Statistic version 25, with initial cleaning of the data prior to the calculation of descriptive statistics (percentages, median, modal and ranges) (Keeney et al. 2011) to summarise the panellists' demographic characteristics.

An Excel spreadsheet™ was used to collate the data from each returned survey. In order to allow a clear comparison, the Excel spreadsheet listed each Delphi round statement alongside the other rounds. Table 32 provides a section of the Excel spreadsheet™ detailing the responses to statements 11-14. Statements were labelled with an S, followed by the round e.g. S12/1 relates to Statement 12 round 1. Blue (round 1), green (round 2) and yellow (round 3) colour coding was used to allow the researcher to easily recognise in which round each statement reached consensus. Analysing the surveys as they came in helped to manage the data efficiently. Strong administrative skills were needed throughout the Delphi process to develop a coding system for tracking the responses of panellists' in multiple rounds, creating a file for the free-text responses and creating emailing databases (Keeney et al. 2001; Sandrey and Bulger, 2008).

Table 32. Part of an Excel spreadsheet detailing Delphi round 1-3 results for the statements for 10-14

S10	S11	S12/1	S12/2	S12/3	S13/1	S13/2	S14/1	S14/2
4	4	4	4	4	4	4	4	4
3	3	3			4		3	
4	4	3	4	4	3	4	3	4
4	4	4	4	3	4	4	4	4
4	4	3			4		3	
4	3	4	4	4	2	4	4	4
4	4	3	3	2	4	4	4	4
4	4	2			2		3	
4	4	4	4	1	4	4	4	4
4	4	3	3	4	3	4	4	4
4	4	3	3	4	4	4	4	4
4	4	3	3	4	3	4	4	4
3	3	3	4	4	2	4	3	4
4	4	4	4	4	4	4	4	4
3	3	2	3	3	2	3	2	3
4	4	4	4	3	3	4	4	4
4	4	4			4		4	
4	4	2			4		4	
4	4	4	4	4	4	4	3	4
4	4	4			3		4	
4	4	3	3		4	4	4	4
4	3	3	3	4	3	4	2	2
4	4	4	4	4	4	4	4	4
4	4	4	4	4	4	4	4	4
4	4	4	4	4	4	4	4	4
4	4	4	4	4	3	4	3	3
4	4	4	4	2	3	4	3	4
4	4	3	4	3	4	4	4	4
25/28	23/28	14/28	15/22	14/21	16/28	21/22	18/28	19/22
yes	yes	no	no	no	no	yes	no	yes
89.20%	82.10%	50.00%	68.20%	67.00%	57.10%	95.40%	64.20%	86.30%

Using descriptive statistics, each statement was analysed individually. Likert scales could not be considered to be uniformly incremental and therefore the data could not be considered to be normally-distributed, so has been described in terms of median and range (Stevens, 1946; Jamieson, 2004).

For statements that did not reach consensus, Fisher's Exact test was used to calculate the probability (two tailed) using a 2x2 contingency table (StataCorp/SE15.1, 2017™) that there was no association between the parents and HCP ratings within each round. The responses were recoded into the ordinal groups of '1' (very high agreement) and '0' (combination of low, medium and high agreement). This test was used instead of the Chi square test because the numbers were relatively small and occasionally the expected frequencies in some cells were < 5 (Petrie and Sabin, 2009). The level of significance was set at $p < 0.05$ (Petrie and Sabin, 2009).
For statements that retained the same wording across round 1 and 2, an interrater reliability was measured using Cohen's weighted Kappa (Landis and Koch, 1977) to examine the stability of rater (HCP and parents) responses across two rounds. A weighted Kappa was calculated for the ordinal data to take into account the extent to which raters disagree (the non-diagonal frequencies) as well as the frequencies of agreement (along the diagonal) (Petrie and Sabin, 2009). Cohen's Kappa ranges from 0 to 1.0, where 1 defines perfect agreement, and almost perfect reliability, and values less than or near zero suggest that agreement is attributed to chance alone (Landis and Koch, 1977).

Once the DS round 1 closed, levels of agreement were calculated, and a separate file was created titled 'statements that have reached consensus'. Thematic analysis, valued for its highly inductive and flexible approach, was applied in the analysis of the Delphi free-text comments (Clarke and Braun, 2013; Braun and Clarke, 2006; Keeney et al. 2011). Content analysis was considered as an alternative approach, however due to its systematic rule-based procedure, often guided by theory (Hsieh and Shannon, 2005), it was felt not to be appropriate in this study.

The following steps were taken to thematically analyse the free-text comments:

1. Free text comments were collated and tabulated in a Word document.

2. Statements with the same meaning were grouped together using an abstract colour coding system, allowing the researcher to easily identify related comments.

3. Themes were developed around comments with similar meanings.

4. The researcher made decisions on the selection of a comment that seemed to incorporate other panellists' views, or the collapse of similar statements into one that captured the overall view. This approach sought to capture the nuances of participants' voice.

5. This decision was shared anonymously with the Delphi Steering Group and supervisory team to ensure that the collapse process did not change the meaning of any of the statements.

6. To develop new, revised or original statements for the round 2 survey, each statement from round 1 that had not yet reached consensus was reviewed using the themes from the free-text comments. Where disparate views were expressed, these were often presented as revised or multiple new statements to replace the previous one. The Delphi Steering Group supported this process.

7. New or revised statements sought to keep the wording true to the expert panel's comments (Keeney et al. 2011).

8. The researcher made a decision about the unique comments or 'outsider' views that were not similar to other panellists' views, whether to include or omit. In order to be impartial and provide trustworthiness in the analysis process members of the Delphi Steering Group supported this process (Thangaratinam & Redman, 2005).

9. The researcher kept a reflective journal throughout the Delphi survey to capture personal thoughts and perceptions in order to increase trustworthiness (Shelton & Creghan, 2015)

After reviewing all the free-text comments and statements, the final list of statements was mapped against the DS categories as informed by the TIDieR checklist (2014) (see Box 7, p. 183). This provided a clear framework for organising the round 2 survey.

6.8.3 Feedback to the panellists

The panellists received feedback after each Delphi round (Keeney et al. 2011; Sandrey and Bulger, 2008). Due to the panellists' (parents and multiple HCPs) perceived limited statistical knowledge (Engels and Kennedy, 2007), a pragmatic decision was made to only feedback the overall percentage levels of agreement for each statement in addition to their individual response to each statement (see section 6.7.3). Box 9, p. 195 provides an example of the information provided to panellists in the round 2 survey.

Box 9. Example of information provided to panellists

Domain:	Statement - why design an EPI?		Very high agreement	High agreement	Moderate agreement	Low agreement
Rationale	5.	'Junk foods' and processed foodstuffs are viewed as problem foods to be limited in the management of a chronic disease				
How **the panel** responded in round 1			46.5%	35.7%	17.8%	
How **you** responded in round 1					✓	
Your round 2 response (to select your response please click inside one box. Please note you do not have to change your response). **Please note this statement has been revised after round 1.**						
Rationale	5.	Processed foods can be high in salt and contain added phosphates; to support the management of many chronic diseases, advice should be given to consume less processed foods, with suggestions regarding nutrient-rich alternatives.	☐	☐	☐	☐
Comment:						

Round 2 included all 48 round 1 statements and 11 new statements (a total of 59 statements). Statements reaching consensus (16 out of 48) in round 1 were clearly marked with a red shaded box, indicating that no further response was necessary (see Box 10), leaving 43 statements to be rated by the panel in round 2. The inclusion of all of the statements gave panellists an understanding of the 'bigger picture', and a clear view of how their involvement was effectively moving the consensus process forward.

Box 10. An example of a statement that had reached consensus requiring no further response in the round 2 survey

Domain:	Statement – why design an EPI?	Very high agreement	High agreement	Moderate agreement	Low agreement
Rationale	6. The provision of early dietary education gives children, young people and families a tool to more confidently manage their own diet during the early stages of a chronic disease				
How **the panel** responded in round 1		75%	21.4%	3.5%	
How **you** responded in round 1			✓		
This statement reached consensus in round 1 (no further response is required).					

6.8.4 Round 2 data collection

All panellists responding to round 1 received round 2 DS by email/post as requested. The covering email and attachment included similar information to round 1 (see section 6.8.1), excluding the invitation to consent and demographic questionnaire. It became apparent that some instructions in round 2 had been misunderstood. When more than one new statement was presented, panellists were advised 'to select your response please click inside one box' (see Box 11, p. 197). Panellists responded by marking only one of the new statements, as opposed to ticking

one box for each new statement. An email was sent to the relevant panellists (n=2) requesting that any unfilled boxes be completed. All panellists still to respond received an email clarifying the need to provide a response for each statement.

Box 11. An example of a misunderstood instruction

Your round 2 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after round 1 into 2 new statements.						
Rationale	7a.	When a child is diagnosed with a chronic disease, knowing and understanding the impact of diet on the disease may motivate families to make diet/lifestyle changes	☐	☐	☐	☐
Rationale	7b.	During the early stages of a chronic disease, diet/lifestyle change can provide a positive focus for the family, knowing that they are making an important contribution to disease management.	☐	☐	☐	☐

6.8.5 Round 2 data analysis

Similar to round 1, round 2 data was entered in the Excel spreadsheet alongside the previous round to allow for a clear comparison (see Table 32, p. 192). Each statement was individually analysed with the calculation of descriptive statistics to determine which statements required further review for consensus in round 3.

Once round 2 closed, levels of agreement were calculated and statements reaching consensus were highlighted and inserted into the separate file. Analysis of the free-text comments followed the stages outlined in section 6.8.2. As the Christmas holiday season approached, an especially busy time for HCPs and parents, there was feeling panellists were displaying a level of 'response exhaustion' after round 2 (McKenna, 1994b). In view of this, and the progress achieved so far, it was necessary to make a decision whether to close the Delphi at round 2, or push out the remaining 10 statements in a final Delphi round 3. The Delphi Steering Group and supervisory team agreed for a round 3.

6.8.6 Round 3 data collection

To reduce the size of the round 3 survey and the corresponding time required for completion – optimising the retention of panellists and to encourage a rapid response time, Green et al. (1999) asserted that researchers should consider taking steps to impose analytical closure. Rather than the 'inclusive' approach taken in round 2, in consultation with the Delphi Steering Group the researcher made the pragmatic decision to adopt a reductionist approach (Green et al. 1999) in the final round DS, including only those statements that needed to reach consensus. Other researchers (Goldberg et al. 2016; Keeney, 2006) report this approach. The covering email emphasised the reduced size of the DS, the anticipated 5 minute completion time, instructions on saving the survey and the 2-week closure date. As feedback, an attachment detailed the 'Statements reaching consensus'.

6.8.7 Round 3 data analysis

The round 3 quantitative data was entered into the Excel spreadsheet™. After cleaning the data, identifying any missing or incorrect values, the data were analysed using descriptive statistics. The LOA remained at ≥70% during round 3. Statements reaching consensus were entered into the final consensus statements document. The free-text comments were tabulated and the data were grouped and analysed as for the second round (see section 6.8.2).

6.9 Summary

This chapter has discussed the research method of the third phase of this MM study. Phase 3 utilised a modified 3-round Delphi survey to facilitate a specialist renal community group of expert EMEESY parents and HCPs, to further explore, negotiate, and build consensus regarding the development of an EPSM strategy. As a heuristic device, Delphi enabled a group of individuals to contribute and learn on an equal footing, despite their geographical and professional differences. In seeking only consensus, many Delphi surveys fail to make full use of panellists' views; with the deviant view not always given a full hearing. Examination of the deviant views provided valuable insights, directly influencing the MMR novel findings.

The statements for Delphi round 1 were primarily informed by the findings of the Phase 1 literature and systematic review, and the qualitative data from the Phase 2 semi-structured interviews with CYP, parents and HCPs. The Delphi survey provided a platform to leverage the expert panel to shape the future of the EPSM strategy. Additional time and finances would have allowed for the development of a Delphi survey that was compatible with an android and iphone mobile platform, making it more accessible and easier to complete on the go. Chapter 7 reports the findings of each Delphi survey round.

Chapter 7: Findings of the Delphi survey

7.1 Introduction

This chapter begins with the results of the statement development for Delphi round 1 (derived from Phase 1 and 2 findings), followed by the sequential presentation of the results for each Delphi round. Delphi panellists were asked to rate their LOA with the 48 statements using four-point Likert items, as described in section 6.7.8.

7.2 Results of the statement development for Delphi round 1

Informed by the Hoffman et al. (2014) TIDieR checklist (as described in section 6.7.7), Delphi round 1 consisted of 54 initial statements which were mapped against six categories (see Box 12).

Box 12. Delphi round 1 survey categories as informed by the TIDieR checklist (2014)

1. **Definition**
2. **Rationale for the development of an EPI**
   - Why design an EPI? (statements related to chronic disease, then CKD)
3. **Intended users**
   - Who should receive the EPI?
4. **Delivery of an EPI**
   - When should we deliver EPI?
   - Where should we deliver EPI?
   - Who should deliver the EPI?
   - How should the EPI be delivered?
5. **Other considerations when planning an EPI?**
   - What else do we need to consider?
     - Relationship-building
     - Support
     - Self-management model
     - Early test undertaken in the out-patient clinic
     - Early dietary blood test
   - Potential concerns
6. **Additional factors or comments**

The source of each statement is detailed in Appendix 14. The original 56 statements were reviewed by the supervisory team and revised or collapsed into the following 48

statements. Each of the statement categories will now be explained. Statements have been abbreviated as S1-48.

7.2.1 Definition category

The definition category sought to provide a name or phrase that best described the early dietary phosphate intervention strategy (referred to as EPI throughout the DS). This would enable easy identification and understanding of the intervention. Three definitions statements were devised.

S1	Early phosphate intervention (EPI) is a program of dietary education, engaging with children, young people and their families during the early stages of chronic kidney disease (CKD 2-3), to support diet and lifestyle change towards early dietary phosphate self-management
S2	Early phosphate intervention (EPI) should be labelled 'early intervention healthy eating'
S3	The EPI diet primarily encourages a healthy eating diet, with a reduced intake of processed food, promotion of home-cooked meals containing meat, fish and dairy, and an increase in plant-based foods such as fruit, vegetables, grains and nuts.

7.2.2 Rationale for the development of an EPI category

The rationale category in the DS form described the theory or goal underpinning the intervention, chronic disease then CKD was addressed in turn. The rationale statements identified elements of theory essential to the intervention. Pragmatic issues, perceptions, the long-term impact on the child and family, eating habits and acceptability of the intervention were highlighted. Starting with chronic disease, four statements were devised.

S4	Diet has a crucial role to play in the management of a chronic disease
S5	'Junk foods' and processed foodstuffs are viewed as problem foods to be limited in the management of a chronic disease
S6	The provision of early dietary education gives children, young people and families a tool to more confidently manage their own diet during the early stages of a chronic disease
S7	When a child is diagnosed with a chronic disease families are more willing to consider dietary/lifestyle changes

For CKD, eight statement were devised.

S8	Diet is one of the hardest parts of CKD, introducing the diet early would help to make it the norm for the whole family
S9	You are going to have medicine lifelong, you are going to need to be careful about your diet lifelong.
S10	Dietary phosphate education is a long term process and should begin in the early stages of CKD
S11	Children and parents need to be told the impact of diet from an early stage, and then they can make an informed choice.
S12	Introducing dietary change early, reduces the dietary 'burden of care' in the later stages of CKD
S13	From diagnosis, parents, children and young people are actively seeking credible information about diet and CKD
S14	Children and young people with early stage CKD are at risk of eating large amounts of phosphates as these are added to processed foods in the form of food additives and preservatives
S15	Early dietary advice should be viewed as important as the medical treatment

7.2.3 Intended users category

Intended user's category described the individual or group that should receive the intervention. In the case of CKD, it identified the individual's specific staging and sought to identify whether the EPI should target specific family members. Three statements were devised.

S16	Early phosphate intervention should be offered to all children and young people with early stage CKD (2-3) who are likely to progress to the later stages of CKD (4-5)
S17	The EPI program should be directed at the whole family
S18	Mothers are often the main leaders of diet and lifestyle change in a family

7.2.4 Delivery of an EPI category

The delivery of an EPI, category, was divided into when, where, who and how statements. The one, 'when' statement spoke to the timing of delivery in relation to the CKD journey.

S19	The EPI program should be offered at, or close to the diagnosis of CKD

The one, 'where' statement spoke to the location best suited to the delivery of the EPI.

S20	The best place to deliver the EPI program is in the children's renal out-patient clinics, across the East Midlands, East of England and South Yorkshire (EMEESY) children's renal network

The following two, 'who' statements sought to identify the individual(s) best placed to be the intervention provider.

S21	Dietary information/education always carries much more authority when it comes from people who have expertise in nutrition.
S22	A children's dietitian (ideally with training in the management of CKD) is the best person to deliver the EPI program

Five 'how' statements described the mode of delivery. Whether it should be delivered face-to-face, in a written format, using social media, generic or tailored to individual needs. Whether it should take account of family household budget, and convey a clear, simple, realistic and positive message.

S23	Written dietary phosphate information is helpful, but it is no substitute for a face-to-face conversation with an expert in children's renal nutrition.
S24	Social media Is not used to its potential as a dietary educational tool during the early stages of CKD
S25	The EPI program needs to be delivered using a clear, simple, realistic and positive message
S26	The EPI program needs to fit within a family's household budget
S27	The EPI program needs to be flexible so that it can be tailored to meet the changing needs of the child or young person and their family

7.2.5 Other considerations and potential concerns category

The other considerations category sought to identify other issues and potential concerns that need attention when planning the development of an EPI strategy, namely, building relationship with the patient/family/clinician; the importance of support, and when and how should it be provided? What did SM look like for the parent/clinician? The practice and perceived use of early tests (blood tests and

assessments), and an early dietary marker in the out-patient clinic. The following four statements were devised for the other considerations category.

S38	Blood results can act as powerful reinforcements to support adherence to diet and lifestyle change
S39	Currently available tests, which give an understanding of disease progression, such as parathyroid hormone (PTH), blood pressure and protein in the urine, should be used to identify those children with CKD 2-3 who may benefit from receiving the EPI program.
S40	Even though an early blood test to support phosphate dietary management is not currently used in out-patient clinics, early dietary phosphate education is essential for all children with progressive CKD.
S41	If a blood test becomes available for use in the early stage, it needs to be something that clearly tells the healthcare professional how to manage the diet

Potential concerns, described factors not already addressed, that needed acknowledgement. These issues spoke to the impact of the intervention, such as workload, anxiety, labelling a child, intrusion to family life, family skillset, the presence of a dietitian and isolation. The following seven statements were devised for the potential considerations section.

S42	It is important to recognise that choosing to follow an EPI diet may be a burden to some families
S43	It is important to recognise that following the EPI diet may create anxiety for some families
S44	It is important to recognise that following the EPI diet may 'label' the child or young person, limiting family expectations of their child or young person
S45	It is important to recognise that following the EPI diet may be intrusive to family life
S46	It is important to recognise, due to a lack of cooking skills, it may be difficult for some families to follow the EPI diet
S47	In order to deliver the EPI diet, children's dietitians needs to have more of a presence in the shared-care clinics
S48	It is important to recognise, due to infrequent hospital/clinic visits during the early stages of CKD, parents and children or young people often feel isolated

At the end of each Delphi round, the additional factors and comments section gave the panellists the opportunity to add their own ideas and suggestions.

7.3 Results of Delphi Survey round 1

The findings for round 1 will be presented sequentially beginning with the response rate and demographic profile, then each category of statements will be discussed in turn, highlighting those statements that achieved consensus. Not all of the statements that failed to reach consensus will be addressed, but the examples provided demonstrate how the qualitative comments of the panellists influenced the revision of the statement that needed further rating in round 2.

7.3.1 Round 1 response rates

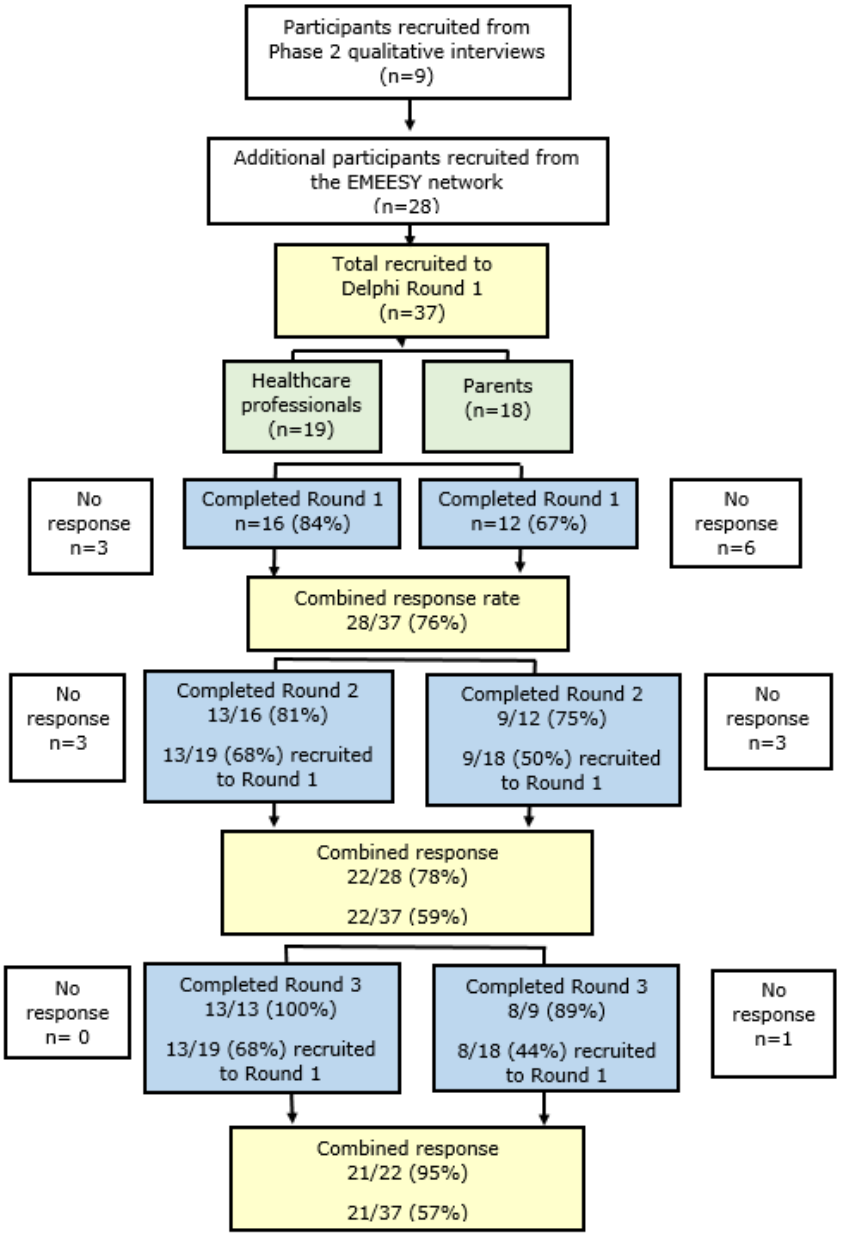
Round 1 surveys were distributed by email to 37 participants (19 HCPs and 18 parents) across the EMEESY network (see section 6.6). In total 28 round one surveys were returned, yielding a response rate of 76% (see Table 33). All three regions of the EMEESY network were represented by the returns, and the cohort comprised a comparable number of parents and HCPs (see Table 33). The attrition rate for round one was higher for the parents (33% n=6) than HCPs (15.7% n=3).

Table 33. Summary of Delphi round one responders and non-responders by region

EMEESY network region	Delphi Round 1 surveys sent out	Delphi Round 1 surveys returned
East Midlands	16 HCP 14 parents	14 HCP (10 [62.5%] based at the tertiary centre) 10 parents
East of England	2 HCP 2 parents	1 HCP 1 parent
South Yorkshire	1 HCP 2 parents	1 HCP 1 parent
Total **Attrition rate**	37 (19 HCP [51%]; 18 parents [49%])	28 (76%) (16 HCP [57%]; 12 parents [43%]) Parents = 33% HCP = 15.7%
% Response per stakeholder group		HCP = 84% Parents = 67%

The CONSORT flow diagram in Figure 18 details the recruitment and response rate across the three Delphi rounds. The figure in the blue boxes relate to each individual round, and the yellow boxes relate to the original cohort.

Figure 18. CONSORT (2010) flow diagram for Delphi round 1-3



7.3.2 Demographic profiles of the panellists

Table 34, p. 207 shows that more females participated in the survey than males, this may reflect the dominance of females in dietetic and nursing positions (Vere-Jones,

2008, BDA, 2014) and the primary role of mothers in childcare and their availability to the complete the survey.

Table 34. The primary role, median age and years of experience with CYP with CKD

Primary role		Participant group	Gender	Percent	Median age category of panelists	Modal years of experience working/ caring for CYP with CKD category
Parent/carer		12 (43%)	12 (2 males)	42.9	40-49 years	7-10
HCPs	Paediatric nephrologist/paediatrician with an interest in nephrology	16 (57%)	9 (5 males)	32.1	40-49 years	>10
	Children's renal nurse		3 (1 male)	10.7	40-49 years	>10
	Dietitian		4 (0 males)	14.3	30-39 years	7-10
Total			n=28 Male 8 (28.6%) Female 20 (71.4%)	100	Modal 40-49 years Min. 30-39 years Max. ≥60 years	Modal 7-10 years Min. < 1 year Max. >10 years

Panellists most frequently had 7-10 years of experience managing CYP with CKD (see Appendix 13). Parents, doctors and nurses all had a modal age category of 40-49 years, and dietitians had a modal age category of 30-39 years. Although all parents attended the tertiary hospital in the East Midlands as their primary treatment centre (to allow face-to-face recruitment), two parents also attended a local shared care clinic in the East of England and South Yorkshire, respectively (see Table 33, p. 205). The primary workplace for the majority of HCP was the tertiary hospital (62.5%) (see Table 33); however, all attended or supported regional clinics across the EMEESY network. Although there was an unequal number of doctors, nurses and dietitians, a mixture of doctors, nurses and dietitians weres represented (see Table 34, p. 207).

7.4 Results of statements in round 1

The results from the statements in Delphi round 1 are presented sequentially, with examples of how the round 1 free–text comments informed the statements going into round 2. As detailed in section 6.8.3, statements reaching consensus in round 1 were identified by a red shaded section. Statements still to reach consensus remained non-red. The following key applies to all tables detailing the Delphi round 1 results:

Dark red shaded box denotes ≥70% of panelist rated the statement as very high agreement, achieving a consensus response to the statement.

Statements have been given an S number e.g. statement 1 is S1

Within the tables % ratings are given in brackets for HCPs (H) and parents (P)

7.4.1 Round 1 results for the definition category (S1-3)

Three definition statements were devised from the integration of Phase 1 and 2 data (see Appendix 14) to best define the EPI (see section 7.2.1). There was consensus that S1 in the definition category should define the EPI, reaching very high agreement at 71.4% (see Box 13, p 211). A comparable % of HCP (68.75%) and parents (P) (75%) rated S1 as 4 – very high agreement.

Box 13. Median LOA rating, % LOA and participant group weighting for the 3 definition statements in round 1

Category: definition (S1-3)	Median LOA rating (1-4)	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S1	Early phosphate intervention (EPI) is a program of dietary education, engaging with children, young people and their families during the early stages of chronic kidney disease (CKD 2-3), to support diet and lifestyle change towards early dietary phosphate self-management				
	4	71.4 (H68.75/P75)	25 (H31.25/P16.6)		3.6 (H0/P8.3)
S2	Early phosphate intervention (EPI) should be labelled 'early intervention healthy eating'				
	3	21.5 (H25/P16.6)	32.2 (H43.75/P16.6)	28.5 (H31.25/P25)	17.8 (H0/P41.6)
S3	The EPI diet primarily encourages a healthy eating diet, with a reduced intake of processed food, promotion of home-cooked meals containing meat, fish and dairy, and an increase in plant-based foods such as fruit, vegetables, grains and nuts.				
	3.5	50 (H62.5/P33.3)	28.6 (H31.25/P25)	17.8 (H6.25/P33.3)	3.6 (H0/P8.3)

Participant group (H = HCPs, P = parents) % weighting for each LOA is given in brackets

Parents comments for S2 suggested the term 'healthy-eating' should be used with caution, as it implied the avoidance of high calorie snacks, something that CYP with CKD often require. One parent felt the term healthy-eating contradicted the message, stating 'I thought I was

providing "Healthy-Eating" by giving my baby steamed vegetables and lots of fresh fruit, which was completely the opposite of what was required once he was diagnosed with CKD' (DEL_PAR19). Furthermore, the removal of 'phosphate' from the definition ('early intervention healthy-eating') created confusion about the primary focus of the intervention, and to combine phosphate control with healthy-eating was felt to be 'not always achievable' (DEL_PAR23). Conversely, a HCP defended this 'healthy-eating' debate, urging for the inclusion of 'healthy-eating for healthy kidneys' (DEL_HCP09) in the definition. Another HCP argued for a definition that featured the term 'kidney', explaining, clear 'marketing is important to aid engagement' (DEL_HCP16). Finally, the term 'intervention' met with disapproval, although no explanation was given (DEL_HCP09). Box 14 gives S2 in round 1 and the revised statement in round 2.

Box 14. Definition S2

Domain:	Statement		Very high agreement	High agreement	Moderate agreement	Low agreement
Definition	2.	Early phosphate intervention (EPI) should be labelled 'early intervention healthy eating'				
How **the panel** responded in round 1			21.5%	32.2%	28.5%	17.8%
Your round 2 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after round 1.						
Definition	2.	The early phosphate intervention (EPI) program should be labelled 'early phosphate management for healthy kidneys'	☐	☐	☐	☐

Furthermore, for definition S3 (see Box 32, Appendix 15), which builds on the above comments from S2, one of the HCP agreeing with the healthy-eating focus of the intervention, stated, in order to 'bring a focus for families, children and young people' the term needs to be used in conjunction with 'phosphate' (DEL_HCP02). Another HCP concurred with the important of an early healthy-eating message, arguing 'all families attending a renal clinic could do with healthy-eating advice as other factors such as salt and calorie intake are important'. He suggested it would be 'a good strategy to combine healthy-eating with early phosphate intervention' (DEL_HCP06).

Two revised definition statements (S2 and S3) went through to round 2. Next, the statements addressing the rationale for the EPI strategy are presented.

7.4.2 Round 1 results for the rationale category (S4-15)

There was consensus that 5 of the 12 rationale statements should support the EPI, reaching very high agreement (see Box 33, Appendix 15). These findings recognised the long-term vital and empowering role of diet in the management of CKD. An equal number of panellists rated rationale S7 about a chronic disease diagnosis and the willingness to consider dietary/lifestyle changes (see Box 33, Appendix 15) with very high agreement, as for high and moderate agreement combined.

However, panellists' comments, which directly shaped the development of the two new statements (S7a and 7b) in round 2 (see Box 34, Appendix 15), suggested the statement required small to significant changes. HCPs provided many suggestions, such as the inclusion of the modal verb 'may' in the statement (DEL_HCP03). Since changing the diet of a child may be one of the few things parents can do in the early stages, the rationale should include 'provides a positive focus for the family' (DEL_HCP06). Although the diagnosis of a chronic disease helps give importance to a healthy diet, one HCP felt that 'it can be hard to change behaviour if the family don't have support from a dietitian' (DEL_HCP09). Contending S7, DEL_HCP11 felt it implied that only those with chronic disease would consider healthy lifestyle choices, and suggested inclusion of the phrase 'knowing and understanding the impact of diet on their disease, makes them more motivated to make changes'. Believing that diet gives a family agency, this parent felt that 'dietary change can give a positive focus' as 'their actions are helping their loved ones at a difficult time' (DEL_PAR22). Considering food

less intimidating than medication, this parent asserted that dietary change places the parent 'more in control than they can be with medications' (DEL_PAR26).

Rationale S13 regarding the seeking of credible information about diet and CKD, failed to reach consensus in round 1 (see Box 35, Appendix 15) with 57.1% of the panellists rating with very high agreement. Panellist comments, suggested S13 required small to significant changes. One HCP who opposed this statement, believed that 'not all actively seek' advice (DEL_HCP16). One mother affirmed in a rebuttal of this opinion that 'families are desperate for information and keen to do what they can to help' (DEL_PAR19). One parent challenged the adjective 'credible', instead encouraging the use of 'helpful' (DEL_PAR22).

Consensus was almost reached for rationale S14, which stated that CYP with CKD are at risk of eating large amounts of added phosphates from processed foods, with a very high agreement of 64.3% (see Box 36, Appendix 15). However, panellists' ratings suggested that the statement required small to significant changes. Only one parent commenting on this statement felt it implied that all parents were giving their children processed foods. Defending her stance she explained how children 'are only at risk if they are given these foods' and that 'her child was not at risk because I made home-cooked meals' (DEL_PAR22).

Eight rationale statements went through to round 2 for further rating by panellists: four were revised (S5, S9, S13 and S14), S7 was made into 2 sub-statements (7a and 7b), and S8 and S12 remained the same.

Next, the results of the statements addressing the individual or group that should receive the EPI are presented.

7.4.3 Round 1 results for the intended user's category (S16-18)

There was consensus that 2 of the 3 intended user's statements (S16 and 17) should support the EPI, reaching very high agreement (see Box 38, Appendix 15).

Recognising the complexity of family configuration, and the increasing involvement of fathers, comments for S18 (the only remaining statement in the intended user's category still to reach consensus) proposed a more inclusive tone to the statement.

Two HCP offered the phrase 'leader(s) of lifestyle and dietary change in a family' (DEL_HCP07; DEL_HCP16) as a replacement for 'mothers'. If the intention is to encourage 'the rest of the family… to sign up too', the mother should not be the key focus (DEL_HCP11). Other HCPs and parents affirmed the need to recognise and elicit support from the wider family – siblings, grandparents and guardians. A revised version of S18 went through to round 2, namely, 'The EPI program should target the main leaders of diet and lifestyle change in a family' (see Box 37, Appendix 15). The next section discusses the results of the statements which addressed delivery of the EPI.

7.4.4 Round 1 results for delivery of an EPI category (S19-27)

There was consensus that 6 of the 9 delivery statements in this category should support the EPI, reaching very high agreement (see Box 15).

Box 15. Median LOA rating, % LOA and participant group weighting for the delivery statements in round 1

Category: delivery (S19-27)	Median level of agreement rating (1-4)	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S19 (when should we deliver EPI?)	The EPI program should be offered at, or close to the diagnosis of CKD				
	4	75 (H62.5/P91.6)	17.9 (H25/P8.3)	7.1 (H12.5/P0)	
S20 where should we deliver EPI?	The best place to deliver the EPI program is in the children's renal out-patient clinics, across the East Midlands, East of England and South Yorkshire (EMEESY) children's renal network				
	4	53.6 (H50/P58.3)	39.3 (H43.75/P33.3)	7.1 (H6.25/P8.3)	
S21 (who should deliver the EPI?)	Dietary information/education always carries much more authority when it comes from people who have expertise in nutrition.				
	4	78.6 (H81.25/P75)	21.4 (H18.75/P25)		
S22	A children's dietitian (ideally with training in the management of CKD) is the best person to deliver the EPI program				
	4	82.1 (H87.5/P75)	17.9 (H12.5/P25)		
S23 how should the EPI be delivered?	Written dietary phosphate information is helpful, but it is no substitute for a face to face conversation with an expert in children's renal nutrition.				
	4	64.3 (H68.75/P58.3)	32.1 (H31.25/P33.3)	3.6 (H0/P8.3)	
S24	Social media is not used to its potential as a dietary educational tool during the early stages of CKD				
	3	28.6 (H43.7/P8.3)	39.3 (H37.5/P41.6)	17.9 (H12.5/P25)	14.2 (H6.25/P25)
S25	The EPI program needs to be delivered using a clear, simple, realistic and positive message				
	4	71.4 (H81.25/P58.3)	28.6 (H18.75/P41.6)		
S26	The EPI program needs to fit within a family's household budget				
	4	75 (H87.5/P58.3)	14.3 (H12.5/P16.6)	7.1 (H0/P16.6)	3.6 (H0/P8.3)
S27	The EPI program needs to be flexible so that it can be tailored to meet the changing needs of the child or young person and their family.				
	4	82.1 (H87.5/P75)	14.3 (H12.5/P16.6)	3.6 (H0/P8.3)	

Only 53.6% of the panel rated the delivery S20, which stated that the best place to deliver the EPI is in the Children's renal out-patient clinics across the EMEESY network (see Box 39, Appendix 15), with very high agreement.

Six panellists (4 HCPs and 2 parents) felt the EPI should be part of a home visit, allowing insight to the 'cooking environment/ability to help with diet plans' (DEL_PAR19). One parent agreed, it would be good to have the information, and as home 'is where most of the patient's relationship with food will stem from', this would seem the ideal location (DEL_PAR26). Other locations offered included: hospital ward; shared care clinic with other team members followed up by a telephone call; Skype call/clinic or group discussions; interactive online dietary advice and games, and an online dietitian to answer questions. One HCP agreed it should be delivered as close to the family home as possible (DEL_HCP15).An invitation to meet the team and learn about the diet should be sent to families in close proximity shortly after diagnosis, suggested a parent (DEL_PAR17). Another parent who requested 'access to timely information' from 'relevant professionals in one place' (DEL_PAR19) echoed this. One parent felt 'access to dieticians with a strong renal leaning is imperative' as part of the regular consultant's review (DEL_PAR23). Highlighting the value of 'peer to peer learning', this parent felt that 'older children can maybe help when you are teaching/advising the younger children and their families' and that they are most 'likely to listen to someone who has been through it' (DEL_PAR27).

Nearly 70% (64.3%) of panellists rated as very high agreement statement 23 that considered 'how' the EPI should be delivered (see Box 40, Appendix 15). The value of written information was recognised by HCPs, but only after a face-to-face discussion. Indeed, 'you can't beat face-to-face input' commented one parent, 'it provides the dietitian with an opportunity to ensure that the messages are delivered and understood accurately, and also provides opportunity for the family to respond with questions and queries' (DEL_PAR19). Comments from one parent spoke of how much she prized the written information because it 'can be referred to time and time again' (DEL_PAR22); another found it 'a huge help as you don't always have people to ask and they make a great reference point' (DEL_PAR23). Recipe books and regular seminar/workshops in the local area were suggested to include dietitian-led cooking

sessions, giving families the opportunity to share ideas (DEL_PAR22). These comments informed the development of two new sub-statements in round 2 (see Box 40, Appendix 15).

Five statements from the delivery category went through to round 2 for further rating by panellists, 1 was revised (S24) and 2 were made into new sub-statements (20a and 20b, and 23a and 23b,).

Next, the results of the statements addressing miscellaneous issues and potential concerns that need attention when planning the development of an EPI strategy are presented. It produced a lower LOA generally than previous sections. Individual sub-sections are presented to understand where the disagreements were.

7.4.5 Round 1 results for the other considerations category (S28-48)

There was consensus that 2 of the 21 statements in the other considerations category should support the EPI, reaching very high agreement (refer to Box 16).

Box 16. Median LOA rating, % LOA and participant group weighting for the other considerations statements in round 1

Category: other considerations (S28-48)	Median LOA rating	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S28 building relationship	It is important to build a trusted relationship with a healthcare professional before diet and lifestyle change can take place.				
	2.5	21.4 (H31.25/P8.3)	28.6 (H31.25/P25)	42.9 (H31.25/P58.3)	7.1 (H6.25/P8.3)
S29	Time is crucial for trust and respect to be built in a relationship				
	3	35.7 (H43.7/P25)	35.7 (H37.5/P33.3)	21.4 (H18.75/P25)	7.1 (H0/P16.6)
S30	The healthcare professional needs time to understand the individual and family needs.				
	3	39.3 (H50/P25)	50 (H50/P50)	7.1 (H0/P16.6)	3.6 (H0/P8.3)
S31 support	Ongoing feedback and support from healthcare professionals are essential to encourage adherence to diet and lifestyle change				
	4	71.4 (H75/P66.6)	17.9 (H18.75/P16.6)	10.7 (H6.25/P16.62)	
S32	Parents of children with early stage CKD need support from other parents who are in or have been in their situation e.g. support groups, social media				
	3	46.4 (H43.7/P50)	25 (H43.75/P0)	28.6 (H12.5/P50)	
S33	An EPI support group should be set up for parents across the shared-care renal network e.g. online or face to face in the clinic				
	4	57.1 (H62.5/P50)	25 (H25/P25)	14.3 (H12.5/P16.6)	3.6 (H0/P8.3)
S34	Patient stories and shared experiences are vital, they provide reassurance and make a difference to how a person feels they are able to manage their diet/lifestyle change.				
	4	64.3 (H62.5/P8)	17.9 (H25/P1)	17.9 (H12.5/P3)	
S35 SM	The healthcare professional should solely direct the delivery of the EPI program.				
	2	10.7 (H12.5/P8.3)	28.6 (H31.25/P25)	28.6 (H43.75/P8.3)	32.1 (H12.5/P58.3)
S36	The healthcare professional should work in partnership with the child or young person and their family in order to tailor the EPI program to the changing needs of the individual				
	4	75 (H75/P75)	25 (H25/P25)		

S37	The child or young person and their family should direct the delivery of the EPI program				
	3	25 (H18.75/P8.3)	28.6 (H50/P25)	14.3 (H25/P0)	32.1 (H6.25/P66.6)
S38 early tests undertaken in the out-patient clinic	Blood results can act as powerful reinforcements to support adherence to diet and lifestyle change.				
	4	57.1 (H7/P9)	25 (H6/P1)	14.3 (H3/P1)	3.6 (H0/P1)
S39	Currently available tests which give an understanding of disease progression, such as parathyroid hormone (PTH), blood pressure and protein in the urine, should be used to identify those children with CKD 2-3 who may benefit from receiving the EPI program.				
	4	57.1 (H37.5/P83.3)	21.4 (H37.5/P0)	14.3 (H25/P0)	7.2 (H0/P16.6)
S40 early dietary blood tests	Even though an early blood test to support phosphate dietary management is not currently used in out-patient clinics, early dietary phosphate education is essential for all children with progressive CKD.				
	4	67.9 (H62.5/P75)	17.9 (H31.25/P0)	10.7 (H6.25/P16.6)	3.5 (H0/P8.3)
S41	If a blood test becomes available for use in the early stage, it needs to be something that clearly tells the healthcare professional how to manage the diet				
	4	57.1 (H50/P66.6)	21.4 (H25/P16.6)	14.3 (H18.75/P8.3)	7.2 (H6.25/P8.3)
S42 potential concerns	It is important to recognise that choosing to follow an EPI diet may be a burden to some families				
	3	42.9 (H56.25/P25)	39.3 (H31.25/P50)	7.1 (H6.25/P8.3)	10.7 (H6.25/P16.6)
S43	It is important to recognise that following the EPI diet may create anxiety for some families				
	3.5	50 (H56.25/P41.6)	28.6 (H31.25/P25)	14.3 (H12.5/P16.6)	7.1 (H0/P16.6)
S44	It is important to recognise that following the EPI diet may 'label' the child or young person, limiting family expectations of their child or young person				
	2	25 (H31.25/P16.6)	21.4 (H25/P16.6)	25 (H31.25/P16.6)	28.6 (H12.5/P50)
S45	It is important to recognise that following the EPI diet may be intrusive to family life				
	3	32.1 (H43.7/P16.6)	35.7 (H37.5/P33.3)	21.4 (H12.5/P33.3)	10.7 (H6.25/P16.6)
S46	It is important to recognise, due to a lack of cooking skills, it may be difficult for some families to follow the EPI diet				
	3	46.4 (H56.25/P33.3)	25 (H37.5/P8.3)	10.7 (H0/P25)	17.8 (H6.25/P33.3)
S47	In order to deliver the EPI diet, children's dietitians needs to have more of a presence in the shared-care clinics				
	4	60.7 (H68.7/P50)	28.6 (H31.25/P25)	10.7 (H0/P25)	
S48	It is important to recognise, due to infrequent hospital/clinic visits during the early stages of CKD, parents and children or young people often feel isolated				
	3	46.5 (H56.25/P33.3)	28.5 (H31.25/P25)	17.8 (H12.5/P25)	7.2 (H0/P16.6)

S28, one of the 'building relationship' statements, was rated by the majority of panellists as moderate agreement (see Box 16). Recognising that it takes time to develop a relationship and trust, one parent prioritised the dietitian's assessment and delivery of dietary information 'as early to diagnosis as possible', once delivered 'trust will then develop' (DEL_PAR19). Similarly, asserting the urgency of receiving information and advice, this parent felt dietary 'changes should be made ASAP', resulting in a sense of confidence and trust which comes as they work with the HCP (DEL_PAR22). Another parent felt strongly that there should be 'a two-pronged approach as soon as you are discharged - dietician and consultant' (DEL_PAR23). Box 17, p. 217 details the three new sub-statements that were developed for S28 in round 2.

Box 17. Other considerations: building relationship, S28

Domain:	Statement – building relationship		Very high agreement	High agreement	Moderate agreement	Low agreement
Other considerations	28.	It is important to build a trusted relationship with a healthcare professional before diet and lifestyle change can take place.				
How the panel responded in round 1			21.4%	28.6%	42.9%	7.1%
Your round 2 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after round 1 into 3 new statements.						
Other considerations	28a.	As part of the early management of CKD the family should be introduced to the doctor and dietitian.	☐	☐	☐	☐
Other considerations	28b.	Diet and lifestyle information, and ongoing support and advice, should be offered as early in the diagnosis as possible.	☐	☐	☐	☐
Other considerations	28c.	Early introduction to the dietitian means trusted relationships will develop over time, making the nutritional message more effective.	☐	☐	☐	☐

Just less than 50% (46.4%) of the panellists felt that S32, which focused on parents receiving 'support' from other parents, should be used to development an EPI (refer to Box 18, p. 218). Comments highlighted the importance of giving parents the option of "accessing" support from other parents, recognising that not everyone wishes to use social media or attend a parents' group (DEL_HCP14). Social media can be a useful tool, but to control unhelpful or incorrect information, it needs close monitoring (DEL_HCP07). One parent felt the idea of connecting with other

parents 'would be great', and even suggested that 'opportunities to get together and cook would be fun and informative' (DEL_PAR22). Box 18 shows how the comment informed the development of the statement in round 2.

Box 18. Other considerations: support, S32

Domain:	Statement - support		Very high agreement	High agreement	Moderate agreement	Low agreement
Other considerations	32.	Parents of children with early stage CKD need support from other parents who are in or have been in their situation e.g. support groups, social media				
How **the panel** responded in round 1			46.4%	25%	28.6%	
Your round 2 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after round 1.						
Other considerations	32.	Parents of children with early stage CKD should have access to support from other parents who are in or have been in their situation.	☐	☐	☐	☐

Support statement 34, statingthe vital role of patient stories and shared experiences in the management of diet/lifestyle change, almost reached consensus with 64.2% of panellists rating it as very high agreement (see Box 41, Appendix 15). Comments from panellists asserted that this type of support is the 'key to long term change' (DEL_HCP02), as there is great value when we 'harness the power of families mentoring each other' (DEL_HCP06). As one parent said 'if we could have found other parents who have been through this with their children, we would have felt less isolated' (DEL_PAR21). This statement remained the same in round 2 as all of the comments affirmed the value of patient stories and shared experiences. The majority of the panel rated S35 and 37 (32.1% for both), describing the medical-model and self-agency SM style, respectively,as low agreement (see Box 42 and Box 43, Appendix 15). Comments implied the HCP should not solely direct the delivery, rather it should be a 'joint

directive' (DEL_HCP07) using a 'partnership' model (DEL_HCP14). Parents felt it was necessary not only for the dietitian to lead the EPI but also for the 'team and other parents' (DEL_PAR17) to be involved, with 'ongoing conversation' to support delivery (DEL_PAR22).

S38 (see Box 19), exploring the value of out-patient blood tests during the early stages of CKD was rated with very high agreement by 57.1%. One comment from a HCP explained that early CKD test results mean more to clinicians than families, and as levels of blood phosphate and parathyroid hormone are not typically problematic in the early stages 'a marker that…reflect[s] the dietary changes is more valuable - demonstrating a reward for their efforts' (DEL_HCP07). Recognising the potential importance of early blood tests, this mother explained the need for blood values to be 'explained in layman's terms so that parents can understand the importance of the results' (DEL_PAR28).

Box 19. Other considerations: early tests undertaken in the out-patient clinic, S38

Domain:	Statement - early tests undertaken in the out-patient clinic		Very high agreement	High agreement	Moderate agreement	Low agreement
Other considerations	38.	Blood results can act as powerful reinforcements to support adherence to diet and lifestyle change.				
How **the panel** responded in round 1			57.1%	25%	14.3%	3.6%
Your round 2 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after round 1.						
Other considerations	38.	Blood results, where they can be explained clearly to families and used to guide management, can act as powerful reinforcements to support adherence to diet and lifestyle change.	☐	☐	☐	☐

Similarly, S39, exploring the use of out-patient blood tests during the early stages of CKD was rated with very high agreement by 57.1% of the panellists (see Box 20). However, two voted with low agreement; one of these was a parent who commented that her lack of knowledge on this matter made it difficult for her to make a valid judgement (DEL_PAR27). The remaining 35.7% felt that small to significant changes were needed. Only one HCP commented on the use of early blood tests, stating that parathyroid hormone rises in late stage 3, so it is 'not an early marker of phosphate balance in the body' (DEL_HCP09). Despite the lack of expressed opinion in HCP comments, one parent felt strongly that in the early stages 'blood results are all families have to go on', she described how they can 'reinvigorate the family to look at diet' (DEL_PAR19). Contending the use of blood tests as a tool for identifying only early-stage CKD children who could benefit from the EPI, this parent felt it should not be exclusive, but that 'all patients should be able to access the program' (DEL_PAR22). Box 20 shows how the comments shaped the revised S39 in round 2.

Box 20. Other considerations: early tests undertaken in the out-patient clinic, S39

Domain:	Statement - early tests undertaken in the out-patient clinic		Very high agreement	High agreement	Moderate agreement	Low agreement
Other considerations	39.	Currently available tests which give an understanding of disease progression, such as parathyroid hormone (PTH), blood pressure and protein in the urine, should be used to identify those children with CKD 2-3 who may benefit from receiving the EPI program.				
How the panel responded in round 1			57.1%	21.4%	14.3%	7.2%
Your round 2 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after round 1.						
Other considerations	39.	Currently available tests which give an understanding of disease progression, such as parathyroid hormone (PTH), blood pressure and protein in the urine, should be explained to parents, and used to identify those children with CKD 2-3 who may benefit from receiving the EPI program.	☐	☐	☐	☐

Over half of the panel (57.1%) rated S41, which explored the role of a dietary blood test during the early stages of CKD, with very high agreement (see Box 44, Appendix 15). Two panellists rated the statement with low agreement. One of these, a HCP, seemed to have rated the statement as low agreement due to feeling the statement lacked clarity on the function of the early blood test, and not because he felt the statement should not be used to support the EPI development (DEL_HCP16). The other, a parent, rated low agreement as an act of rejecting the idea of a dietary blood, saying 'the healthcare professional should know how to manage the diet without the blood test' and that 'good practice will be the same regardless' (DEL_PAR22). Describing the ideal function of a dietary blood test, this HCP felt 'it needs to tell the family and health professional how to manage diet' (DEL_HCP07). Again, this parent seemed to be convinced that regardless of a dietary blood test, 'all patients should be offered it [EPI education] from the start' (DEL_PAR17).

Nearly half of the panellists (42.9%) rated statement 42, which described the EPI as potentially burdensome, as very high agreement (see Box 45, Appendix 15). Three panellists (10.7%) gave it a low agreement rating. One of these, a HCP, seemed to oppose the idea that the EPI was a 'burden', removing factors he felt might be associated with being burdensome, he explained, 'early CKD diet is a healthy diet. It is not restrictive. It does not have to be expensive' (DEL_HCP09). In line with this belief that diet is not the problem, parent DEL_PAR22 affirmed that 'having kidney disease is the burden!' Similarly, parent DEL_PAR23 vowed that diet 'should not be seen as a burden but a necessity', she explained, 'we all want the best well-being for our children and if EPI helps them maintain a healthy outlook then so be it.' Likewise, after the initial shock of diagnosis, parent DEL_PAR19 explained that families 'want to do all they can to help their loved one', and 'if the information is simple and provided in the right way it does not need to be onerous. Give people simple changes they can make at first and then build it up. Offer cooking skills advice or refer to adult learning classes.' Recognising potential concerns of an EPI, one parent felt so long as HCPs are aware of these issues, it could be factored into the delivery and care plan. She explained that 'change is difficult for anyone and with correct planning and understanding this can be achieved' (DEL_PAR17).

Statement 44, describing how the EPI could 'label' the CYP, received parity across the agreement levels (see Box 21). Of the 28.6% of panellists that rated it with low agreement, one of these, panellist DEL_PAR226, speaking of their child, declared 'they've already got the label of being unwell', implying that they are not labelled by the diet. Parent DEL_PAR23 also rejected the idea that diet might create a label; he explained that the diet 'limits the amount of food stuffs they can eat, but nothing else.' Box 21 shows how the comments shaped the revised S44 for round 2.

Box 21. Other considerations: potential concerns, S44

Domain:	Statement - potential concerns		Very high agreement	High agreement	Moderate agreement	Low agreement
Other considerations	44.	It is important to recognise that following the EPI diet may 'label' the child or young person, limiting family expectations of their child or young person				
How **the panel** responded in round 1			25%	21.4%	25%	28.6%
Your round 2 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after round 1.						
Other considerations	44.	Following the EPI diet may identify the child or young persons as having different dietary needs. This does not have to limit their own, and their family expectations as they grow up.	☐	☐	☐	☐

Agreement ratings for statement 45, which identified the EPI diet potentially intrusive to family life, suggested uncertainty (see Box 46, Appendix 15). Dismissing the idea of the EPI as intrusive, parent DEL_PAR22 argued 'the changes go with the territory'. Again, parent DEL_PAR23 contended the diet as intrusive, explaining 'it doesn't need to be, once your child accepts some stuff they can have and some they can't then it just about adapting'.

Almost half of the panel (46.5%) rated statement 46 (see Box 22), which recognised a lack of cooking skills as a potential concern, with very high agreement.

Box 22. Other considerations: potential concerns, S46

Domain:	Statement - potential concerns		Very high agreement	High agreement	Moderate agreement	Low agreement
Other considerations	46.	It is important to recognise, due to a lack of cooking skills, it may be difficult for some families to follow the EPI diet				
How the panel responded in round 1			46.5%	25%	10.7%	17.8%
Your round 2 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after round 1.						
Other considerations	46.	The EPI program will encourage more home-cooked meals. Some families may benefit from cookery workshops to inspire and support them to follow the program.	☐	☐	☐	☐

Interestingly, one of the panellists who rated it with a low agreement showed agreement with the statement in her comments, with a suggested solution, she explained, 'there is an assumption that there is a lack of cooking skills amongst some families. This is why workshops would be a good idea' (DEL_PAR22). Rating the statement with a very high LOA, one HCP asked 'who is going to deliver or fund them?' (DEL_HCP01).

Over 60% of panellists rated S47, which recognised a need for the dietitian to have a greater presence in the shared-care clinics, with very high agreement (see Box 47, Appendix 15). One parent, affirming the statement with a very high agreement, commented 'absolutely' (DEL_PAR22). Nearly half of the panel rated S48, which identified isolation during the early stage of CKD as a potential concern, with very high agreement (see Box 23, p. 224).

Box 23. Other considerations: potential concerns, S48

Domain:	Statement - potential concerns		Very high agreement	High agreement	Moderate agreement	Low agreement
Other considerations	48.	It is important to recognise, due to infrequent hospital/clinic visits during the early stages of CKD, parents and children or young people often feel isolated				
How the panel responded in round 1			46.5%	28.5%	17.8%	7.2%
Your round 2 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after round 1.						
Other considerations	48.	With the right level of education and support during the early stages of CKD, children and young person and their parents should not feel isolated.	☐	☐	☐	☐

Parent DEL_PAR22 again insisted 'this is why local hubs for parents would be good. An invitation to come along to a diet/cooking workshop.' For one of the parents who rated the statement with a low agreement, this seemed to reflect the fact that her child was diagnosed with advanced stage CKD, and therefore never attended clinic during the early stages. While recognising isolation as a concern, she affirmed that 'dieticians are always available at clinic or via phone/email… there is always the option to contact someone with questions' and therefore no one needs to feel isolated (DEL_PAR26).

Twenty-three statements from the other considerations category went through to round 2 for further rating by panellists, 13 were revised (S29, S32, S35, S37, S38, S39, S41, S42, S43, S44, S45, S46 and S48), 3 were made into 7 new sub-statements (28a, 28b and 28c, 30a and 30b, 33a and 33b) and 3 remained the same (S34, S40, S47).

7.4.6 Summary of Delphi round 1

Consensus was achieved on a definition for EPI. The term 'healthy-eating' and 'burden' in the context of the CKD diet were met with contention. Diet has a vital long-term role in the management of CKD. Rationale and other considerations statements required greater emphasis on the value of early engagement and partnership with HCPs and other parents/families to provide long-term education and support. Additionally, an early dietary blood test needs to clearly guide dietary management. The leaders of lifestyle and dietary change are more diverse than just mothers, and EPI should be offered at or close to diagnosis of CKD by the dietitian, in an appropriate and accessible format.

7.5 Results of statements in round 2

This section reports the development and findings of the round 2 Delphi survey, and examples of how the round 2 free-text comments shaped the round 3 statements.

7.5.1 Development of the round 2 survey

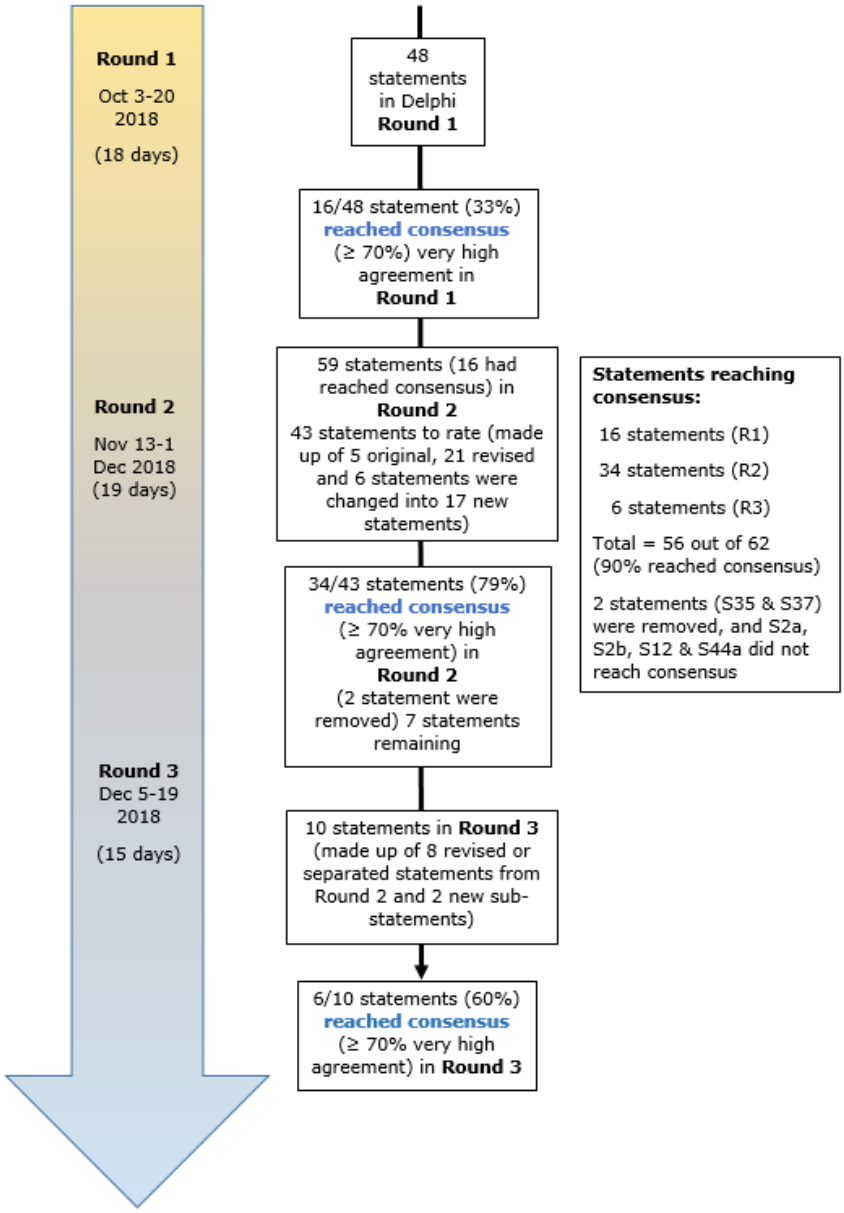
The outcome of the statements at the start of round 2 are listed in Table 35.

Table 35. Statements at the end of round 1

Statements that achieved consensus	Statements that remained unchanged	Statements that were revised	Statements that were changed into new statements
S1, 4, 6, 10, 11, 15, 16, 17, 19, 21, 22, 25, 26, 27, 31 and 36	S8, 12, 34, 40 and 47	S2, 3, 5, 9, 13, 14, 18, 24, 29, 32, 35, 37, 38, 39, 41, 42, 43, 44, 45, 46 and 48	S7, 20, 23, 28, 30 and 33

Fifty-nine statements (including sub-statements) went into round 2 (see Figure 19, p. 226). Statements that had reached consensus were clearly marked in the Delphi Survey (see section 6.8.3, Box 10), indicating that no further response was necessary, leaving 43 statements to be rated by the panel (refer to Figure 19).

Figure 19. Statements reaching consensus across 1-3 Delphi rounds



7.5.2 Round 2 response rate

Round 2 surveys were distributed by email or posted to the 28 participants (16 HCPs, 12 parents) across the EMEESY network who completed and returned the round 1 survey (see section 6.6). In total, 22 round 2 surveys were returned, yielding a response rate of 78% (see Table 36). The return time for round 2 DS was slower than round 1. This may have reflected time restraints, and the longer length of the round 2 survey. Using the dynamic strategy (see section 6.7.12), attrition was minimised and the response rate reached the target of >70% (Walker and Selfe, 1996; Sumsion, 1998). An attrition of 22% was comparable to the 28% recorded by Alexandrov et al. (1996), and less than the 50% recorded by Lambert et al. (2019). All regions of the EMEESY network were represented by the returns.

Table 36. Summary of Delphi round 2 responders and non-responders by region

EMEESY network region	Delphi Round 2 surveys sent out	Delphi Round 2 surveys returned
East Midlands	14 HCP 10 parents	12 HCP (10 based at the tertiary centre) 7 parents
East of England	1 HCP 1 parents	1 HCP 1 parent
South Yorkshire	1 HCP 1 parents	0 HCP 1 parent
Total Attrition	28 (16 HCP [57%]; 12 parents [43%])	22 (78%) (13 HCP [59%]; 9 parents [41%]) Parents 25% HCP 19%
% Response per stakeholder group		HCP = 81% Parents = 75%

The round 2 cohort comprised more HCPs than parents (see Table 37). Although the number of doctors, nurses and dietitians was not equal, a mixture of doctors, nurses and dietitians was represented. Similar to round 1, more females participated in round 2 than males.

Table 37. Primary role and gender of round 2 participants

Primary role		Participant group Delphi Round 2 responders	Gender	Percent
Parent/carer		9 (41%)	9 (1 males)	40.9
HCPs	Paediatric nephrologist/paediatrician with an interest in nephrology	13 (59%)	7 (4 males)	31.8
	Children's renal nurse		2 (0 male)	9.1
	Dietitian		4 (0 males)	18.2
Total			n=22 Male 5 (22.7%) Female 17 (77.3%)	100

The following key applies to all tables detailing the Delphi round 2 results:

Yellow shaded box denotes ≥70% of panelist rated the statement as very high agreement, achieving a consensus response to the statement.

In addition to the S number, round 2 is abbreviated as R2 e.g. statement 2 round 2 is S2/R2

7.5.3 Round 2 results for the definition category

There was consensus that S3/R2 in the definition category should define the EPI, reaching very high agreement at 77.3% (see Box 24, p. 229), compared to 50% for the corresponding statement in round 1.

Box 24. Median LOA rating, % LOA and participant group weighting for the definition statements in round 1 and 2

Category: definition (S1-3)	Median score	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S1	4	71.4 (H68.75/P75)	25 (H31.25/P16.6)		3.6 (H0/P8.3)
S3	3.5	50 (H62.5/P33.3)	28.6 (H31.25/P25)	17.8 (H6.25/P33.3)	3.6 (H0/P8.3)
S3/R2	4	77.3 (H77/P77.7)	22.7 (H23/P22.2)		

This left S2/R2 definition still to reach consensus (see Box 25 p. 230). Despite the use of the term 'healthy kidneys' in the revised S3, its use in S2 provoked comments from both HCPs and parents (see Box 25). The three HCP who rated S2/R2 with low agreement emphatically disagreed with the implication of this statement. One HCP explained 'the kidneys are NOT healthy otherwise you would not be intervening!' he went on to propose an alternative definition for EPI, namely, 'early phosphate management and healthy-eating in CKD' (DEL_06HCP). Another HCP likewise explained that the revised statement 'implies there is no problem with the kidneys' (DEL_07HC). The parent who rated S2/R2 with low agreement concurred with these remarks; saying 'if we are on the program by default we don't have healthy kidneys' (DEL_23PAR). She suggested using the words 'to reduce kidney decline or maintain kidney health'. While another HCP thought the focus on phosphate alone, limited the potential of an early dietary intervention (DEL_09HCP). He clarified that phosphate education should be nested within a healthy-eating message. This was affirmed by a parent who advocated for other dietary changes to be implemented alongside the phosphate message that would be useful for kidney health (DEL_19PAR).

Box 25. Definition S2 in round 1-3

Domain:	Statement 2		Very high agreement	High agreement	Moderate agreement	Low agreement
Definition	Round 1.	Early phosphate intervention (EPI) should be labelled 'early intervention healthy eating'				
How **the panel** responded in round 1			21.5%	32.2%	28.5%	17.8%
Definition	Round 2.	The early phosphate intervention (EPI) program should be labelled 'early phosphate management for healthy kidneys'	Very high agreement	High agreement	Moderate agreement	Low agreement
How **the panel** responded in Round 2			45.4%	31.8%	4.5%	18.3%
Your round 3 response (to select your response please click inside one box. Please note you do not have to change your response). **Please note this statement has been revised after round 2 into 2 new statements.**						
Definition	Round 3. 2a.	The EPI program should be labelled as 'early phosphate management and healthy eating for kidney health'				
Definition	Round 3 2b.	The EPI program should be labelled as 'early phosphate management and healthy eating for CKD'				

As a result of these contentions, this statement failed to reach consensus. In response to the comments, S2 was made into two new sub-statements for round 3 (see Box 25).

7.5.4 Round 2 results for the rationale category

There was consensus that S5, about recognising and consuming less processed food, and 7a and 7b regarding understanding the impact of diet and how early dietary changes can provide a positive focus for the family, respectively, should support the rationale for the EPI (see Table 38, p. 232).

Additionally, S8 which recognised how diet is one of the hardest parts of CKD, and S9 which distinguished the lifelong nature of CKD requiring dietary change, also reached consensus. S13 and 14, which recognised the families need to access credible advice, and CYPs increased exposure to phosphates in processed foods, respectively, also reached consensus (see Table 38).

These findings demonstrate panellist's agreement on the pivotal role of diet in the management of CKD. In particular, the availability and consumption of processed foods rich in inorganic phosphates is recognised as a problem that needs to be addressed early, with better education and support, harnessing buy-in from the whole family.

Statement 12/R2 regarding the 'burden of care', almost reached consensus at 68.2% (see Table 38), however panellists indicated by their rating that small changes were required.

One HCP who was uncertain whether early dietary intervention would 'make it easier later on' seemed to measure the 'burden of care' in terms of a hard or easy load. Explaining that starting dietary change early, would make more dietary changes relatively easier later on when the CYP neared dialysis, and the demands on the family were great: 'they would already be used to' the diet (DEL_12HCP). This was echoed by a parent who thought it would be 'better to change diet early on, than carry on as you have been doing and then have to change diet drastically during the later stages of CKD' (DEL_19PAR). Another parent contended the idea that early changes ease the strain later on, stating 'although early [dietary] introduction will help', as the CKD progressed the increasingly restrictive demands of the diet meant that 'it hasn't fully eased the burden in the later stages' (DEL21PAR).

Table 38. Median LOA rating, % LOA and participant group weighting for the rationale statements in round 2

Category: rationale (S4-15)	Median level of agreement rating	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S4	**4**	**89.3 (H93.75/P83.3)**	**10.7 (H6.25/P16.6)**		
S5/R2	4	81.8 (H69/P100)	18.2 (H31/P0)		
S6	**4**	**75 (H75/P75)**	**21.4 (H25/P16.6)**	**3.6 (H0/H8.3)**	
S7a/R2	4	72.7 (H61.5/P8)	22.7 (H30.74/P1)	4.5 (H7.7/P0)	
S7b/R2	4	77.3 (H61.5/P100)	18.2 (H30.7/P0)	4.5 (H7.7/P0)	
S8/R2	4	77.3 (H69.2/P88.8)	22.7 (H30.7/P11.1)		
S9/R2	4	90.9 (H92.3/P88.8)	4.5 (H7.6/P0)	4.5 (H0/P11.1)	
S10	**4**	**89.3 (H81.25/P100)**	**10.7 (H18.74/P0)**		
S11	**4**	**82.2 (H75/P91.6)**	**17.8 (H254/P8.3)**		
S12/R2	4	68.2 (H61.5/P77.7)	31.8 (H38.4/P22.2)		
S13/R2	4	95.5 (H92.3/P100)	4.5 (H7.7/P0)		
S14/R2	4	86.4 (H92.3/P77.7)	9.1 (H7.7/P11.1)	4.5 (H0/P11.1)	
S15	**4**	**78.6 (H81.25/P75)**	**17.9 (H18.75/P16.6)**	**3.6 (H0/P8.3)**	

S12/R2 was the only statement in the rationale category still to reach agreement in round 3.

Box 26, p. 233 shows how the round 2 comments shaped S12 in round three

Box 26. Rationale S12 round 1-3

Domain:	Statement 12 – why design an EPI?		Very high agreement	High agreement	Moderate agreement	Low agreement
Rationale	Round 1.	Introducing dietary change early, reduces the dietary 'burden of care' in the later stages of CKD				
How **the panel** responded in round 1			50%	39.3%	10.7%	
Rationale	Round 2.	Introducing dietary change early, reduces the dietary 'burden of care' in the later stages of CKD	Very high agreement	High agreement	Moderate agreement	Low agreement
How **the panel** responded in round 2			68.2%	31.8%		
Your Round 3 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after round 2.						
Rationale	Round 3.	Introducing dietary changes early may not reduce the 'burden of care' in the later stages of CKD, but may help to establish a better understanding of diet/lifestyle to inform long term change.				

7.5.5 Round 2 results for the intended user's category

There was consensus that S18/R2, the final statement in the intended users' category, which suggested the EPI should target the main leaders in the family, reached consensus with very high agreement at 86.4% (84.6 and 88% of HCPs and parents, respectively) (see Box 48, Appendix 15).

7.5.6 Round 2 results for the delivery category

There was consensus that statements S20a and b, which considered the best location or forum to offer the EPI, respectively, should support the EPI. S23a and b, which elevated face-to-face as the preferred mode of delivery, supported by written information that can be frequently accessed

for future reference, respectively, also reached consensus (see Table 39). This left only S24/R2, regarding the use of social media as a tool to support the delivery of EPI education, still to reach agreement in this category.

Building on the rationale statements, these findings validate the desire of panellists to have easy of access to EPI dietary advice, whether locally or in the tertiary hospital. A face-to-face consultation with the children's renal dietitian is desired, reinforced by written resources; strengthened by a workshop with other families.

Table 39. Median LOA rating, % LOA and participant group weighting for the delivery statements in round 1 and 2

Category: delivery (S19-27)	Median LOA rating	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S19 when should we deliver EPI?	4	75 (H62.5/P91.6)	17.9 (H25/P8.3)	7.1 (H12.5/P0)	
S20a/R2	4	90.9 (H84.6/P100)	4.5 (H7.6/P0)	4.5 (H7.6/P0)	
S20b/R2	4	86.4 (H77/P100)	13.6 (H23/P0)		
S21 who should deliver the EPI?	4	78.6 (H81.25/P75)	21.4 (H18.75/P25)		
S22	4	82.1 (H87.5/P75)	17.9 (H12.5/P25)		
S23a/R2	4	86.4 (H77/P100)	13.6 (H23/P0)		
S23b/R2	4	81.8 (H77/P88.8)	12.1 (H23/P11.1)		
S24/R2	4	54.5 (H69.3/P33.3)	31.8 (H23/P44.4)	13.6 (H7.7/P22.2)	
S25	4	71.4 (H81.25/P58.3)	28.6 (H18.75/P41.6)		
S26	4	75 (H87.5/P58.3)	14.3 (H12.5/P16.6)	7.1 (H0/P16.6)	3.6 (H0/P8.3)
S27	4	82.1 (H87.5/P75)	14.3 (H12.5/P16.6)	3.6 (H0/P8.3)	

S24/R2 focused on the use of social media as a tool to support the delivery of the EPI (see Box 49, Appendix 15). This statement reached 54.5% (very high agreement) predominantly from HCP (69.2%), with only 33.3% of the parents rating it accordingly. This parental lack of agreement was reflected in the following comments. Recognising that not all parents choose to access social media, one parent felt it 'must be an extra tool rather than an expected tool' (DEL_19PAR). Another parent felt engagement with social media would be intrusive to the privacy of some families, stating 'people are more private about family health than to want to share it on social media' (DEL_26PAR). Finally, another parent felt it would be easier to 'follow' EPI educational updates on Facebook or Twitter, as 'once you have liked a site or follow on Twitter you automatically get the updates, versus logging on to a website' (DEL_23PAR).

7.5.7 Round 2 results for the other considerations category

There was consensus that the remaining statements in the building relationship section, namely S28a, b and c, which focused on the early introduction of the family to the doctor and dietitian, the offer of diet and lifestyle change, all of which will support the development of trusted relationships to improve the effectiveness of dietary advice, respectively, should support the EPI. S29, echoing the importance of building a therapeutic relationship with HCP over time, also reached consensus. Likewise, S30a and b, which acknowledged the importance of HCP making time to understand the child and family needs, and allowing time for the family to come to terms with the CKD diagnosis, respectively, reached consensus (see Table 40, p. 236).

These findings reinforced panellists' desire for early engagement with the doctor and dietitian in order to build trust and efficacy of treatment.

Table 40. Median score, % levels of agreement and participant group weighting for the other considerations statements in round 1 and 2

Category: other considerations (S28-48)	Median score	Very high agreement (4) (%)	High agreement (3) (%)	Moderate agreement (2) (%)	Low agreement (1) (%)
S28a/R2	4	95.5 (H92.3/P100)		4.5 (H7.7/P0)	
S28b/R2	4	95.5 (H92.3/P100)	4.5 (H7.6/P0)		
S28c/R2	4	81.8 (H76.9/P88.8)	9.1 (H15.3/P0)	9.1 (H7.7/P11.1)	
S29/R2	4	86.4 (H92.3/P77.7)	13.6 (H7.6/P22.2)		
S30a/R2	4	81.8 (H92.3/P66.6)	18.2 (H7.6/P33.3)		
S30b/R2	4	81.8 (H84.6/P77.7)	18.2 (H15.3/P22.2)		
S31	4	71.4 (H75/P66.6)	17.9 (H18.75/P16.6)	10.7 (H6.25/P16.62)	
S32/R2	4	81.8 (H84.6/P77.7)	18.2 (H15.3/P22.2)		
S33a/R2	4	86.4 (H84.6/P88.8)	9.1 (H7.7/P11.1)	4.5 (H7.7/P0)	
S33b/R2	4	68.2 (H53.8/P88.8)	27.3 (H38.4/P11.1)	4.5 (H7.7/P0)	
S34/R2	4	77.3 (H77/P7)	9.1 (H15.3/P0)	13.6 (H7.7/P2)	
S35/R2	3	45.5 (H38.4/P55.5)	22.7 (H30.7/P11.1)	13.6 (H23/0)	18.2 (H7.7/P33.3)
S36	4	75 (H75/P75)	25 (H25/P25)		
S37/R2	3	40.9 (H5/P4)	45.5 (H7/P3)	9.1 (H1/P1)	4.5 (H0/P1)
S38/R2	4	81.8 (H69.2/P9)	18.2 (H30.7/P0		
S39/R2	4	81.8 (H77/P88.8)	9.1 (H15.3/P0)	4.5 (H0/P11.1)	4.5 (H7.7/P0)
S40/R2	4	90.9 (H92.3/P88.8)	4.5 (H0/P11.1)	4.5 (H7.7/P0)	
S41/R2	4	81.8 (H77/P88.8)	4.5 (H7.7/P0)	4.5 (H7.7/P0)	9.1 (H7.7/P11.1)
S43/R2	4	90.9 (H84.6/P100)	9.1 (H15.3/P0)		
S46/R2	4	77.3 (H77/P77.7)	13.6 (H15.3/P11.1)	9.1 (H7.7/P11.1)	
S48/R2	4	72.7 (H69.2/P77.7)	27.3 (H30.7/P22.2)		
S49/R2	4	77.3 (H77/P77.7)	22.7 (H23/P22.2)		
S50/R2	4	77.3 (H70/P88.8)	18.2 (H30/P0)	4.5 (H0/P11.1)	
S51/R2	4	90.9 (H100/P77.7)	9.1 (H0/P22.2)		
S52/R2	4	95.5 (H100/P88.8)	4.5 (H0/P11.1)		

Statement S35 and S37 shaded in grey were removed and superseded by S36

Of the 'other the considerations' statement, that addressed support, S32 which recognised the need for parents to access peer support from other parents, reached consensus. Likewise, S33a and S34, which suggested the use of an EPI local support network, and the value of patient stories and shared experiences to CKD management, respectively, also reached consensus (see Table 40). Leaving only S33b in this category, which suggested an annual education meeting as a forum to support the EPI (see Box 27, p. 237). S33b required one more panellist to rate it as 4, in order to reach consensus. Panellists rating it with very high agreement were predominantly parents (88.8%) with 53.8% agreement from HCPs (see Table 40). The only comment received on S33b was from the one parents who did not rate it with a 4. He felt, in order to appreciate the potential benefits of an annual EPI education meeting, it was necessary to gain clarity on 'what it aims to deliver and communicate' (DEL_23PAR).

These findings complement those from the rationale and delivery category, reinforcing the desire of parents to reach other families using an 'early support platform'.

Box 27. Other considerations: support S33b

Domain:	**Statement 33 was revised into 33a and 33b in Round 2 – statement 33b did not reach consensus**		**Very high agreement**	**High agreement**	**Moderate agreement**	**Low agreement**
Other considerations	**Round 1. 33.**	An EPI support group should be set up for parents across the shared-care renal network e.g. online or face to face in the clinic				
How **the panel** responded in round 1			**57.1%**	**25%**	**14.3%**	**3.6%**
Other considerations	**Round 2. 33b.**	For those who would choose to access it, an annual EPI education meeting should be set up for parents, children and young people.	**Very high agreement**	**High agreement**	**Moderate agreement**	**Low agreement**
How **the panel** responded in round 2			**68.2%**	**27.3%**	**4.5%**	
Your round 3 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after round 2						
Other considerations	**Round 3. 33b.**	For those who would choose to access it, an annual EPI education workshop should be set up for parents, carers, children and young people.				

In the SM section, S35 and S37/R2 describing the medical-model and self-agency SM model failed to reach consensus, with 45.5 and 40.9%, respectively (see Table 40**,** p. 236). In view of the strong opposition to these statements (see section 7.4.5), in consultation with the Delphi Steering Group, these statements were removed and superseded by S36, which advocated a 'partnership' or 'joint directive' model, reaching consensus in round 1.

In the section covering the early tests undertaken in the out-patient clinic, the remaining S38 and 39 reached consensus, both achieving 81.8% very high agreement (see Table 40, p. 236). S40 and 41/R2 in the early dietary blood tests section (see Table 40), also reached consensus with 90.9 and 81.8%, respectively.

These findings highlight panellists' recognition and readiness for early dietary intervention despite the lack of a specific early nutritional phosphate blood test. Currently available blood tests play a powerful role in supporting adherence to treatment and should be used to flag up early-stage patients who may benefit from EPI.

In the potential concerns section, S43, which recognised the potential of the EPI to create anxiety, and S46, which acknowledged cooking skills as a limiting factor, both reached consensus. S47 which advocated that the dietitian should have a greater presence in the shared-care clinics, and S48, which recognised isolation as a potential concern, likewise reached consensus.

These potential concerns findings speak to those from the rationale and delivery categories; by providing the family with a positive focus, early EPI education and support through early engagement consultations and workshops would address issues of anxiety, isolation and cooking skills set.

The final four statements 49, 50, 51and 52/R2, developed in response to new ideas not previously captured, addressed the development of educational/cookery workshops, offered at a venue close to the family home; the involvement of families in the development of educational resources; and the opportunity for HCP to learn about, and thus reinforce the EPI message, respectively, all reached consensus (see Table 40)**.** Statement 42, which proposed the 'burden' of the EPI may be ameliorated by targeted education and support, still did not reach consensus. In fact, the consensus was little different from round 1, despite revisions (see Box 28, p. 239). Reflecting again on the free-text comments from round 1, participants felt diet was not the source of the burden. Rather, it was confronting the need to change that was the key burden. Simple education and support to make the necessary adjustments would reduce the onerous nature of the task. These factors shaped the revised S42 in round 3.

Box 28. Other considerations: potential concerns S42

Domain:	Statement 42 - potential concerns		Very high agreement	High agreement	Moderate agreement	Low agreement
Other considerations	Round 1.	It is important to recognise that choosing to follow an EPI diet may be a burden to some families				
How **the panel** responded in round 1			42.9%	39.3%	7.1%	10.7%
Other considerations	Round 2.	With the right level of education and support, choosing to follow the EPI diet should not become a burden to the family	Very high agreement	High agreement	Moderate agreement	Low agreement
How **the panel** responded in round 2			45.4%	36.4%	18.2%	
Your round 3 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after round 2.						
Other considerations	Round 3.	Choosing to follow the EPI diet requires family change, which is not easy, and can be a burden. The right level of education and support would help families to make sense of the diet and support change.				

Likewise S44, while acknowledging the CYP's dietary needs, recognised that it did not have to limit future aspirations, also failed to reach consensus. No new comments were received for S44/R2, but in order to separate important components of the statement, two sub-statements were created to capture the strength of opinions expressed (see Box 29, p. 240).

These additional comments findings again reinforced panellists' desire for an 'early support platform' through local educational food preparation workshops. The potential 'burden' associated with EPI remains an important issue, with costs and 'labelling of the CYP' highlighted as perceptible factor. If the whole family is to follow the EPI, not differentiating the CKD CYP, it must be accessible, practical and affordable.

Box 29. Other considerations: potential concerns S44

Domain:	Statement 44 - potential concerns		Very high agreement	High agreement	Moderate agreement	Low agreement
Other considerations	Round 1.	It is important to recognise that following the EPI diet may 'label' the child or young person, limiting family expectations of their child or young person				
How **the panel** responded in round 1			25%	21.4%	25%	28.6%
Other considerations	Round 2.	Following the EPI diet may identify the child or young persons as having different dietary needs. This does not have to limit their own, and their family expectations as they grow up.	Very high agreement	High agreement	Moderate agreement	Low agreement
How **the panel** responded in round 2			59%	22.7%	18.3%	
Your round 3 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after round 2 into 2 separate statements.						
Other considerations	Round 3. 44a.	Following the EPI diet may identify the child or young persons as having different dietary needs.				
Other considerations	Round 3. 44b.	Following the EPI diet does not have to limit their own, or their family expectations as they grow up.				

S45/R2, which addressed the value of support in reducing the intrusive impact of the EPI, still required agreement. No new comments were received for this statement, but after consultation with the Delphi Steering Group and reflection on round 1 comments (see section 7.4.5), two new sub-statements were created for round 3 which divided what were effectively two statements in S45/R2 in order to identify where agreement could be found (see Box 30, p. 241).

Box 30. Other considerations: potential concerns S45

Domain:	Statement 45 - potential concerns		Very high agreement	High agreement	Moderate agreement	Low agreement
Other considerations	Round 1.	It is important to recognise that following the EPI diet may be intrusive to family life				
How **the panel** responded in round 1			32.2%	35.7%	21.4%	10.7%
Other considerations	Round 2.	Following the EPI program will require the child or young person and their family to adapt to the necessary dietary changes. This may take time, but with support it does not need to be intrusive to family life.	Very high agreement	High agreement	Moderate agreement	Low agreement
How **the panel** responded in round 2			59.1%	27.3%	13.6%	
Your Round 3 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after round 2 into 2 separate statements.						
Other considerations	Round 3. 45a.	Following the EPI program will require the child or young person and their family to adapt to the necessary dietary changes and can take time.				
Other considerations	Round 3. 45b.	Adequate support to manage the EPI diet may reduce intrusion upon family life.				

Six statements from the other considerations category went through to round 3 for further rating by panellists; two were revised (S33b and S42), and two were made into four sub-statements (44a and b, and 45a and b).

7.5.8 Stability of agreement across round 1 and 2

Five statements (S8, 12, 34, 40 and 47) remained the same between R1 and R2, with no changes to the wording. Weighted Cohen's Kappa (1968) was calculated to determine interrater (between HCP and parents) reliability across the 2 rounds (see Table 41), showing the extent to which raters disagree (the non-diagonal frequencies) and agree (along the diagonal) (Petrie and Sabin, 2009). A Kappa statistic of '1' defined perfect agreement - where there was no change in the LOA rated. A zero indicated a large interrater variation between rounds, such that agreement was no better than chance (Landis and Koch, 1977) (see Table 41).

Table 41. Kappa statistic (Landis and Koch 1977)

Kappa statistic	Agreement
<0.0	Poor agreement
0.0- 0.20	Slight agreement
0.21- 0.40	Fair agreement
0.41-0.60	Moderate agreement
0.61-0.80	Substantial
0.81-1.0	Almost perfect agreement

Table 42, p. 243 details the results of the Cohen's Kappa for S8, 12, 34, 40 and 47, demonstrating an overall interrater reliability across rounds 1 and 2 of moderate to substantial. S40 had a Kappa = 0.3 (<p=0.085), 95% CI (0.000-0.519) demonstrating a fair interrater reliability that was not significant. Refer to Appendix 16 for the cross tabulation frequencies.

Table 42. Results of Cohen's Kappa interrater reliability analysis

Statements	Weighted kappa	Agreement	*p*-value	Kappa (95% CI)
S8	0.7	Substantial	.0001	(0.473-1.0)
S12	0.6	Moderate	.0001	(0.512-0.900)
S34	0.7	Substantial	.0001	(0.663-0.901)
S40	0.3	Fair	.085	(0.000-0.519)
S47	0.5	Moderate	.003	(0.205-0.531)

7.5.9 Summary of Delphi round 2

During Delphi round 2 another EPI definition reached consensus. The term 'healthy-eating', 'kidney health' and the 'burden of care' associated with EPI continued to provoke a response from panellists. The rationale for EPI gained clarity, with a focus on the need to reduce processed foods through proactive education and support. EPI provides a positive role for parents, with buy-in from the whole family. EPI should target the main leaders of change in the family. Early education and support should be delivered face-to-face in a location that best suits the family, using a workshop format which provides opportunity to network with other families, and build trusted therapeutic relationships with HCPs. Although social media was valued, it should be optional and requires supervision. Owing to strong opposition, and in consultation with the Delphi Steering Group, S35 and S37 advocating a medical and self-agency model, were removed, in favour of the 'partnership' model of SM. The desire for an early dietary blood test to direct and support dietary management reached consensus. Limited cooking skills, isolation, anxiety and singling out CYP as different, were acknowledged as potential concerns of EPI.

7.6 Results of statements in round 3

This section reports the development and findings of the round 3 Delphi survey.

7.6.1 Development of the round 3 survey

The outcome of the statements at the start of round 3 are listed in Table 43, p. 244.

Table 43. Statements at the end of round 2

Statements that achieved consensus	Statements removed	Statements that were revised or separated	New sub-statements
S3, 5, 7a, 7b, 8, 9, 13, 14, 18, 20a, 20b, 23a, 23b, 28a, 28b, 28c, 29, 30a, 30b, 32, 33a, 34, 38, 39, 40, 41, 43, 46, 47, 48, 49, 50, 51 and 52	S35 and 37 *(superseded by S36 see Table 42 in section 9.4.5)*	S12, 24, 33b, 42, 44a, 44b, 45a and 45b	S2a and S2b

Ten statements/sub-statements (shaded in grey) remained to be rated by the panel in round 3 (see Figure 19, p. 226).

7.6.2 Round 3 response rate

On the 5th December 2018, round 3 surveys were distributed by email or posted to the 22 participants (13 HCPs, 9 parents) who completed and returned the round 2 survey, with planned closure on the 19th December before the Christmas break. In total 21 round 3 surveys were returned (13 HCPs, 8 parents), only one parent failed to return the survey, yielding a response rate of 95% (see Table 44, p. 245). This high response rate in the third and final round suggested that the reductionist approach (section 6.8.6) had proved effective in retaining interest and achieving a rapid response time. Additionally, it suggested that the 21 experts who participated were highly committed to the study topic (Fefer et al. 2016).

All regions of the EMEESY network were represented by the returns. For a consort flow diagram detailing the recruitment and response rates across each Delphi round refer to Figure 18, p. 206.

Table 44. Summary of Delphi round 3 responders and non-respond by region

EMEESY network region	Delphi round 3 surveys sent out	Delphi round 3 surveys returned
East Midlands	12 HCP (10 based at the tertiary centre) 7 parents	12 HCP (10 based at the tertiary centre) 6 parents
East of England	1 HCP 1 parents	1 HCP 1 parent
South Yorkshire	0 HCP 1 parents	0 HCP 1 parent
Total	22 (13 HCP [59%] 9 parents [41%])	21 (95.4%) (13 HCP [62%] 8 parents [38%])
% Response/primary role		HCP = 100% Parents = 88.8%

Similar to round 2, the round 3 cohort comprised more HCP than parents, and more females than males (see Table 45).

Table 45. Primary role and gender of round 3 participants

Primary role		**Participant group Delphi round 3 responders**	**Gender**	**Percent**
Parent/carer		8 (38%)	8 (1 male)	38
HCPs	Paediatric nephrologist/paediatrician with an interest in nephrology	13 (59%) Ratio of doctors: nurse/dietitians (7:6)	7 (4 males)	33.3
	Children's renal nurse		2 (0 male)	9.7
	Dietitian		4 (0 males)	19
Total			21 Male 5 (23.8%) Female 16 (76.2%)	100

The following colour key applied to all tables detailing the Delphi round 3 results.

Green shaded box denotes ≥70% of panelist rated the statement as very high agreement, achieving a consensus response to the statement.

In addition to the S number, round three is abbreviated as R3 e.g. statement 24 round 3 is S24/R3

7.6.3 Round 3 results for the definition category

Both of the two remaining sub-statements in the definition category, which sought to capture the phosphate management and healthy-eating elements of EPI (S2a/R3 and S2b/R3) (see Table 46, p. 247) failed to reach consensus. This reflected the panellists' disagreement and lack of clarity regarding the association of 'healthy-eating' with 'kidney healthy'. Only one comment was received in round 3 for S2a and b, which expressed a preference for the term 'healthy kidneys' rather than 'kidney health'. This comment directly opposed the majority of comments in round 2, where both HCP and parents disagreed with the implication of the statements, believing that by nature of having CKD, the kidneys cannot be healthy (see section 7.5.3).

For those statements that did not reach agreement in round 3, inferential statistics were used to assess if differences in the within-round ratings between the parents and HCP's were statistically significant. To correct for the small sample size in the study, where expected frequencies in some cells were <5, Fisher's exact test was used (Petrie and Sabin, 2009). The level of significance was set at $p<0.05$ (Petrie and Sabin, 2009). Levels of agreement were 'very high agreement', 'high agreement', 'moderate agreement' and 'low agreement'.

Table 46. Median LOA rating, % LOA and participant group weighting, and Fisher's Exact test for the definition S2a and b in round 1-3

Category: definition (S2)	Median level of agreement rating	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)	Fisher's Exact Test
S2	3	21.5 (H25/P16.6)	32.2 (H43.75/P16.6)	28.5 (H31.25/P25)	17.8 (H0/P41.6)	*p=0.037
S2/R2	3	45.5 (H53.84/P33.3)	31.8 (H23/P44.4)	4.5 (H0/P11.1)	18.2 (H23/P11.1)	p=0.390
S2a/R3	The EPI program should be labelled as 'early phosphate management and healthy eating for kidney health'					
	3	47.6 (H37.5/ P33.3)	19 (H6.25/P25)	19 (H18.75/P11.1)	14.3 (H18.75/P0)	p=0.335
S2b/S3	The EPI program should be labelled as 'early phosphate management and healthy eating for CKD'					
	3	42.9 (H38.4/P50)	52.4 (H53.8/P50)	4.8 (H7.7/P0)		p=1.0

*p=statistical significance

For S2 in round 1, the stakeholder group did have an effect on the LOA rating (p=0.037); parents were significantly more likely than HCP to rate S2 as low agreement, affirming the misleading use of the term 'healthy-eating' in the early CKD context. In round 2 and 3, for the revised S2 there was no significant difference between the parents and HCPs within-round ratings (see Table 46).

7.6.4 Round 3 results for the rationale category

Similar to R2, S12/R3 regarding the 'burden of care' of EPI was close to reaching consensus, with 66.7% very high agreement rating (see Table 47). Two HCP comments, disagreeing with the wording of this statement, thought it was 'less powerful' and 'diluted', which affirmed the value of early dietary phosphate control, but also recognising that the term 'burden of care' has a 'negative connotation' which families may not like, especially those coming to term with a new diagnosis of CKD. A Fisher's Exact test showed no statistically significant difference for S12 in round 1, 2 and 3, between the parents' and HCPs' and their within-round rating (Table 47).

Table 47. Median LOA rating, % LOA and participant group weighting, and Fisher's Exact test for the rationale S12 in round 1 and 3

Category: rationale (S12)	Median LOA rating	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)	Fisher's Exact Test
S12	3.5	50 (H37.5/P66.6)	39.3 (H50/P25)	10.7 (H12.5/P8.3)		p=0.306
S12/R2	4	68.2 (H61.5/P77.7)	31.8 (H38.4/P22.2)			p=0.648
S12/R3	Introducing dietary changes early may not reduce the 'burden of care' in the later stages of CKD, but may help to establish a better understanding of diet/lifestyle to inform long term change.					
	4	66.7 (H61.5/P75)	19 (H23/P12.5)	9.5 (H7.6/P12.5)	4.8 (H7.6/P0)	p=1.0

7.6.5 Round 3 results for the delivery and other considerations category

There was consensus that S24/R3 (see Box 50, Appendix 15), in the delivery category regarding the use of social media, should support the EPI, reaching very high agreement rating at 90.5%. This affirmed the panellist's desire for an online 'early support network'. There was also consensus that S33b/R3 (see Box 51, Appendix 15), in the other considerations category, regarding the development of an annual EPI education workshop, should support the EPI, reaching very high agreement rating at 85.7%. There was consensus that S42/R3 (see Box 52, Appendix 15), in the other considerations: potential concerns category, regarding the use of education and support to alleviate any EPI dietary burden, should support the EPI, reaching very high agreement rating at 71.4%. S44a/R3 (see Table 48) in the other considerations: potential concerns category, which raised the issue of the CYP following the EPI as having different dietary needs, failed to reach consensus, with a very high agreement rating of 57.1%. Only one comment was offered for this statement, affirming that early stage CKD patients 'have different dietary needs and need to be made aware of them to stay healthy', however these meals 'can be enjoyed by all the family without necessarily being thought of as renal meals' (DEL_22PAR). Fisher's Exact test showed no statistical significance for S44 in R1-3, between the parents and HCP and their chosen rating (see Table 48).

Table 48. Median LOA rating, % LOA and participant group weighting, and Fisher's Exact test for the other considerations: potential concerns S44a and b in round 1-3

Category: other considerations (S44a and b)	Median LOA rating	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)	Fisher's Exact Test
S44 (potential concerns)	2	25 (H31.25/P16.6)	21.4 (H25/P16.6)	25 (H31.25/P16.6)	28.6 (H12.5/P50)	p=0.215
S44/R2	4	59 (H46/P77.7)	22.7 (H23/P22.2)	18.3 (H31/P0)		p=0.838
S44a/R3	Following the EPI diet may identify the child or young persons as having different dietary needs.					
	4	57.1 (H53.8/P62.5)	33.3 (H38.5/P25)	9.5 (H7.6/P12.5)		p=0.830
S44b/R3	4	71.4 (H69.2/P75)	14.3 (H23/P0)	14.3 (H7.6/P25)		

There was consensus that S44b/R3 (see Table 48, p. 249), regarding concerns that the EPI may limit the CYP expectations as they grow up, should support the EPI, reaching very high agreement rating at 71.4%.

There was consensus that S45a and b/R3 (see Box 53, Appendix 15), regarding the adaptation to change and the intrusion to family life, should support the EPI, reaching a very high agreement rating at 85.7% and 71.4%, respectively. A final comment from a HCP in support of S45b, stated 'people misunderstand EARLY phosphate intervention. It is not a restriction. It is about healthy choices' (DEL_09HCP), seemed to capture a key message from the Delphi survey.

7.6.6 Summary of Delphi round 3

The findings form round 3 highlighted a need to be cautious with the us eof the term 'healthy-eating', and with the suggestion that the EPI may alleviate the later 'burden of care'. They also emphasised the need for clarity and a reframing of the role of healthy-eating and EPI in the early management of kidney health. Although the EPI was recognised as a potential 'burden', the term conferred different messages to different people. Once the burden of 'change' rather than diet, was acknowledged, and the need for tailored education and support was incorporated, S42 reached consensus. An online platform is desired as part of the 'early support network', and the EPI needs to avoid singling out the CYP with CKD as having different needs, rather with long-term education and support, the program seeks to encourage the whole family to adopt healthier dietary choices.

The next section details the statements that reached consensus across the 3-round DS.

7.6.7 Statements reaching consensus across Delphi rounds 1-3

The following Table 49, p. 251 details the statements that reached consensus during Delphi rounds 1-3.

Table 49. Statements reaching consensus

S1	Early phosphate intervention (EPI) is a program of dietary education, engaging with children, young people and their families during the early stages of chronic kidney disease (CKD 2-3), to support diet and lifestyle change towards early dietary phosphate SM
S2a (48%)	The EPI program should be labelled as 'early phosphate management and healthy eating for kidney health'
S2b (43%)	The EPI program should be labelled as 'early phosphate management and healthy eating for CKD'
S3	The EPI program will encourage a diet for healthy kidneys, with a reduced intake of processed foods (which are high in added phosphate), parents and carers will be empowered to prepare home-cooked meals containing meat, fish and dairy, with an increase in fruit, vegetables, grains and nuts.
S4	Diet has a crucial role to play in the management of a chronic disease
S5	Processed foods can be high in salt and contain added phosphates; to support the management of many chronic diseases, advice should be given to consume less processed foods, with suggestions regarding nutrient-rich alternatives.
S6	The provision of early dietary education gives children, young people and families a tool to more confidently manage their own diet during the early stages of a chronic disease
S7a	When a child is diagnosed with a chronic disease, knowing and understanding the impact of diet on the disease may motivate families to make diet/lifestyle changes
S7b	During the early stages of a chronic disease, diet/lifestyle change can provide a positive focus for the family, knowing that they are making an important contribution to disease management.
S8	Diet is one of the hardest parts of CKD, introducing the diet early would help to make it the norm for the whole family.
S9	Chronic kidney disease is a lifelong condition, which is managed by both medication and diet.
S10	Dietary phosphate education is a long-term process and should begin in the early stages of CKD.
S11	Children and parents need to be told the impact of diet from an early stage, and then they can make an informed choice.
S12 (67%)	Introducing dietary changes early may not reduce the 'burden of care' in the later stages of CKD, but may help to establish a better understanding of diet/lifestyle to inform long term change.
S13	From diagnosis, parents, children and young people should have access to helpful and credible information about diet and CKD

S14	Due to the availability of processed foods, children and young people with early stage CKD are at risk of eating large amounts of phosphates as these are added to processed foods in the form of food additives and preservatives.
S15	Early dietary advice should be viewed as important as the medical treatment
S16	Early phosphate intervention should be offered to all children and young people with early stage CKD (2-3) who are likely to progress to the later stages of CKD (4-5)
S17	The EPI program should be directed at the whole family.
S18	The EPI program should target the main leaders of diet and lifestyle change in a family.
S19	The EPI program should be offered at, or close to the diagnosis of CKD.
S20a	The EPI program should be delivered in a location that best suites the family, this may be the home, a shared-care renal clinic close to home, hospital ward or another suitable venue.
S20b	During the early stages of CKD, parents, children and young people could be invited to attend a group session with other families close to their home, to learn about the EPI program.
S21	Dietary information/education always carries much more authority when it comes from people who have expertise in nutrition.
S22	A children's dietitian (ideally with training in the management of CKD) is the best person to deliver the EPI program.
S23a	The EPI program should ideally be delivered initially during a face to face consultation with an expert in children's renal nutrition. This would affirm understanding of the advice and provide an opportunity for the family to ask any questions.
S23b	After a face to face conversation with an expert in children's renal nutrition, written dietary phosphate information (including recipe information) is vital, and provides a useful reference that can be frequently accessed at home.
S24	The use of social media (e.g. EMEESY, Twitter or Facebook) in the EPI program would provide users with a useful additional tool and source of information or support.
S25	The EPI program needs to be delivered using a clear, simple, realistic and positive message.
S26	The EPI program needs to fit within a family's household budget.
S27	The EPI program needs to be flexible so that it can be tailored to meet the changing needs of the child or young person and their family.
S28a	As part of the early management of CKD the family should be introduced to the doctor and dietitian.
S28b	Diet and lifestyle information, and ongoing support and advice, should be offered as early in the diagnosis as possible.
S28c	Early introduction to the dietitian means trusted relationships will develop over time, making the nutritional message more effective.
S29	It is important that a therapeutic relationship built on trust and respect, empathy and understanding, is established over time between the family and healthcare professional.
S30a	It is important for the healthcare professional to understand the individual and family needs.
S30b	It is important for the family to be given time to come to terms with the diagnosis, and what this means for them and their child or young person.
S31	Ongoing feedback and support from healthcare professionals are essential to encourage adherence to diet and lifestyle change
S32	Parents of children with early stage CKD should have access to support from other parents who are in or have been in their situation.
S33a	For those who would choose to access it, an EPI support network should be set up for parents, children and young people in their local area or outpatient clinic.

S33b	For those who would choose to access it, an annual EPI education workshop should be set up for parents, carers, children and young people.
S34	Patient stories and shared experiences are vital, they provide reassurance and make a difference to how a person feels they are able to manage their diet/lifestyle change.
S35	In view of the consensus reached for statement 36, this statement was closed.
S36	The healthcare professional should work in partnership with the child or young person and their family in order to tailor the EPI program to the changing needs of the individual
S37	In view of the consensus reached for statement 36, this statement was closed
S38	Blood results, where they can be explained clearly to families and used to guide management, can act as powerful reinforcements to support adherence to diet and lifestyle change.
S39	Currently available tests which give an understanding of disease progression, such as parathyroid hormone (PTH), blood pressure and protein in the urine, should be explained to parents, and used to identify those children with CKD 2-3 who may benefit from receiving the EPI program.
S40	Even though an early blood test to support phosphate dietary management is not currently used in out-patient clinics, early dietary phosphate education is essential for all children with progressive CKD.
S41	If a blood test which gives an understanding of phosphate control, becomes available for use in the early stages of CKD, it needs to be something that clearly informs the healthcare professional and family of the impact of dietary change.
S42	Choosing to follow the EPI diet requires family change which is not easy, and can be a burden. The right level of education and support would help families to make sense of the diet and support change.
S43	Having kidney disease creates anxiety. With the right level of dietary education and support, the EPI program would give the family a positive focus and a way to help their loved one.
S44a (57%)	Following the EPI diet may identify the child or young persons as having different dietary needs.
S44b	Following the EPI diet does not have to limit their own, or their family expectations as they grow up.
S45a	Following the EPI program will require the child or young person and their family to adapt to the necessary dietary changes and can take time.
S45b	Adequate support to manage the EPI diet may reduce intrusion upon family life.
S46	The EPI program will encourage more home-cooked meals. Some families may benefit from cookery workshops to inspire and support them to follow the program.
S47	In order to deliver the EPI diet, children's dietitians needs to have more of a presence in the shared-care clinics
S48	With the right level of education and support during the early stages of CKD, children and young person and their parents should not feel isolated.
S49	The provision of EPI workshops for families (including carers and grandparents) could provide information to manage CKD in different age groups, advance their cooking skills and provide social support.
S50	EPI workshops could be offered at local schools/shared-clinics/or other suitable venues close to the family home.
S51	EPI educational material (including recipe books) should be developed and updated with the support of families.
S52	Medical, nursing and other healthcare professionals need to be given opportunity to learn about the EPI program, so that they can reinforce the message.

56 out of 62 statements reached consensus

Statements S35 and S37 were removed

Statements S2a, S2b S12 and S44a did not reach consensus

7.7 Summary

This chapter has presented the results of the EPI Delphi survey for round 1-3. Key findings to emerge are that long-term the HCPs and parents recognise the vital and empowering role that diet plays in the management of CKD. There is a readiness for an EPSM strategy. Early engagement with the doctor and dietitian would build trust and efficacy of treatment, advocating a partnership dietary self-management model. EPI education and support need to be offered in the diagnosis period in the form of an initial renal dietetic face-to-face consultation, strengthened by accessible resources and cookery workshops close to home or in the tertiary hospital. These educational workshops, promoting buy-in from the whole family would provide opportunity to meet other families and HCP, and address issues of anxiety, information overload, isolation and cooking skills set. An online support network would give further access to resources and connections, consolidating education and support. Currently available blood tests play a powerful role in supporting adherence to treatment and should be used to flag up early-stage patients who may benefit from EPI. The Delphi survey highlighted the value of free-text comments, and the importance of giving them due attention, particularly if they are voicing a nuanced or deviant viewpoint. For example, across the rounds thecommented highlighted a need to be cautious with the use of the term 'healthy-eating'. Definition S2a and 2b, by failing to reach consensus, elevated the importance of this key issue, and the need for clarity and a reframing of the role of healthy-eating and EPI in the early management of kidney health, which directly informed key novel findings from this MMR study (see section 9.2.1 and 9.2.2).

Next, the key findings of the systematic review, semi-structured interviews and Delphi survey, presented in chapter 3, 5, and 7, respectively, will be integrated in chapter 8.

Chapter 8: Integration of the findings from Phase 1-3

8.1 Introduction

The findings and results from the qualitative and quantitative phases of this research study were provided in Chapter 3, 5 and 7 of this thesis. This chapter draws on these empirical research outputs, providing an integration of the findings to answer the research question and address the objectives that formed the foundation for this study. The five core tenets of Lorig *et al's* (2003) SM theoretical framework (Table 5, p. 16) underpinned these integrative findings.

The integration of the data or 'conversation between the qualitative and quantitative components' is considered to be the most important aspect of any mixed-methods study, as this is where the two approaches become 'greater than the sum of the parts' (O'Cathain et al. 2010; Farquahar et al. 2011). As informed by Andrew and Halcomb (2009), and Tashakkori and Creswell (2007), in this doctoral study the term integration was used, as opposed to synthesis. These terms are often used synonymously in the literature.

8.2 Response to research question and objectives

Firstly, the main research question and the seven research objectives (see Box 31, p. 256), which delineate the scope of this research study, are revisited. The research question and each of these objectives will be addressed in turn, using empirical research findings from all three phases of the investigation to respond.

Box 31. The seven research objectives

1. To achieve a consensus definition for Early Phosphate Intervention (EPI)
2. To explore and gain consensus on how we should deliver the dietary phosphate education, skills and support to children, young people and their families
3. To explore and gain consensus on who should deliver the EPI education, skills and support
4. To explore and gain consensus on who should receive the EPI strategy
5. To explore and gain consensus on when the EPI education and support should be delivered
6. To explore and gain consensus on where the EPI education, skills and support should be delivered
7. To identify the barriers and enablers to implementing an EPI strategy

8.2.1 Overview of the key stakeholders' perspectives towards the development of an EPI

The primary research question that determined the seven principal objectives of this thesis was:

> *What are the perspectives of children and young people, parents and key healthcare professionals towards the development of an early dietary phosphate SM strategy, to delay disease progression for children with CKD stages 1-3, within a managed paediatric renal network?*

This question sought to navigate the study towards the exploration of key stakeholders' perspectives regarding the development of an EPSM strategy, to delay disease progression, for CYP with early stage CKD. By gaining a multiple stakeholders' perspective, this study provided an integrated viewpoint on the how, who, when and where of developing and delivering an EPSM strategy, with identification of the challenges and enablers to its implementation.

Establishing a platform to learn from empirical research, Phase 1 - the literature review and mixed-studies systematic review with a narrative synthesis, exposed a wealth of insight from the experience and views of CYP, parents and other stakeholders engaged in dietary interventions to manage a range of chronic diseases.

The principal Phase 1 findings, presented in Chapter 3, were categorised under four themes. Most notably, the themes of *education, parents/caregivers, early dietary SM,* and *challenges and enablers* featured as key elements to dietary intervention strategies for chronic disease. Aspects of the research objectives are addressed by these themes.

Before presenting the integration and discussion, it is important to reiterate that the findings in this research study emerged from the literature review, and systematic review narrative synthesis, and QUALITATIVE interview themes, corroborated by the QUALITATIVE free-text comments and statements reaching QUANTITATIVE statistical agreement in the three rounds of the DS. The findings were not mapped to the study in an *a priori* manner. As such, the order in which they appear in this chapter will not be chronological; rather the findings and proposals will be introduced according to the constructs being discussed in response to addressing each research objective.

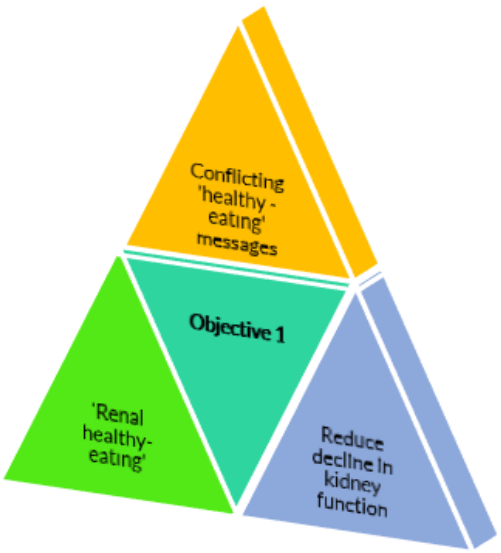
8.2.2 Objective 1: Achieve a consensus definition for EPI

This section addresses the first research objective (RO) for this study.

RO1: To achieve a consensus definition for EPI

In response to the objective, the focus is on those statements that reached consensus; although some of the nuances provided by lower agreement are also considered. Figure 20, p. 258 captures the themes for objective 1 and each theme will be presented in turn overleaf.

Figure 20. Objective 1 themes



As informed by the CREDES framework (Jünger et al. 2017), this objective sought to pragmatically describe the EPI, identifying its target audience and main purpose. The objective was addressed primarily during the quantitative Delphi phase with a consensus agreement for 2 of the 3 definition statements. The construction, refinement and redefining of the three definition statements (S1-3) (refer to section 7.2.1) captured elements of the literature from Phase 1 and the qualitative findings from Phase 2. Participants had co-constructed an EPI definition capturing the early vital role of diet and lifestyle changes to promote kidney health.

The EPI definition statements 1 and 3 reached ≥70% consensus agreement during Delphi round 1 and 2, as shown below, p. 259.

Category: definition	Median LOA rating (1-4)	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S1/R1	Early phosphate intervention (EPI) is a program of dietary education, engaging with children, young people and their families during the early stages of chronic kidney disease (CKD 2-3), to support diet and lifestyle change towards early dietary phosphate SM				
	4	71.4 (H68.75/P75)	25 (H31.25/P16.6)		3.6 (H0/P8.3)
Category: definition	**Median LOA rating (1-4)**	**Very high agreement (4)**	**High agreement (3)**	**Moderate agreement (2)**	**Low agreement (1)**
S3/R2	The EPI program will encourage a diet for healthy kidneys; with a reduced intake of processed foods (which are high in added phosphate), parents and carers will be empowered to prepare home-cooked meals containing meat, fish and dairy, with an increase in fruit, vegetables, grains and nuts.				
	4	77.3 (H77/P77.7)	22.7 (H23/P22.2)		

NB. Sections shaded in yellow show where definition consensus was reached.

Continuing with RO1, the term 'healthy-eating' proved contradictory.

Healthy-eating portrays conflicting messages

Inclusion of the label 'healthy-eating' in two of the original round 1 EPI definition statements (see S2/R1 and S3/R1 below) (refer to section 7.4.1), generated multiple diverse responses.

S2/R1	Early phosphate intervention (EPI) should be labelled 'early intervention healthy eating'
S3/R1	The EPI diet primarily encourages a healthy eating diet, with a reduced intake of processed food, promotion of home-cooked meals containing meat, fish and dairy, and an increase in plant-based foods such as fruit, vegetables, grains and nuts.

Although not dichotomous, the expert panel of parents and HCP expressed conflicting opinions on this label. For many parents, early phosphate intervention 'healthy-eating' seemed paradoxical. For example, they understood healthy-eating to encourage a greater consumption of fruit and vegetables, but the renal diet restricted a lot of high-potassium fruits and vegetables. Parents understood their children required high-energy meals and snacks to optimise growth, but healthy-eating

discouraged these foods. The conventional message of decreased saturated fats, refined sugar and salt, increased fruit, vegetables and fish, and the avoidance of high-calorie meals and snacks was reportedly causing confusion, portraying a misleading message.

> *'I think you have to be careful about labelling CKD diet "healthy-eating". I thought I was providing "healthy-eating" by giving my baby steamed vegetables and lots of fresh fruit which was completely the opposite of what was required once he was diagnosed with CKD.'*
>
> ***(DEL_19PAR)***

The confusion lay in the fact that parents assumed the restriction of potassium-rich fruit and vegetables, and the promotion of high energy/nutrient-dense foods prescribed during the later stages of CKD was appropriate for the early stages of CKD. During the early stages when renal impairment is low to moderate, and biochemistry is usually stable, these dietary restrictions are not routinely necessary (Shaw, 2015). Because parents of CYP with early stage CKD do not consistently receive dietary advice, there is a lack of communication between HCP and parents, and thus a lack of parental knowledge of the most beneficial food decisions during this important early period.

Renal-healthy-eating

By contrast, HCP defended the EPI healthy-eating message as relevant to patients with all stages of CKD; indeed its suitability for society's health.

> *'This [EPI diet] is a way of…framing it in a very positive way…it is very much promoting a healthy diet. You know I think there's a big focus on phosphate within that, for all CKD patients.'*
>
> ***(INT_04HCP)***

> *'…you know a lot of the [EPI] changes… are healthy changes and actually there's a lot of benefits for people's general health, their cardiovascular health which we know is something that's important with our society on a whole, but even more so if you've got chronic kidney disease, and even with the earlier stages there's some suggestion that there may be changes in childhood and some for kidney stage 2 level that also there's definitely going to be some health benefits for them.'*
>
> ***(INT_08HCP)***

Reduce decline in kidney function

In addition to the 'healthy-eating' label contentions, the redefining of Delphi statement 2, from round 1 to round 2, provoked a reaction to the term 'healthy kidneys'.

> S2/R1: Early phosphate intervention (EPI) should be labelled 'early intervention healthy- eating'
>
> In response to the expert panel's comments, this statement was amended to:
>
> S2/R2: The early phosphate intervention (EPI) program should be labelled 'early phosphate management for healthy kidneys'

The emphatic reaction from both parents and HCPs, vehemently opposed to the idea that the strategy could encourage 'healthy kidneys', again reflected a nuanced understanding of the term 'healthy' in the context of chronic kidney disease.

> *'If we are on the program, by default we don't have healthy kidneys."*
>
> ***(DEL_23PAR)***

In this context, this parent's interpretation of health was arguably similar to the Oxford dictionary's definition of health, 'the state of being free from illness or injury' (Lexico, 2019). The realisation that CKD can be delayed is a notion that is not regularly communicated to parents of children with early stage CKD. Thus, the belief that by delaying disease progression their child may be less exposed to the associated co-morbidities of progressive CKD, such as bone and cardiac complications, is poorly understood. Rather, as captured in this parent's comments, the EPI strategy seeks the retention of current kidney function (health), with some possible improvements, and the prevention of disease decline (Carrero and Cozzolino, 2014).

> *'It should say something like "early phosphate management to reduce kidney decline" or "early phosphate management to maintain kidney health"'*
>
> ***(DEL_23PAR)***

8.2.3 Objective 2 and 3: Exploration and consensus on 'how' and 'who' should deliver the EPI strategy

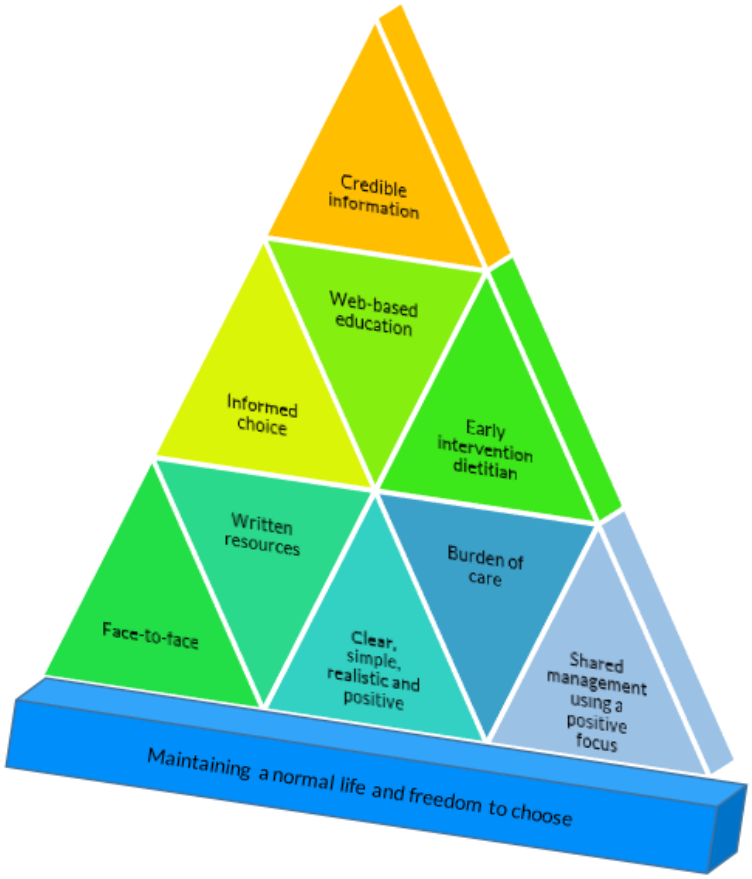
This section presents an integrative discussion of some of the pragmatic issues that require consideration when planning and delivering an EPI education strategy. Because the 'how' element of delivering the EPI strategy's education, skills and support is closely linked to 'who' should deliver it, this section collectively discusses the second and third research objective for this study.

RO2: To explore and gain consensus on *how* we should deliver the dietary phosphate education, skills and support to children, young people and their families.

RO3: To explore and gain consensus on *who* should deliver the EPI education, skills and support

These objectives were addressed during all three phases of the study. The Phase 1 findings exposed a wealth of insight from the experience and views of CYP, parents and other stakeholders involved in delivering and receiving early dietary interventions to manage chronic diseases, presented in Chapter 3. The key messages, detailed in Box 3, section 3.12 were carried forward to inform the interview guide instrument development, shaping the structure of the semi-structured interviews, and the resultant qualitative themes, which directly informed the Delphi survey statements. Firstly, in order to address the 'how we should deliver' focus of this objective, it is necessary to state 'what' the EPI will deliver. Drawing on the definitions in S1 and S3 (see section 8.2.2), the separate study phases and analysis procedures, the EPI strategy seeks to deliver the following three elements: education, skills and support. Each of these elements as aligned to RO2 and 3 will now be discussed. Beginning with the education component of the EPI, Figure 21, p. 263 captures the education themes for objective 2 and 3, which will be addressed in turn.

Figure 21. Objective 2 and 3 education themes



8.2.3.1 Education

The parent and young person's need for credible information

Dietary education is marked out in the findings of the Phase 1 systematic review as playing a pivotal role in building the foundations of knowledge in the early dietary management of chronic disease. In fact, parents are often so desperate for information to inform their actions in the early stages, their search for understanding can lead to misguided information (Ball et al. 2005). Accessing credible, evidence-based information from experts in the field is crucial (discussed in more detail later in this objective), and sign posting to reputable written or online advice is necessary and important. When education is not readily available, parents resort to 'self-management through Google' (INT_04HCP). For example, anxious to do the best for her daughter, this parent purchased an online renal course.

'… after she'd been diagnosed, I did a 5 night [online renal course] … I just Googled anythin' I could… they gave me intermittent information to read… I had to then like answer questions to see if I was like taking it in. And at the end of it I got like a recipe book online, they sent to me. It was dead good…'

(INT_01PAR)

The following of quasi-web-based guidance (Geense et al. 2017), with its possible inaccurate recommendations, is reported as a source of concern to many young people with CKD (Huby et al. 2016). Speaking of his uncertainty about accessing misguided online information, this young man explained:

'I would probably rather have it…from healthcare rather than a booklet [online], cos' anyone could right get it off the internet…Rather get it from a healthcare person's perspective, cos' they know more…'

(INT_06CYP)

The Delphi consensus statement 13 (S13/R2) firmly supported the desire of parents and CYP for early access to credible dietary information, as shown below. Action needs to be taken to produce, identify and make this desired evidenced-based dietary information available to CKD users and service providers.

Category: rationale	Median LOA rating (1-4)	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S13/R2	From diagnosis, parents, children and young people should have access to helpful and credible information about diet and CKD				
	4	95.5 (H92.3/P100)	4.5 (H7.7/P0)		

Informed choice

Early access to evidence-based information provides an essential foundation to build upon. It enables individuals to evaluate the benefits of an intervention, supporting them in making an informed choice.

'People [who] don't realize the impact food can have… they're not making it an informed choice'

(INT_03HCP)

Delphi consensus statements 11 (S11/R1) strongly endorsed this desire for parents and children to be educated on the impact of diet and early stage CKD to facilitate an informed choice, as shown below.

Category: rationale	Median LOA rating (1-4)	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S11/R1	Children and parents need to be told the impact of diet from an early stage, and then they can make an informed choice.				
	4	82.2 (H75/P91.6)	17.8 (H254/P8.3)		

Web-based education

In today's electronic world many CYP are using their mobile devices (Gutiérrez et al. 2016; Majeed-Ariss et al. 2015) for social networking and gaming, desiring education to be tailored to their virtual world, offering them instant 24/7 access whenever and wherever they choose.

> *'As somebody who watches YouTube 24/7', and on 'Instagram, Snapchat, WhatsApp… having a social media platform… would be extremely helpful'*
>
> ***(INT_03CYP)***

Attentive to this desire for web-based information and the risk of CYP and parents accessing quasi self-help sites and 'doing their own thing' (INT_04HCP), CYP, parents and HCP in this study proposed a purpose-built digital EPI education and support package, including a recipe book, via the EMEESY website (INT_01PAR) (refer to section 5.1.4.4 Responding to different learning needs). The upward trend in CYP mobile phone ownership suggests they have greater potential as an accessible, convenient and interactive platform to support chronic disease SM (Majeed-Ariss et al. 2015; CMR, 2014). In order to ensure this passive dietary education is correctly understood and translated into practical lifestyle changes, HCP suggested providing a 'feedback loop', enabling users to ask questions, and receive a timely response (INT_01HCP).

Development of web-based applications to address the information and support needs of CYP with CKD is emerging in the literature (Huby et al. 2016; Majeed-Ariss et

al. 2015). Young people often want to share their own, and hear the experience of others.

> *'If I was scrolling through and I see Instagram and it says how to stick to your diet with CKD or something, then like, I would look at that page, and then see what other people have to go through'*
>
> ***(INT_04CYP)***

The Delphi statements 24 (S24/R3) and 34 (S34/R2) firmly corroborated this desire for more information and support to be available online, and the pivotal role of shared experiences in the dietary management of CKD.

Category: delivery	Median LOA rating (1-4)	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S24/R3	The use of social media (e.g. EMEESY, Twitter or Facebook) in the EPI program would provide users with a useful additional tool and source of information or support.				
	4	90.5 (H92.3/P87.5)	9.5 (H7.6/P12.5)		
Category: delivery	**Median LOA rating (1-4)**	**Very high agreement (4)**	**High agreement (3)**	**Moderate agreement (2)**	**Low agreement (1)**
S34/R2	Patient stories and shared experiences are vital; they provide reassurance and make a difference to how a person feels they are able to manage their diet/lifestyle change.				

Early intervention dietitian

When considering 'who' is best placed to deliver the EPI strategy, the Totadri et al. (2005) research on the dietary perceptions and beliefs among families with children undergoing cancer therapy, and Stapleton *et al's* (2001) 'Go and grow with CF' research (included in the Phase 1 systematic review) demonstrate a positive commitment from multiple HCPs to their education strategies. Reflecting on this wider team involvement, INT_06HCP, a specialist dialysis nurse, suggested a children's 'early intervention CKD nurse' could deliver the EPI education. On the contrary, INT_02HCP, a renal specialist nurse, defended the already overextended role of the nurse. By questioning this notion of the renal nurse leading the EPI education, she emphasised parent's desire and expectation for a renal dietary expert.

'…I think if they're informed … I think nurses can deliver [the EPI education], but it's just one little aspect of that huge amount of stuff that we do. If I was a parent sitting in the clinic and I knew that there was a paediatric renal dietitian there delivering that information and guiding me along the way, then I would be very very happy and very confident that I was getting the right specialist professional advice.'

(INT_02HCP)

Likewise, both CYP, parents and HCPs almost unanimously agreed the children's dietitian, with training in the management of CKD, carried the greatest authority and expertise to deliver the EPI education strategy.

'It should be the dietitian…information should come first hand from the expert'

(INT_02CYP)

That person who can tailor the complex information to the individual child and family needs, is essential.

'I think because it's quite complex, and because it needs to be tailored to the individual you know, you can't just give the leaflet and expect people to just, like change their whole lifestyle. I think, and obviously it needs to have follow up, obviously, and for all the reasons it does need to be a dietitian, or someone trained in that dietary role.'

(INT_04HCP)

Delphi consensus statements S21/R1 and S22/R1, as shown below, resolutely identified the children's dietitian with training in the management of CKD as the best person to deliver the EPI program.

Category: delivery	Median LOA rating (1-4)	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S21/R1 (who should deliver the EPI?)	Dietary information/education always carries much more authority when it comes from people who have expertise in nutrition.				
	4	78.6 (H81.25/P75)	21.4 (H18.75/P25)		
S22/R1	A children's dietitian (ideally with training in the management of CKD) is the best person to deliver the EPI program				
	4	82.1 (H87.5/P75)	17.9 (H12.5/P25)		

In agreement with the literature (McLaughlin, 2004), these findings suggest the specialist renal dietitian, or the children's dietitian with training in the management of

CKD, as a member of the multi-professional team, could take on the role of an 'early intervention dietitian'.

Face-to-face input is the cornerstone

Returning to 'how' we should deliver the EPI education, parents on the Delphi expert panel and Phase 2 interviews stated:

> *'you can't beat face-to-face input' (**DEL_PAR19)** with 'dieticians with a strong renal leaning, [it] is imperative'*
>
> ***(DEL_PAR23)***
>
> *'having that face-to-face contact I think makes a huge difference in terms of helping people to make the lifestyle changes'*
>
> ***(INT_08HCP)***

Not only does 'it provide the dietitian with an opportunity to ensure that the messages are delivered and understood accurately', it also 'provides opportunity for the family to respond with questions and queries' (DEL_PAR19). Especially considering the dynamic nature of CKD where 'things can change all the time' (INT_03PAR), tailored specialist dietary advice can respond to the emergent and changing needs of the child and family (Beto et al. 2016). Delphi statement 23a and 27 corroborate this desire for a face-to face consultation with the children's renal dietitian, who can tailor advice to the changing needs of the individual and their family.

Category: delivery	Median LOA rating (1-4)	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S23a/R2 how should the EPI be delivered?	The EPI program should ideally be delivered initially during a face-to-face consultation with an expert in children's renal nutrition. This would affirm understanding of the advice, and provide an opportunity for the family to ask any questions.				
	4	86.3 (H77/P100)	13.6 (H23/P0)		
Category: delivery	**Median LOA rating (1-4)**	**Very high agreement (4)**	**High agreement (3)**	**Moderate agreement (2)**	**Low agreement (1)**
S27/R1	The EPI program needs to be flexible so that it can be tailored to meet the changing needs of the child or young person and their family.				
	4	82.1 (H87.5/P75)	14.3 (H12.5/P16.6)	3.6 (H0/P8.3)	

Written information

To strengthen face-to face contact, both parents and HCP affirmed the value of follow-up written information. It 'can be referred to time and time again' (DEL_PAR22), especially at home when 'you don't always have people to ask… they make a great reference point' (DEL_PAR23). However, when the children's renal dietitian is absent from the shared-care clinics, renal doctors may encourage parents to engage with passive dietary education, signposting them to EMEESY website dietary resources, or ask the dietitian to post out some written information. For parents who choose not to follow recommendations to seek out online advice, a feedback loop as mentioned earlier, would provide accountability and support the translation of advice into practical everyday lifestyle changes. Additionally, as families come to terms with a CKD diagnosis, taking on a lot of information involves a 'filtering' process as they hold 'on to what's really important at that point', and allow the less important information to be 'pushed back' (INT_03PAR). In order to improve the recognised poor long-term retention of knowledge (Stapleton et al. 2001), and bring to the fore what is essential, face-to-face dietary education, resources and activities needs to be provided regularly and long-term, and tailored to the age/developmental needs of the child/family (Murimi et al. 2018). Interviewees in Phase 2 affirmed education to involve the 'drip feeding' of information over time (INT_01HCP; INT_03PAR), with some 'sort of practical kind of input' (INT_03PAR) to consolidate the learning (discussed further in 8.2.3.2 – Skills).

Statements 23 (23b/R2) and 47 (S47/R2) corroborated these qualitative findings, requesting written resources to support verbal advice, and for the shared-care clinics to be attended by the children's renal dietitian.

Category: delivery	Median LOA rating (1-4)	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S23b/R2	After a face-to-face conversation with an expert in children's renal nutrition, written dietary phosphate information (including recipe information) is vital, and provides a useful reference that can be frequently accessed at home.				
	4	81.8 (H77/P88.8)	12.1 (H23/P11.1)		
S47/R2	In order to deliver the EPI diet, children's dietitians needs to have more of a presence in the shared-care clinics				
	4	77.3 (H84.6/P66.6)	13.6 (H7.7/P22.2)	4.5 (H0/11.1)	4.5 (H7.7/P0)

Clear, simple, realistic and positive

To positively influence engagement and promote adherence to preferred CKD lifestyle choices, the EPI education needs to be 'clear, simple, realistic and positive' (S25/R1) (see below). Offering a practically relevant message that can be replicated in the home/life environment (Coleman et al. 2000) was recognised as important.

Category: delivery	Median LOA rating (1-4)	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S25/R1	The EPI program needs to be delivered using a clear, simple, realistic and positive message				
	4	71.4 (H81.25/P58.3)	28.6 (H18.75/P41.6)		

Burden of care

Delivering an early dietary change message was proposed as a way to enable families to better understand the early CKD diet/lifestyle choices, and thus potentially reduce the 'burden of care' in the later stages of CKD. Despite a statistically significant interrater reliability (see section 7.5.8) across Delphi rounds 1 and 2 for statement 12, moderate Kappa = 0.6 (p <0.0001), 95% CI (0.512-0.900), agreement on early dietary intervention as a means of alleviating the later 'burden of care' was met with contention, and failed to reached consensus.

Categor: rationale	Median LOA rating (1-4)	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)	Fisher's Exact Test
S12/R3	Introducing dietary changes early may not reduce the 'burden of care' in the later stages of CKD, but may help to establish a better understanding of diet/lifestyle to inform long-term change.					
	4	66.7 (H61.5/P75)	19 (H23/P12.5)	9.5 (H7.6/P12.5)	4.8 (H7.6/P0)	P=1.0

Parents and HCP felt the 'burden of care' associated with 'having kidney disease' (DEL_PAR22) was different to the 'dietary burden of care', although the two become interrelated during the later stages of CKD. As described by the mother of a young person with advanced CKD, diet 'should not be seen as a burden but a necessity' (DEL_PAR23). Early dietary intervention, rather than alleviating the later 'burden of care' proposed a vital opportunity for parental involvement and control. It also provided an early opening for parents to build trusted relationships with HCPs.

Shared management using a positive focus

At this early stage of CKD when anxiety runs high, it feels like 'parents can do very little' (INT_07HCP), despite 'want[ing] to do all they can to help their loved one' (DEL_PAR19). It is clear that the EPI 'dietary change [would give them] a positive focus', knowing that 'their actions [are] helping their loved ones at a difficult time' (DEL_PAR22). It is apparent that a triadic mismatch can exist between the clinicians drive to best manage the child's condition, the parent's desire to embrace the EPI advice completely or separate 'health…and the rest of their lives' (INT_08HCP), and the child's desire to get on with 'normal life'. The EPI education strategy needs to work for all parties in this triad – providing a bridge to best manage a child's health, whilst offering a tool to allow the parent and child to take control and find a new level of normality in their everyday life. HCP recognised the 'empowering' value of the EPI strategy. Speaking of CYP and parents' desire to be proactive, one paediatric nephrologist explained, 'A lot do say, can we do anything to make a difference?' and 'having that [the EPI strategy] is really helpful' (INT_08HCP).

However, both parents and HCP were concerned about the frequency and volume of information exchange, believing 'it's a fine line, it's a balance' (INT_01HCP), neither withholding information nor giving too much that they feel 'completely overwhelmed'

(INT_03PAR). The focus should be on 'help[ing] them to self-manage well and for it to be empowering rather than something that becomes sort of a burden or a focus' (INT_08HCP). Delphi consensus statement 7a (S7a/R2) and 43 (S43/R2) captured these constructs of providing a positive focus and empowering parental involvement. These results were corroborated by Delphi statements 7b/R2 and S43/R2, which confirmed that diet/lifestyle change can provide a positive focus and significant contribution to disease management in the early stages when anxiety is high.

Category: rationale	Median LOA rating (1-4)	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S7b/R2	During the early stages of a chronic disease, diet/lifestyle change can provide a positive focus for the family, knowing that they are making an important contribution to disease management.				
	4	77.3 (H61.5/P100)	18.2 (H30.7/P0)	4.5 (H7.7/P0)	
Category: rationale	**Median LOA rating (1-4)**	**Very high agreement (4)**	**High agreement (3)**	**Moderate agreement (2)**	**Low agreement (1)**
S43/R2 (other considerations)	Having kidney disease creates anxiety. With the right level of dietary education and support, the EPI program would give the family a positive focus and a way to help their loved one				
	4	90.9 (H84.6/P100)	9.1 (H15.5/P0)		

Maintaining a normal life and freedom to choose

This desire for involvement and control by parents and CYP was also apparent in decisions that affected their ability to have a 'normal life'. The mother of a child on dialysis felt this decision to get involved and take control, reflected an individual's vantage point. Some parents would think, 'it will be OK, we won't get to that point' and therefore would take a laissez-faire approach, 'I'm not going to take much notice' and leave things to take their own course (INT_03PAR). By contrast, others would 'go completely the other end of the scale and get a bit obsessive' (INT_03PAR). One parent, convinced by the EPI diet, pledged to never serve 'any remotely processed meat' at home, as 'I'm against' it (INT_05PAR). This conviction was a way of making

the EPI diet a 'normal' part of family life. Her son, however, in a bid to create normality when 'having his friend stay the night in the holidays', negotiated an occasional treat, 'mum said I might be able to have some [pepperoni] pizza' (INT_05CYP).

Young people spoke of their interest in following the EPI diet 'so long as it doesn't really affect anything else', or mark them out as being 'different' from their peers (INT_03CYP). For example, for one young person who pointed out that most of her friends like to 'meet up at McDonald's', following the EPI diet 'it's a bit weird knowing that I might be different from everyone else' (INT_03CYP). By contrast, another young person felt confident that he could suggest an alternative place to meet up '…yeah like Subway is healthy, cos' it's bread and there's loads of healthy vegetables and stuff, so that would be a good place' (INT_04CYP). Additionally, once offered the EPI diet, CYP felt strongly the advice should not be prescriptive; rather 'a choice would be best'. Otherwise 'it would be waste of NHS time' (INT_04CYP). After all, remarked one young person, 'diet is part of their freedom…you can't tell a person exactly what they need to eat' (INT_03CYP).

Statement 6 (S6/R1) validated the emphasis on providing a tool that would build confidence to manage their own diet during the early stages of CKD.

Category: rationale	Median LOA rating (1-4)	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S6/R1	The provision of early dietary education gives children, young people and families a tool to more confidently manage their own diet during the early stages of a chronic disease				
	4	75 (H75/P75)	21.4 (H25/P16.6)	3.6 (H0/H8.3)	

Table 50, p. 274 provides a summary of potential EPI education strategies for parents and CYP during the early stages of CKD

Table 50. Summary of EPI education strategies for early CKD

Target area	Education strategies
Multi-professional team	Early intervention dietitians take on the role EPI education
Web-based education program	Online EPI dietary education program tailored to the needs of parents, and children and young people with CKD
Dietary resources	EPI dietary resources available as a hard copy and online
YouTube	CYP share their stories and experience of diet and CKD
Early parental and CYP involvement	Parents and CYP are empowered to engage in the early dietary management of CKD
Clear, simple, realistic and positive message	The EPI diet is communicated using a clear, simple, realistic and positive message
Partnership model of SM	The HCPs share the early CKD management with the parent, child and young person

8.2.3.2 Skills

The skills components of the EPI strategy will now be discussed.

Figure 22 captures the skills themes for objective 2 and 3, each theme will be addressed in turn.

Figure 22. Objective 2 and 3 skills themes



Regular cookery workshops

In order to adopt the EPI's 'renal-healthy-eating' diet with its reduced consumption of processed foods and more home-prepared meals, families will need some basic cooking skills. For those who regularly cook from fresh ingredients this will be second nature, but for those who 'heavily rely on convenience foods' (INT_03HCP), and have 'busy lives', often the 'last thing [they] want to do is actually stand in the kitchen and cook [a] meal from scratch' (INT_03PAR). The 'limited' or 'lack of cooking skills amongst some families' (INT_22PAR) was noted as a factor restricting them from consuming home-cooked meals (INT_10HCP). To address these practical prerequisites, some basic cooking skills training needs to be offered, and flexibly adapted to the needs of each family.

Because of their primary involvement in meal preparation, women in the Coleman et al. (2010) tailored diabetes prevention programme (from Phase 1) were invited to participate in two cookery workshops, demonstrating how affordable, culturally relevant healthy meals can be prepared in 10-20 minutes. Recipes were made available to the parents on request. The workshops provided education on the benefits of healthy food, budgeting and shopping; and parents valued the social support as they shared experiences and ideas.

Qualitative data from parents participating in Phase 2 and 3, established their desire to attend a 'local hub' or 'workshop' (DEL_22PAR) with other parents and families in a 'similar situation to [them]self' (INT_01PAR), to learn about food labels, plan meals for different ages, engage in cooking demonstrations and 'share ideas' and 'renal recipes' (DEL_22PAR). One paediatric renal dietitian in Phase 2 described how 'amazing' it would be to do 'cooking sessions with a group' and see parents 'bounce [ideas] off each other' (INT_03HCP). Parents felt it would be important to include the children, as 'doing it together' would be 'fun' and make them 'feel important' (INT_04PAR). One mother, INT_03PAR, recognising that some children remain in the early stages of CKD for many years, would give them plenty of time to learn new skills. She described the importance of early skills-based workshops for families using a horticultural metaphor:

'… early [skills] preparation…does work', it is about 'setting that seed early in the mind of the child or…family', in a 'fun' and 'easy' way

(INT_03PAR)

'… some of these [early CKD] children have gone for years aren't they and so you've got lots of time to learn new ways of cooking and just [be] more prepared for it [later stages of CKD], that's what it's about being more prepared.'

(INT_03PAR)

Rather than making 'sudden big changes' explained a renal doctor, cookery workshops are about 'making small changes' in the early stages that become habits 'that turn into a lifestyle' (INT_08HCP). As one parent commented, an 'annual renal family cooking workshop' would be 'fun and informative', with 'education…drip fed in as cooking demonstrations take place' (DEL_22PAR). However, 'why only hold workshops on an annual basis?' a renal doctor questioned (DEL_16HCP), rather, 'regular workshops…would be wonderful', proposed a parent (DEL_22PAR). Alternatively, a renal dietitian voted for a bespoke 'home visit' or 'supermarket shop' where information and skills can to be tailored to the individual's circumstances – allowing 'more practical' and realistic advice to be offered (INT_03HCP).

EPI champions

Harnessing the value of peer education, one paediatric nephrologist proposed a 'model of peer support', where 'motivated' individuals, who 'do very well' with the EPI diet are profiled in the workshops as EPI champions to 'help and support others' (INT_08HCP). As the EPI champions share their ideas and experiences, the group learn from each other how to 'cook healthy' meals, passing on valuable 'life skills'. Additionally, this approach retains the HCP's conviction that it is 'easier to hear and take on board' dietary education and skills from peers 'rather than a health professional' (INT_08HCP).

Delphi statement 46 (S46/R2) and 49 (S49/R2) overleaf, corroborated these qualitative findings.

Category: other considerations	Median LOA rating (1-4)	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S46/R2	The EPI program will encourage more home-cooked meals. Some families may benefit from cookery workshops to inspire and support them to follow the program.				
	4	77.3 (H77/P77.7)	13.6 (H15.3/P11.1)	9.1 (H7.7/P11.1)	
Category: Other considerations	**Median LOA rating (1-4)**	**Very high agreement (4)**	**High agreement (3)**	**Moderate agreement (2)**	**Low agreement (1)**
S49/R2	The provision of EPI workshops for families (including carers and grandparents) could provide information to manage CKD in different age groups, advance their cooking skills and provide social support.				
	4	77.3 (H77/P77.7)	22.7 (H23/P22.2)		

Table 51 provides a summary of potential EPI skills strategies for parents and CYP during the early stages of CKD.

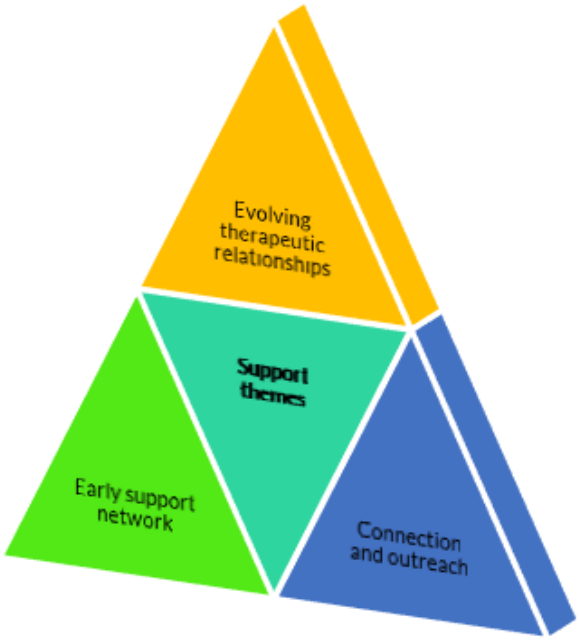
Table 51. Summary of EPI skills strategies for early CKD

Target area	Education strategies
'Local hub' or 'cookery workshop'	EPI 'renal-healthy-eating' workshops demonstrating basic cooking skills, and providing education on food labels, meal planning for different ages, and renal recipes ideas
Regular skills workshops	Provide skills/cookery workshops regularly (several times a year)
Home visit or supermarket shop	Provide bespoke education and skills advice to an individual/family following a home visit or supermarket shop
EPI champions	Learn from those who have developed the skills and gained experience EPI champions support the facilitation of the cookery workshops

8.2.3.3 Support

The support component of the EPI strategy will now be discussed. Figure 23, p. 278 captures the support themes for objectives 2 and 3, each theme will be addressed in turn.

Figure 23. Objective 2 and 3 support themes



Evolving therapeutic relationships

In order to impact lifestyle change, dietetic-led EPI education and skills-based cooking workshops need to be delivered within the framework of a growing therapeutic relationship with the dietitian and other HCPs. The diagnosis of 'chronic kidney disease…changes the family prescription forever!' remarked a consultant nephrologist, and tailoring advice to the unique needs of the child and family, requires 'building relationship' (INT_07HCP). As one mother stated, 'How can you support a family if you don't really know the kind of dynamics, or what's going on for them at that point?' (INT_03PAR). Lifestyle change demand motivation and support, and 'that's hard to give if you don't have... [a] relationship that you've built...with that element of trust and respect' (INT_08HCP). As one young person commented, he didn't want to be a dumping ground for 'a load of information', rather an initial introduction would provide a 'more personal' opportunity to meet the dietitian who was going to play a fundamental part in his renal management.

Delphi consensus statement 28c (S28c/R2) substantiated these findings (see overleaf).

Category: other considerations	Median LOA rating (1-4)	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S28c/R2	Early introduction to the dietitian means trusted relationships will develop over time, making the nutritional message more effective.				
	4	81.8 (H76.9/P88.8)	9.1 (H15.3/P0)	9.1 (H7.7/P11.1)	

Therapeutic relationships are established over time. With the current scarce, if any, specialist paediatric renal dietetic support for CYP attending shared-care clinics (see section 1.3.2), families are required to depend on telephone advice. Most often, however, the consultant engages patients in passive methods such as signposting them to dietary resources available on the EMEESY website, dietary information posted out by the dietitian, or information handed out by the paediatric nephrologist.

This posed a problem for INT_03HCP, the renal dietitian, because people 'need to know that they are valued', and 'you can't do that by giving them a piece of paper'. Furthermore, as one renal consultant explained, it's harder to build that therapeutic relationship 'if you're calling out of the blue' (INT_08HCP). This is where either the specialist renal dietitian needs to have more of presence in the local shared-care clinics, or local shared-care dietetic input, working 'with the same [EPI] ethos' would be a vital link (INT_08HCP). Delphi statement 47 (S47/R2) substantiated the need for more dietetic input in the shared-care clinics.

Category: other considerations	Median LOA rating (1-4)	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S47/R2	In order to deliver the EPI diet, children's dietitians needs to have more of a presence in the shared-care clinics.				
	4	77.3 (H84.6/P66.6)	13.6 (H7.7/P22.2)	4.5 (H0/P11.1)	4.5 (H7.7/P0)

Early support network

Once the diagnosis of early CKD had sunk in, and the initial flurry of appointments and hospital contacts became less frequent, many parents recognised their need for

an ‘early support network’. As little or no medication is required for many early stage patients, medical input is usual scheduled at 6-12 months intervals. Between these hospital contact points, the routine of life threw up questions about CKD, diet and lifestyle, and parents desperate for reassurance that they were ‘doing [it] correctly’ wanted to reach out to others in the same situation as them. ‘I mean, [CKD] kids don’t come with a manual’ remarked one mother (INT_04PAR). The season that followed diagnosis left 05PAR feeling vulnerable and uncertain how she was going to manage, ‘does anyone really care? and ‘will anyone be in touch?’. Despite attending clinic and ‘seeing’ lots of different people, there was no opportunity to really engage at a meaningful level. She wanted ‘an actual support group…not [a] social media one’, where families come together and shared their experiences (INT_05PAR).

By contrast, some parents felt a social media support group would be a ‘good idea’ if managed appropriately (DEL_27PAR). For others, however, access to a virtual or real support group was either not desired, or engagement from the side-line was enough. One mother commented ‘I enjoy following it myself, but feel that people are more private about family health than to want to share it on social media’ (DEL_26PAR).

Delphi consensus statement 24 (S24/R3), 32 (S32/R2), and 34 (S34/R2) below corroborated these desires for psychosocial support through therapeutic relationships with HCPs, and access to online to face-to-face support from other parents.

Category: delivery	Median level of agreement rating (1-4)	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S24/R3	The use of social media (e.g. EMEESY, Twitter or Facebook) in the EPI program would provide users with a useful additional tool and source of information or support.				
	4	90.5 (H92.3/P87.5)	9.5 (H7.6/P12.5)		
Category: other considerations	**Median level of agreement rating (1-4)**	**Very high agreement (4)**	**High agreement (3)**	**Moderate agreement (2)**	**Low agreement (1)**
S32/R2	Parents of children with early stage CKD should have access to support from other parents who are in or have been in their situation.				
	4	81.8 (84.6/P77.7)	18.2 (H15.3/P22.2)		
Category: delivery	**Median level of agreement rating (1-4)**	**Very high agreement (4)**	**High agreement (3)**	**Moderate agreement (2)**	**Low agreement (1)**
S34/R2	Patient stories and shared experiences are vital; they provide reassurance and make a difference to how a person feels they are able to manage their diet/lifestyle change.				
	4	77.3 (H77/P7)	9.1 (H15.3/P0)	13.6 (H7.7/P2)	

Connection and outreach

During the early post diagnosis stage, a young person's intimation that all is well may be covering up feelings of 'anxiety' and 'stress'. Friends and family become pillars of strength, yet there are times when the young person desires support from a CKD specialist without having to wait for an appointment or engage in a lengthy conversation. A teenager suggested a CKD 'text hotline' as an anonymous platform for posting a query and connecting with a specialist (INT_03CYP). Additionally, an 'outreach program' where your diagnosis does not define you, could provide a social networking opportunity. She explained, where CYP with CKD 'can just gather together, relax, and do whatever, just hang out together' (INT_03CYP). If you wanted it, you could access non-judgemental professional support. It would be a safe, informal space, with opportunity to engage in activities, or one-to-one with people

your age, and HCP. There would be no rush or agenda, people had time for you and were genuinely interested in getting to know and support you (INT_03CYP).
Table 52 provides a summary of potential EPI support strategies for parents and CYP during the early stages of CKD.

Table 52. Summary of EPI support strategies for early CKD

Target area	Education strategies
Early dietetic introduction	Meet the dietitian during the diagnosis period, and start to build a therapeutic relationship.
Shared-care renal dietetic input	Children's (renal) dietitians to attend the EMEESY shared-care clinics
Building therapeutic relationship	Addresses a vital psychosocial need of parents
Early support network	Parents desire to meet with others either virtually or in person, to share experiences and ideas about CKD management
Text hotline	Young people can anonymously post a query and connect with a CKD specialist
Outreach program	CYP can access non-judgemental psychosocial and lifestyle support

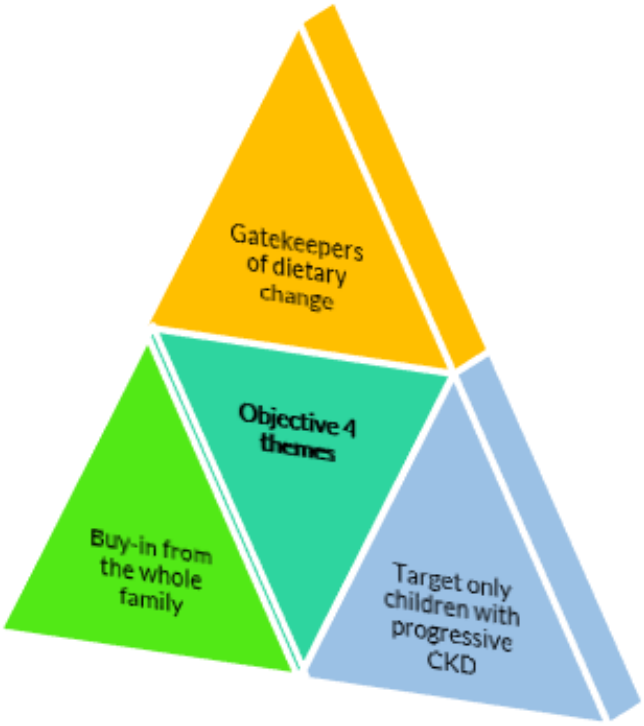
8.2.4 Objective 4: Exploration and consensus on the recipients of the EPI

This section addresses the fourth research objective for this study.

RO4: To explore and gain consensus on 'who' should receive the EPI strategy.

Figure 24, p. 283 captures the themes for objective 4, each theme will be discussed in turn.

Figure 24. Objective 4 themes



Gatekeepers of dietary change

This objective was addressed during all three phases of the study. The education and dietary intervention strategies detailed in the Phase 1 systematic review studies were tailored to appeal to all family members. However, studies detailing parent/carer participant characteristics predominantly recruited women. This likely reflected the culturally dominant involvement of women in childcare and meal preparation in the Californian, Hispanic, (Coleman et al. 2010) and Indian (Totadri et al. 2017) study population. In these cultures, these findings suggest that mothers are often the gatekeepers of family dietary change.

In this study there was a similar tendency towards mothers, with 86% of all recruited parents being women, despite invitations extended to both parents. As the child's prime caretakers, this likely reflects their predominant attendance during hospital/clinic visits (Walters et al. 2001) and their availability for interview. However despite this, in the Delphi survey, both parents and HCPs advocated for a more inclusive agenda concerning the drivers of dietary change in a family. This difference

in opinion may reflect the wider family contribution at home, and not just the availability for an interview.

Delphi consensus statement 18 (S18/R2) corroborated this finding.

Category: other considerations	Median LOA rating (1-4)	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S18/R2	The EPI program should target the main leaders of diet and lifestyle change in a family				
	4	86.4 (H84.6/P88.8)	4.5 (H7.7/P0)	9.1 (H7.7/P11.1)	

Buy-in from the whole family

Once the 'leader' or food 'gatekeeper' has been identified, and EPI education, skills and support has been provided, they can provide an in-road to encourage buy-in from the entire family. There is potential that 'it becomes a habit for the whole family' and that means 'everybody being healthy together' (INT_08HCP). No one is 'singled' out as having different needs because of CKD, instead, 'it encourages a healthy mind-set rather than a disease mind set' (INT_08HCP). 'Little changes' in the family diet, form 'habits' that 'become the norm', which are 'much more likely to be imbedded in the long term' (INT_08HCP). Any perceived early dietary burden of care is disarmed as the EPI diet becomes the 'norm' for the whole family.

In the Phase 1 systematic review studies, education was esteemed not only as leverage for enhancing knowledge to drive change, but it held persuasive powers with family and peers. It raised awareness of chronic disease, increasing recognition of the need for change, motivating action towards better health. Education empowered parents/carers, as the leaders of family change, to take control of their children and family eating habits (Coleman et al. 2010).

Delphi statement 17 (S17/R1) (see overleaf) corroborated the whole family focus of the EPI strategy.

Category: other considerations	Median LOA rating (1-4)	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S17/R1	The EPI program should be directed at the whole family.				
	4	78.6 (H75/83.3)	21.4 (H25/P16.6)		

Target only children with progressive CKD

As a main objective of the EPI strategy is to delay disease progression, it is appropriate that the diet should be offered only to CYP with active disease decline, and their families. However, parents opposed this approach, claiming that the EPI 'diet is a key component of keeping well', and therefore appropriate 'not only for slowing progression of the disease, but also important to help prevent complicating illnesses, such as coughs [and] colds' (DEL_19PAR). Moreover, as one mother asserted, as '…it's not going to do any harm, so it's only going to help anyway… you know, crack on with it. It seems silly not too, when it could help' (INT_05PAR).

Conversely, HCPs were almost united in the belief that 'an element of screening' is required 'to identify people at risk of the condition progressing' (INT_08HCP). However, there remained a difficulty with those 'children [where you] don't know if they are going to progress' (INT_08HCP). In these children, the nephrologists concurred with parents in their belief that '…early advice can do no harm, and may do good' (INT_07HCP). The 'healthy-eating' principle of the EPI diet would establish some good lifestyle habits, and from the 'phosphate perspective any little changes would mean the habits are there …so if [the] condition is progressing then … it's actually just making little steps … rather than having to make a sudden big change to [their] lifestyle' (INT_08HCP).

This contentious issue reached consensus in the Delphi survey, with the majority of parents and HCP agreeing in S16/R1 that only children with progressive CKD should be offered EPI (see overleaf).

Category: other considerations	Median LOA rating (1-4)	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S16/R1	Early phosphate intervention should be offered to all children and young people with early stage CKD (2-3) who are likely to progress to the later stages of CKD (4-5)				
	4	78.6 (H75/P83.3)	17.9 (H25/P8.3)		3.6 (H0/P8.3)

Table 53 provides a summary of the main recipients of the EPI strategies during the early stages of CKD

Table 53. Summary of the recipients of the EPI strategy

Recipients of the EPI strategy	
Leaders of family dietary change	Mothers, fathers, grandparents, guardians and siblings can all be leaders of family dietary change
Whole family engagement	EPI diet will seek buy-in from the whole family
Children with progressive CKD	Target only children with progressive CKD

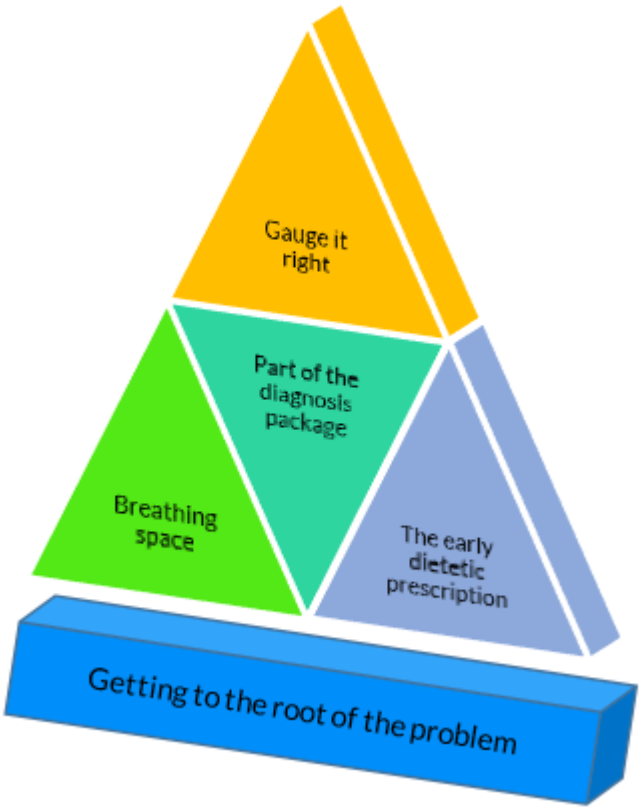
8.2.5 Objective 5: Exploration and consensus on 'when' to deliver the EPI

This section addresses the fifth research objective for this study.

RO5: To explore and gain consensus on 'when' the EPI education and support should be delivered

Figure 25, p. 287 captures the themes for objective 5, each theme will be discussed in turn.

Figure 25. Objective 5 themes



This objective, which focused on the best time to deliver EPI education and support, was met during all three study phases.

Gauge it right

All participants agreed that many important variables need to be considered when trying to identify the best time to employ the strategy. While recognising that the EPI 'information is vital' (INT_02PAR) most parents, CYP and HCPs agreed there is no perfect delivery time. 'You've got to gauge it right' (INT_01HCP). Parents have a lot of information to assimilate at diagnosis, and diagnoses can vary so much, and 'different families all require different amounts of [information]' (INT_01HCP). Although it is current practice for dietary advice to be given in the later stages of CKD, some professionals indicated this practice needs to be revised.

'The perturbations of bone mineral metabolism … occur down in CKD level 3, particularly in level 2. Most dietary advice is given for level 5 - obviously for those on dialysis, and those approaching dialysis. But in fact, we [renal doctors] feel that the advice should be given much earlier on … each family needs to be assessed on its merits …The families deserve it.'

(INT_07HCP)

Delphi consensus statement 10 (S10/R1) endorsed the long-term focus of dietary phosphate education, beginning in the early stages of CKD.

Category: rationale	Median LOA rating (1-4)	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S10/R1	Dietary phosphate education is a long term process and should begin in the early stages of CKD				
	4	89.3 (H81.25/P100)	10.7 (H18.74/P0)		

Breathing space

Leaving some 'breathing room for the family' post-diagnosis is 'important', remarked one young person (INT_02CYP). Time is needed 'to come to terms with the disease' (INT_02CYP) before more information can be received. If you are 'given everything at once … it's not all going to go in … so turning that information over time, giving it after… a week, few weeks, maybe a month. It's important to get the information' (INT_02CYP).

Despite the early 'bombardment' of information, and the 'overwhelming' nature of a CKD diagnosis, some parents and CYP felt strongly that they deserve to be given the EPI information close to diagnosis. Receiving difficult information is not easy. Similarly, providing the EPI information 'may be too much to put on somebody … [but] diagnosing someone sometimes is too much to put on somebody' (INT_02PAR). However, 'it would be better to let people know earlier, like the sooner the better' (INT_04CYP).

Part of the diagnosis package

Many parents, CYP and professionals agreed that the EPI education and support should be given as 'part of the diagnosis' package. Food is part of everyday life, and if

EPI proposes benefits in the early management of CKD, parents and CYP felt strongly that despite the large volume of information already received, this information should not be left until stage 4 'that's too late', rather it should be part of the diagnosis package (INT_03CYP). One mother described how she might feel if denied this information.

> *'Now if somebody would have said to me, we'll leave [the EPI information] because she's got too much to take on. So months passed. In that month… like the damage it could be beyond…it doesn't bear thinking about. I genuinely think that we need to be told from day one.'*
>
> ***(INT_02PAR)***

Many parents believed that making the EPI strategy a part of the 'diagnosis package' would increase motivation, people would be 'more inclined to do it' (INT_05PAR). Additionally, having the information and support 'as early as possible' would help parents 'bring it into [their] normal life without it feeling like a chore' (INT_03PAR).

Moreover, as discussed in objective four, many professionals emphasised, if a child is diagnosed with progressive early stage CKD, they should be given the EPI information and support 'right from the outset, the first time that you encounter them' (INT_07HCP). Although there might not be the opportunity to go through the EPI information in detail at the first consultation, people should be told there is a dietary aspect to the management, and 'within the first 3 months … actually go through things with them' (INT_03HCP).

Delphi consensus statement 19 (S19/R1) corroborated parents and professionals desire for the EPI strategy to be introduced at or close to diagnosis, and 7a (S7a/R2) recognised EPIs function in motivating patients and families to modify their diet and lifestyle early (see overleaf).

Category: rationale	Median LOA rating (1-4)	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S19/R1	The EPI program should be offered at, or close to the diagnosis of CKD.				
	4	75 (H62.5/P91.6)	17.9 (H25/P8.3)	7.1 (H12.5/P0)	
Category: rationale	**Median LOA rating (1-4)**	**Very high agreement (4)**	**High agreement (3)**	**Moderate agreement (2)**	**Low agreement (1)**
S7a/R2	When a child is diagnosed with a chronic disease, knowing and understanding the impact of diet on the disease may motivate families to make diet/lifestyle changes				
	4	72.7 (H61.5/P88.8)	22.7 (H30.74/P11.1)	4.5 (H7.7/P0)	

The early dietetic prescription

Another reason professionals, parents and CYP argued for the early introduction of the EPI strategy was reflected in their views on its essential position alongside medicine in the management of CKD. This often reflected the impact that diet would have on everyday life.

> *'I honestly think dietary is a bigger part as any type of [medical] treatment you are gonna' give. It's a massive part.*
>
> ***(INT_02PAR)***

Many professionals agreed on the parallel significance of the dietetic, medical and psychosocial prescription, with one renal doctor believing they need to be joined up (INT_07HCP).

> *'The management of children with chronic kidney disease requires three basic prescriptions…the medical prescription…the dietary prescription and the…psychosocial prescription, and all of them need to be interlinked.'*
>
> ***(INT_07HCP)***

Delphi consensus statement 15 (S15/R1) and 28a (S28a/R2) corroborated these findings (see overleaf).

Category: rationale	Median LOA rating (1-4)	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S15/R1	Early dietary advice should be viewed as important as the medical treatment				
	4	78.6 (H81.25/P75)	17.9 (H18.75/P16.6)	3.6 (H0/P8.3)	
Category: rationale	**Median LOA rating (1-4)**	**Very high agreement (4)**	**High agreement (3)**	**Moderate agreement (2)**	**Low agreement (1)**
S28a/R2	As part of the early management of CKD, the family should be introduced to the doctor and dietitian.				
	4	95.5 (H92.3/P100)		4.5 (H7.7/P0)	

Getting to the root of the problem early

Additionally, professionals' believed by making the 'early renal-healthy-eating' diet a key part of the early CKD management package, this would be 'tackling the root of the [phosphate] problem' (INT_04HCP). As one HCP explained,

> *'It's far better to try to ameliorate the diet at an earlier stage … You build up a relationship, and that's why I would strongly believe that the dietary involvement in the predialysis stage, at an early stage, is really worthwhile'.*
>
> ***(INT_07HCP)***

Table 54, p. 292 provides a summary of when the EPI education, skills and support should be delivered.

Table 54. Summary of when the EPI should be delivered

Key considerations	When to deliver the EPI strategies
Gauging it right	The timing can be different for each family. Parents have a lot of information to embrace - diagnoses can vary so much, and different families all require different amounts and type of information
Part of the diagnosis package	EPI strategy should be employed during the diagnosis period
Breathing space	Families may need time to comprehend and come to terms with CKD, before receiving the EPI dietary advice
Dietetic prescription	Medical and dietary management need to be addressed during the early stages of CKD
Getting to the root of the problem	Introducing EPI early seeks to address the problem of excess dietary phosphate early on

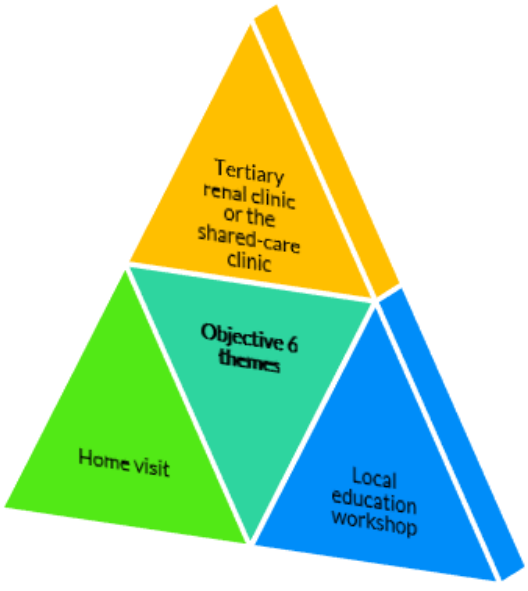
8.2.6 Objective 6: Exploration and consensus on 'where' to deliver the EPI

This section addresses the sixth research objective for this study.

RO6: To explore and gain consensus on where the EPI education and support should be delivered

Figure 26 captures the themes for objective 6, each theme will be discussed in turn.

Figure 26. Objective 6 themes



This objective, which sought to identify the best location for delivering the EPI education, skills and support, was addressed during all three phases of the study.

Tertiary renal clinic or the shared-care clinic

The studies included in the systematic review involved heterogeneity of settings, one in an elementary school, three in a specialist clinic for chronic disease, two in a children's in-patient ward, and one included an educational home visit. Location was a key facilitator in the CF trial (Stapleton et al. 2001), where the educational intervention involved a home-based course, sandwiched between an introductory, and final group workshop with other families at the hospital. This offered the opportunity to adapt the program to the unique needs of each family, while promoting social and peer interaction during the hospital workshops.

Parents and HCPs in this doctoral study agreed the location for delivering the EPI should be carefully considered, to minimise absenteeism from school (CYP) and work (parents), and the cost of travel and parking (DEL_14HCP).

Parents argued for the delivery of EPI in a location that gave the family optimal access to 'timely information', with opportunity to ask questions of the relevant professionals all 'in one place' (DEL_19PAR). In view of the limited availability of the multi-professional team in the EMEESY shared-care clinics, the tertiary renal clinics is currently the only location to provide this 'one-stop' service. However, for many families, this would mean long journeys of up to 3-4 hours, with the associated travel costs and absence from school.

In the words of a renal nurse specialist

> *'… the gold standard would be having a renal dietitian there [shared care clinics]. The next best is having the local dietitian'*
>
> ***(INT_02HCP)***

Corroborating these findings, limited access and an inability to attend were recognised challenges in the Coleman et al. (2010) study in the Phase 1 systematic review.

Planning the EPI delivery to coincide with a child's routine appointment in the shared-care clinic, close to the family home would minimise travel and absence from school.

Indeed, for all of the parents in the Delphi survey this would be a preferred option, with 100% agreement (see below). These findings suggest parents desire a 'one-stop' service at a shared-care clinic close to their home.

Delphi consensus statement 20a (S20a/R2) corroborated these findings for the EPI to be delivered in a location that best suites the family.

Category: rationale	Median LOA rating (1-4)	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S20a/R2	The EPI program should be delivered in a location that best suites the family, this may be the home, a shared-care renal clinic close to home, hospital ward or another suitable venue.				
	4	90.6 (H84.6/P100)	4.5 (H7.5/P0)	4.5 (H7.5/P0)	

Home visit

Many parents and professionals suggested the 'home environment' as an ideal 'place to deliver the intervention' (DEL_02HCP). It would allow the teaching to be tailored to the specific circumstances and needs of the child and family, encouraging buy-in from the whole family. One mother recalled the long-standing impact of a home visit from the play therapist, the personal touch and hands-on engagement made a real difference.

> *'What really helped us … was when [the play therapist] came to our house, and she spoke to [our daughter] and her sisters about what kidney failure was. That were brilliant. And then it was like 'Oh, yeah ok then'. How they took it was…so doing like a hands on kind of thing with them. Like we made pizza …, and doing stuff like that with them…yeah, they loved it… they always say, when is that lady coming again, because she was so good with them.'*
>
> ***(INT_04PAR)***

Local education workshop

As previously discussed in objective two and three, when discussing 'how' to deliver the education, support and skills, the parents, CYP and professionals suggested 'cookery workshops', 'local hubs' and 'support groups'. Parents suggested these

ventures could be held at a local school, the shared-clinic, or another venue that provides easy access close to the family home. If the children's renal dietitian and EPI champions facilitated these workshops, with support from other members of the multi-professional team, this could offer parents a forum to access 'one-stop' professional support. In addition, as families learn together, exchange experiences and co-create new ideas and skills, peer support would be realised.

Delphi consensus statement 33a (S33a/R2) and 50 (S50/R2) corroborated parents' and HCPs' desire for EPI education and support networks to be held at a venue close to the family home or shared-care clinic

Category: delivery	**Median LOA rating (1-4)**	**Very high agreement (4)**	**High agreement (3)**	**Moderate agreement (2)**	**Low agreement (1)**
S33a/R2	For those who would choose to access it, an EPI support network should be set up for parents, children and young people in their local area or outpatient clinic.				
	4	86.4 (H84.6/P88.8)	9.1 (H7.7/P11.1)	4.5 (H7.7/P0)	
Category: other considerations	**Median LOA rating (1-4)**	**Very high agreement (4)**	**High agreement (3)**	**Moderate agreement (2)**	**Low agreement (1)**
S50/R2	EPI workshops could be offered at local schools/shared-clinics/or other suitable venues close to the family home.				
	4	77.3 (H70/P88.8)	18.2 (H30/P0)	4.5 (H0/P11.1)	

Table 55, p. 296 provides a summary of the ideal locations to deliver the EPI education, support and skills strategy workshops.

Table 55. Summary of the best locations for delivering the EPI

Where to deliver the EPI strategy	
A 'one stop' clinic	Parents requested an appointment at a clinic close to their home which gave access to the relevant professionals in one place
Home visits	Bespoke home visits would allow advice to be tailored to the unique needs of the child and family
Education and support workshops/hubs	A venue should be selected that best suites the family i.e. the shared-care clinic, tertiary centre or a local school

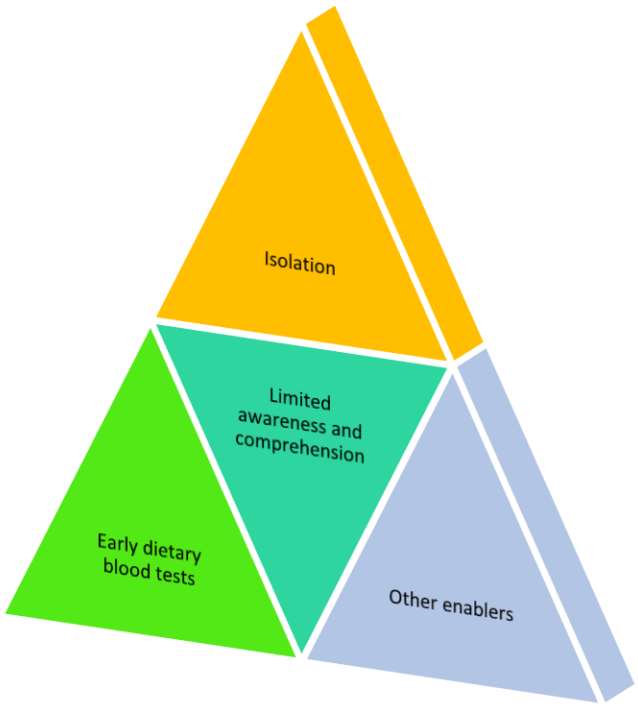
8.2.7 Objective 7: Identifying challenges and enablers

This section addresses the seventh research objective for this study.

RO7: To identify the enablers and challenges to implementing an EPI strategy

Figure 27 captures the themes for objective 7, with each theme addressed in turn.

Figure 27. Objective 7 themes



This objective, which sought to identify enablers and challenges to the development and delivery of an EPI strategy, was addressed during all three phases of the study.

This doctoral study identified some actual or perceived challenges to EPI and suggested solutions to address these issues.

Isolation

Once the intensity of the early CKD diagnosis period subsided, with the often-chaotic timetable of appointments and information bombardment, parents felt overwhelmed by a sense of vulnerability and uncertainty as to how they could impact change in the early stage, or how to access psychosocial support from HCPs or other parents. These feelings of dis-connect, wondering 'will anyone be in touch?' and 'does anyone really care?' created feelings of isolation (INT_05PAR). In an attempt to make sense of her feelings, this mother of twins concluded her 'landlocked' feeling was compounded by the prematurity of online search engines.

> *'…I think it was quite a big responsibility, and we just kind of tried to do what we could, really. I think, now…in them days Google and things like that wasn't a big thing. Like now I'd probably be able to research things myself… for me personally I was still, I had the fact that I had twins, so I needed, I was fairly isolated in the fact that I had twins and then I had this other issue as well.'*
>
> ***(INT_05PAR)***

Isolation was exemplified in the disappointment of this mother over the poor public perception of CKD, which was often framed through skewed stories in the media.

> *'It was like hitting a brick wall, and having all this information and thinking, 'oh my goodness' just completely overwhelmed … and you get really rubbish storylines on Coronation Street where somebody has been transplanted in about a week, and they're up and about… I can't watch it'*
>
> ***(INT_03PAR)***

She had to confront widespread ignorance from her family and friendship groups on a regular basis. Although they were sympathetic towards her daughter's situation, she still experienced a profound sense of isolation and loneliness because people could not relate to, or understand what she was going through.

> *'Unless they'd experienced it themselves … it almost feels as if when you've got this diagnosis there is not a lot of information out there, and I don't really know who to talk to about it'*
>
> ***(INT_03PAR)***

Corroborating these findings, parents in the Coleman et al. (2010) tailored diabetes prevention program in the Phase 1 systematic review, expressed frustrations over the lack of understanding and reciprocity from family members.

Beyond the early stages, isolation continued to present as a challenge. Despite attending regular clinic reviews for advancing CKD or home peritoneal dialysis, there was no opportunity to 'really connect' with anyone. For this mother, to be 'seen' implied connecting meaningfully with those that could offer her the much needed help and support that she was 'seeking'. She implied that clinic appointments involved a checklist of events: after clerking in at reception, you're seated in a crowded, noisy waiting area with numerous other families attending different specialist clinics. When your child's name is called you're escorted to the weighing room and given a urine sample bottle, before heading back to the waiting area. Finally, after what can be more than an hour of waiting, you're taken to 'the doctor's corridor' and seated outside the consultation room. Once called in, you are 'seen' by the doctor, often with another member or two of the multi-professional team. This is often followed by another delay as you wait to be called for blood tests. Finally, without hesitation you head home.

> *'…if you've got children that are perhaps either not on dialysis but are coming to clinic or even the children that are on PD [peritoneal dialysis] you're a bit isolated really because you don't really see, you come to clinic you have bloods'*
>
> ***(INT_03PAR)***

Delphi consensus statement 48 (S48/R2) (see overleaf) corroborated this feeling of isolation during the early stages of CKD.

Category: other considerations	Median LOA rating (1-4)	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S48/R2	It is important to recognise, due to infrequent hospital/clinic visits during the early stages of CKD, parents and children or young people often feel isolated				
	4	72.7 (H69.2/P77.7)	27.3 (H30.7/P22.2)		

These prevailing feelings of isolation reported by parents in the Phase 2 interviews, and strongly corroborated in the Delphi consensus statement 48, can be addressed by improving early parent-CYP-HCP engagement, and developing opportunities to harness support. Implementation of the following proposed EPI education, skills and support ventures could help to reduce feelings of isolation, fostering better patient and family engagement and connectedness:

- web-based education (see section 8.2.3.1)
- Early face-to-face meeting with the dietitian to sow the seeds of a therapeutic relationship (see section 8.2.3.1 and 8.2.3.3)
- Local hubs or cookery workshops (see section 8.2.3.2)
- Liaising with EPI champions (see section 8.2.3.2)
- Early support networks online and face-to-face (see section 8.2.3.3)

Early dietary blood tests

While discussing the concept of EPI, many parents thought it would be 'helpful' to have an early blood test to guide dietary management. Much like the measurement of parathyroid hormone (PTH) in the Shroff et al. (2012) study in the Phase 1 systematic review, blood tests can direct dietary goals. Likewise, they can provide reassurance that parents' endeavours are making a difference - 'know[ing] that you are doing something correctly is a massive weight lifted off your shoulders' (INT_04PAR). Additionally, professionals in the Delphi survey recognised that a blood result can act 'as a powerful reinforcement with some patients' (DEL_10HCP).

Attentive to the absence of clear FGF-23 clinical interpretation measures, and more importantly resources to respond, many consultants in the Delphi intensely objected to its use. Using the metaphor of a 'firefighter' directing broad strokes to maximise

effect, one renal doctor felt the current absence of dietetic expertise in the early shared-care clinics addressing the early 'triggers' and 'root issue' of phosphate intake, means their efforts in the later stages of CKD when the damage is done, were often too late.

> *'… what's the point in finding out that these early markers are there if you haven't got people who can use them … you just can't do it with restricted resources. If you've got restricted resources then you're firefighting. So what's the point in measuring FG-23, if you can't do anything with it?'*
>
> ***(INT_07HCP)***

Interestingly, one mother in the Delphi survey who believed 'all patients should be able to access the [EPI] programme' (INT_22PAR) contested the need for a blood test when you already have a team of clinical experts.

> *'The healthcare professionals should know how to manage the diet without the blood test. Good practice will be the same regardless'*
>
> ***(DEL_22PAR)***

Delphi consensus S40 (S40/R2) firmly corroborated these findings, recognising the strong desire for EPI even without an early blood test to guide and affirm its intervention.

Category: other considerations	Median LOA rating	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S40/R2	Even though an early blood test to support phosphate dietary management is not currently used in out-patient clinics, early dietary phosphate education is essential for all children with progressive CKD.				
	4	90.9 (H92.3/P88.8)	4.5 (H0/P11.1)	4.5 (H7.7/P0)	

Limited awareness and comprehension

Practical challenges to the implementation and take-up of an EPI strategy could include a limited awareness and comprehension. As discussed in 8.2.2 when seeking to achieve a consensus definition for EPI, many parents were confused about its purpose and potential effect. The term 'healthy-eating' within the context of early

CKD is not universally understood. EPI needs to be clearly defined and reframed, with explanatory information emphasising its potential benefits.

Other enablers

Other enablers already discussed include the use of age-appropriate materials and 'home-visits' (Stapleton et al. 2001) (see section 8.2.3.2). The use of a clear, simple, realistic and positive EPI educational message (refer to section 8.2.3.1).

8.3 Summary of the integrative findings

This chapter has presented the empirical integrative findings from this exploratory sequential three phase mixed–methods study, as summarised. The perspectives of stakeholders' in this doctoral study suggest that the landscape of CKD dietary management is changing; an increased knowledge and awareness of the potential benefits of early dietary change has brought a greater emphasis on early intervention to promote 'kidney health' and delay disease progression. The early dietary phosphate self-management strategy needs to be framed within the healthy-eating message, ensuring clarity of purpose; preventing misunderstanding amongst service users and providers. The EPSM strategy, comprising a package of education, skills and support needs to be offered to all CYP with progressive CKD, and delivered by the children's renal dietitian, using evidence-based information in a clear, simple, realistic and positive format that best serves its users: written hand-outs, web-based information giving instant 24/7 access, and downloadable resources via a credible portal, such as the EMEESY website. Timely face-to-face dietetic input remains the cornerstone of dietary education and support; tailored to the changing, individual's needs, with opportunity for questions to ensure clarity and translation into everyday life. Ideally, this needs to take place in conjunction with the paediatric renal consultant during the diagnosis period in the tertiary or shared-care clinic , while leaving some 'breathing room' for the familiy to come to terms with the chronic disease. The EPSM strategy will provide CYP and parents the opportunity to make an informed choice, inviting early proactive management and control to tackle the root of the problem, rather than conceding to the CKD's demise. By encouraging buy-in from the whole family, the EPSM diet and lifestyle changes will provide families with a new framework of normality. Parents and CYP desire early engagement with the

dietitian and other HCP, and families on a similar CKD journey, with the opportunity to establish trusted relationships, and long-term support.

As a practical prerequisite to the EPSM strategy, family leaders of diet and lifestyle change desire cookery skills training at a convenient venue. Regular education/cookery workshops would provide a forum for families and HCPs to come together to receive a managable amount of information in a practical format. Peer support would be apprehended as families learn together, exchange experiences and co-create new ideas and skills. Additionally, an early support network for CYP and parents, via face-to-face and social media platforms is desired; increasing patients' and families engagement and connectedness; ameliorating feelings of isolation, anxiety and information overload.

Galvanising this doctoral study's unique contributions to the early dietary phosphate management landscape of CYP with CKD, three novel MMR themes were developed as follows:

- Theme 1 - Reframing the Early dietary Phosphate Intervention (EPI) strategy to create a new paradigm of early CKD management
- Theme 2 - The early intervention 'renal-healthy-eating' diet needs to be offered to all CYP with progressive CKD, by the paediatric renal dietitian as part of the diagnosis package, and adopted by the whole family
- Theme 3 - Culinary medicine workshops offer education, support and upskilling

Chapter 9 presents a discussion of these three novel MMR themes within the wider literature. Before presenting the conclusions of this thesis, the contributions of this study to knowledge and clinical practice, a reflexive account of the researcher's positionality, research integrity, strengths and limitations of the study, and the directions for future research, are presented.

Chapter 9: Discussion and conclusion

9.1 Introduction

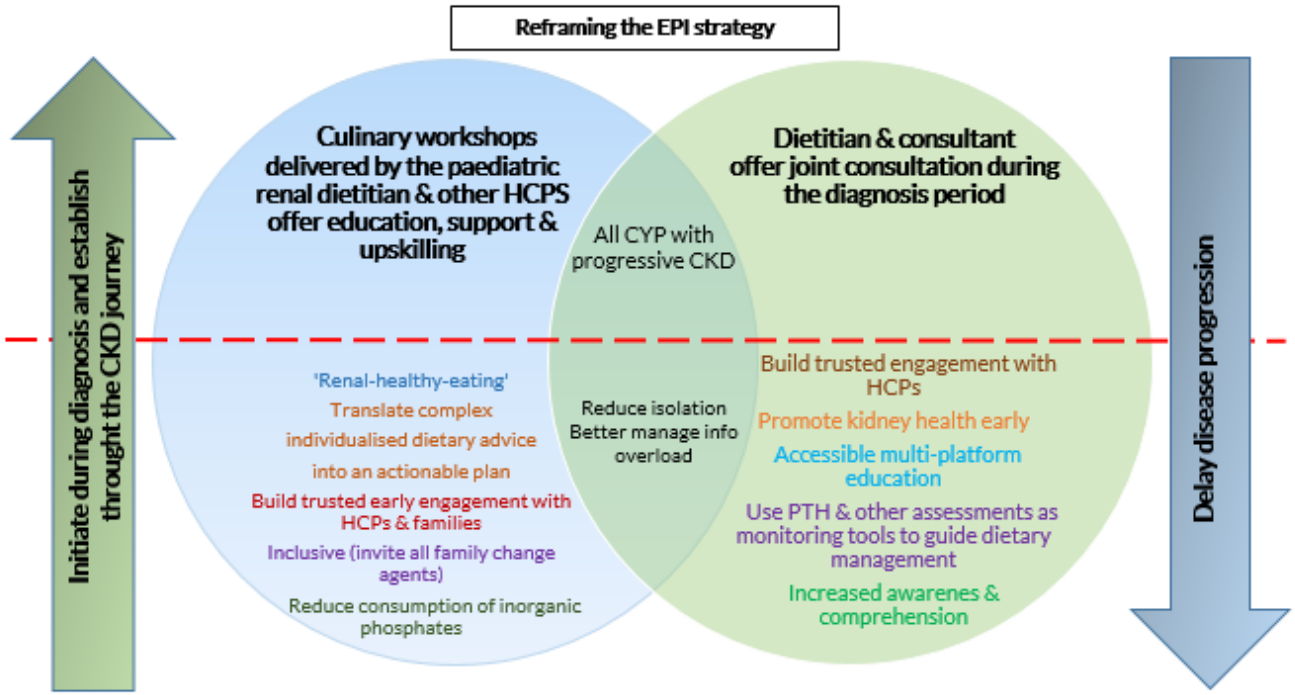
This study set out to explore the perspectives of CYP, parents and HCPs regarding the development of an early dietary phosphate self-management strategy, to delay disease progression for CYP with CKD stages 1-3, within a managed paediatric renal network. Aligned with Lorig et al's (2013) self-management theoretical framework (see section1.5.3), this aim has been achieved, with the output of some early modelling on the operationalisation of an EPSM strategy. This chapter presents this early modelling through three novel MMR themes (see below) from the integrative findings of this exploratory sequential mixed-methods study.

- Theme 1 - Reframing the Early dietary Phosphate Intervention (EPI) strategy to create a new paradigm of early CKD management
- Theme 2 - The early intervention 'renal-healthy-eating' diet needs to be offered to all CYP with progressive CKD, by the paediatric renal dietitian as part of the diagnosis package, and adopted by the whole family
- Theme 3 - Culinary medicine workshops offer education, support and upskilling

It also highlights the contributions of this research to theory and practice, and the implications for CKD clinicians and policymakers. A reflexive account of the researcher's positionality and 'insider' perspective make explicit the many nuanced power dynamics present when undertaking research with colleagues/patients, and how the researcher mitigated these challenges. The steps taken to maintain the quality of the research are detailed. Next, the limitations of this study and directions for future research are discussed. Finally, the conclusion section marks a culmination of the ideas, insights and outcomes of this thesis.

Figure 28 overleaf presents a concept model of the novel MMR themes.

Figure 28. Concept model of the novel MMR themes



As outlined in the MMR design in Figure 2, p. 21, each phase of this study informed and shaped the next; data interaction and integration resulted in the development of some early intervention modelling for the dietary management of CYP with CKD, as will now be discussed.

9.2 MMR Theme 1 – Reframing the Early dietary Phosphate Intervention (EPI) strategy to create a new paradigm of early CKD management

Integrative findings from this MMR study captured two of the centrally defining features of the EPSM strategy, education and support. This definition statement (S1), achieving consensus by the expert Delphi panellists (see section 7.4.1), captured these features:

- Early phosphate intervention (EPI) is a program of dietary *education*, engaging with children, young people and their families during the early stages of chronic kidney disease (CKD 2-3), to *support* diet and lifestyle change towards early dietary phosphate self-management

The EPSM strategy aims to provide a *dietary education* program to inform and equip CYP and parents with the tools to utilise the protective potential of diet, in delaying the progression of CKD and its related complications. The importance of introducing a dietary phosphate intervention early is aligned to the new Pediatric Renal Taskforce recommendation for children with CKD stages 2-5 and on dialysis (McAlister and Pugh et al. 2020). These pragmatic clinical practice recommendations recognised the role of EPI in the early stages of CKD to attenuate the progression of secondary hyperparathyroidism and associated cardiovascular damage in children. Paediatric and adult literature (Warady et al. 2015; Di lorio et al. 2012; Moe et al. 2011) also recognise its potential role in delaying CKD progression. It is necessary to reframe the early dietary management of CKD; the next two sub-sections outline the justifications for reframing (9.2.1 and 9.2.2).

9.2.1 Renal healthy-eating for early CKD

The EPI diet, with its 'healthy-eating-like' principles was cause for concern in this study. Many parents felt it paradoxically went against their understanding of 'diet

and CKD'. Healthy-eating encourages a greater consumption of fruit and vegetables, but the renal diet restricts a lot of high-potassium fruits and vegetables (NKF, 2009). Parents of CYP with CKD seek to provide energy-rich meals and snacks to optimise growth, but healthy-eating discouraged such foods (Shaw, 2015). The conventional healthy-eating message of decreased saturated fats, refined sugar and salt, increased fruit, vegetables and fish, and the avoidance of high-calorie meals and snacks was reportedly causing confusion, portraying a misleading message.

However, in accordance with the literature, the healthy-eating message is relevant and necessary for CYP with early stage CKD (Carrero and Cozzolino, 2014). While acknowledging the need to tailor the conventional healthy-eating message to the therapeutic needs of individual renal patients, Carrero and Cozzolino (2014) extol the many merits of healthy-eating as relevant and effective nutritional therapy for all stages of CKD; in particular, delaying its progression. In addition to the management of phosphate, which has been a key focus of this study, there are many other factors aligned to a healthy-eating diet that can delay CKD progression. In the adult literature, high consumption of saturated fat (Lin et al. 2010) and refined sugars correlate with CKD progression (Lin and Curhan, 2011). Dietary fat modifications to reduce negative saturated fats and increase n-3 and n-6 polyunsaturated fatty acids 6 (present in fish and vegetable oils) carries renal protective benefits (Huang et al. 2013). Although no current evidence relates a decreased salt intake to delayed CKD progression, it has been directly correlated with a reduction in blood pressure (Yu et al. 2012).

The literature affirms that healthy-eating CKD tailored advice should focus on a decreased consumption of processed foods, with its high inorganic phosphate content (refined sugars, salt and unhealthy fats) (McAlister and Pugh, 2020; Rose and Strombom, 2019; Isakova et al. 2013), an understanding of the ratio of phosphate-to-protein to guide optimal fish and meat choices (Miller et al. 2009), and an increase in plant-based foods. A plant-based diet (vegan) or the inclusion of small amounts of animal derived products (semi-vegetarian) (Fraser, 2009), predominantly encouraging an increased consumption of fruits (particularly favouring high-alkali fruit), vegetables, tree nuts, legumes and wholegrains, has been shown to safely and

reliably attenuate kidney injury (Goraya et al. 2013; Rose and Strombom, 2019; Passey, 2017). In the context of the present growing plant-based trend, it may be easy for patients to adopt this approach (Laurence, 2019).

Epidemiological studies in adults have consistently demonstrated that those following a predominantly plant-based diet (as advocated in the EPI diet), compared to a rich consumption of animal derived products (Haring et al. 2017; Bosch et al. 1983; Wiseman et al. 1987), have a lower glomerular filtration rate (GFR), which predisposes to a reduced risk of CKD (prevention of glomerular sclerotic changes). In one epidemiological study of adults with moderate CKD (stage 2-3) with median follow-up period of 23 years, red and processed meat consumption was significantly associated with increased CKD risk, compared with a consumption of plant-based foods (Haring et al. 2017). Similarly, in the 3-year Chilean study of 124 adults with CKD stage 3, timely, frequent and individualised dietary counselling based on general health eating guidelines (as suggested in the EPSM strategy) was proposed as the key to the preservation of renal function (Matus et al. 2019). By contrast, in the largest CKD study comparing the consumption of a low and very-low protein diet (Levey et al. 1996), minimal benefits were shown (see section 3.1.3), however the absence of detail regarding the protein source and long-term dietary compliance issues exposed legitimate concerns regarding the internal and external validity of the observational data (Evenepoel and Vervloet, 2016; Selamet et al. 2016; Menon et al. 2009). It can be concluded that a tailored renal healthy-eating message is relevant and necessary for CYP with early stage CKD.

9.2.2 EPI promotes kidney health to delay CKD progression

Similar to the concerns regarding healthy-eating for early CKD management, the notion of "early phosphate management for healthy kidneys" created a strong reaction. The fact that both parents and HCPs were vehemently opposed to this definition (see section 8.2.2), reflected the nuanced interpretation. The use of the preposition 'for', before healthy kidneys, implying the dietary intervention was for CYP with healthy kidneys, seemed to be the catalyst for the many objections, as it contradicted the core premise that, by nature of having CKD the kidneys are not

healthy. The statement sought to use 'for' as a conjunction, implying the EPSM strategy seeks the retention of current kidney function (health) and the prevention of disease decline (Carrero and Cozzolino, 2014). This realisation that CKD can be delayed is a notion not routinely communicated to parents of CYP with early stage CKD. Similarly, the belief that by delaying disease progression, their CYP may be less exposed to the associated co-morbidities of progressive CKD, such as bone and cardiac complications, is poorly understood, and by controlling phosphate early on, even if CKD continues to decline, CKD-MBD control may well be better. The term "kidney health" is gaining acceptance in the global adult CKD literature as a term to capture the scope of CKD, including those at risk for and affected by CKD (Bello et al. 2017; Levin et al. 2017). Consequently, 'kidney health' needs to be reframed within the context of paediatric early CKD. Congruently, in the Stapleton et al. (2001) study which evaluated the impact of a dietary education and behavioural change program for children with cystic fibrosis, an improvement in 'health' through better nutrition and enzyme management, positively improved weight gain, which in turn can prevent decline in pulmonary function and disease progression (McDonald et al. 2013; Levy et al. 1985). Thus, cystic fibrosis was not absent, but its complications were reduced, improving the 'health' and well-being of the children.

Therefore, long-term diet and lifestyle modification in line with the EPSM strategy will involve a paradigmatic shift in the family's eating habits (Carrero and Cozzolino, 2014), and the HCP's mind set towards the management of early CKD. The EPSM strategy will encourage the EPI diet as a tool to promote kidney health, delaying disease progression and its associated co-morbidities.

9.2.3 Theme summary

Integrated empirical research outputs from this MMR study substantiate the desire of CYP, parents and HCP for an early intervention 'renal-healthy-eating diet' for CYP with early stage CKD; promoting an increased consumption of plant-based food and decreased inorganic phosphates from processed foods. Mounting evidence in the literature supports the benefits of such a diet in the management and delay of CKD progression. Consequently, to avoid ambiguity and misunderstanding, early 'renal-healthy-eating' as a tool to promote 'kidney health' and delay disease progression

needs to be better communicated to CYP and parents, and all those involved in the management of CKD. A new paradigm of early intervention, as shown below in Table 56, justifies the early CKD 'renal-healthy-eating' plan to promote kidney health, reduce inorganic dietary phosphate load and delay disease progression.

Table 56. Comparison between the current and the proposed new paradigm of early CKD 'renal-healthy-eating' dietary intervention

	Current early CKD paradigm (CKD 2-3)	**New early CKD paradigm (CKD 2-3)**
Rationale	A normal serum phosphate level suggests there is no need for dietary intervention until serum levels start to rise.	Early renal-healthy-eating is for all patients with progressive CKD, to promote kidney health, reduce inorganic dietary phosphate load, and delay disease progression. Blood tests and assessments, such as PTH and blood pressure, can be used as monitoring tools to guide dietary management, but should not determine when to start dietary intervention. When FGF-23 becomes available as a clinical tool, its role in the early dietary management of CKD needs to be considered.
Goal	Treat life normally, no dietary changes needed.	To delay elevation of PTH and serum phosphate. Promotion of kidney health, to delay disease progression and the onset of associated co-morbidities, such as bone and cardiac complications.
	Minimal referral for paediatric renal dietary advice	The family should be motivated and empowered by the upskilling and support to prepare home-cooked meals containing meat, fish and dairy, with an increased consumption of vegetables, fruits, grains and nuts, reducing intake of inorganic phosphates from processed foods.

9.3 MMR Theme 2 – The early intervention 'renal-healthy-eating' diet needs to be offered to all CYP with progressive CKD, by the paediatric renal dietitian as part of the diagnosis package, and adopted by the whole family

If early nutrition education is a critical component in developing healthy-eating habits and behaviours for healthy children (Murimi et al. 2018), it is even more important for children with CKD and their predicted reduced life expectancy (Warady and Chadha,

2007). The earlier the 'renal-healthy-eating' message is introduced, the earlier CYP and their family can build the knowledge and skills needed to perform desired behaviours that track to later stages of the disease and into adulthood. Establishing good eating and lifestyle habits to prevent and manage CKD aligns with *Next Steps on the Five-Year Forward View* (2017), which is also echoed in *The NHS Long Term Plan* (2019) that seeks to promote a strong start in life for CYP. Failure to provide early dietary intervention for CKD, with its primary focus on inorganic phosphate management, until the late stages of CKD, may be a missed opportunity to slow progression of the disease and its associated morbidities (Evenepoel and Vervloet, 2016). The four elements of this second MMR theme (timing, target group, family buy-in and intervention provider), are discussed in the following four sub-sections (9.3.1-9.3.4).

9.3.1 Part of the diagnosis package

Parents unanimously agreed (see section 7.4.2) that 'dietary phosphate education is a long-term process and should begin in the early stages of CKD'. More specifically, CYP, parents and HCP were united in their belief that it should be introduced during the diagnosis period. Analogous to the literature on dietary management in diabetes (Smart et al. 2009; Dyson et al. 2011; Gilbertson et al. 2010), which states that 'all people with diabetes should be provided with quality professional education on medical nutrition therapy upon diagnosis, and at regular intervals thereafter' (Gilbertson et al. 2010), stakeholders in this doctoral study agreed that the EPSM strategy is fundamental to the effective management of CKD, and should be introduced as part of the CKD diagnostic package. However, in accordance with the literature (Tong et al. 2009), it was also noted that during chronic diagnoses, some parents may need more time to comprehend and come to terms with CKD before receiving the EPI dietary advice.

Accordingly, EPI dietary education needs to be offered during the early diagnosis period and on frequent occasions thereafter, responding to the changing and evolving (growth and development) needs of each child and their family (Gilbertson et al. 2010). This 'drip feeding' of information and education, frequently, over time, in

an engaging manner, using accessible multi-platform, age-appropriate resources (see section 9.5.2.3 for the implications for clincial practice) is affirmed in the literature (Huby et al. 2016; Beto et al. 2016). As patients become more and more familiar with a message through regular exposure, adaptation is increased causing the message to become so ingrained or habitual, they think less and less about it (Beto et al. 2016). Additionally, this early knowledge exchange and role negotiation between CYP, parents and HCP is pivotal to patients shared management (Swallow et al. 2013).

9.3.2 EPI is for CYP with all stages of CKD

Similar to diabetes management, specialist dietetic education is required at the initial diagnosis of type 1 diabetes, with follow up 2-4 weeks later and regular (at least annual) reviews thereafter (Gilbertson et al. 2010). Unlike in CKD where the function of the nephrons can either remain stable for many years, or rapidly decline, in type 1 diabetes, once diagnosed, there is a progressive and predictable loss in insulin secretory function (Atkinson, 2012). The unpredictability of CKD decline was the reason why the majority of HCP interviewed in Phase 2 of this study were united in the belief that a measure of screening is required to identify those CYP most at risk of the disease progressing. A key issue here is to establish clarity regarding CKD disease progression. All CYP with CKD have progressive disease. Variability lies between CKD that remains stable for a while and that where there is active decline. One of the risk factor for CKD decline is the level of GFR at diagnosis (Cerqueira et al. 2014). The lower the GFR at the start, the quicker the deterioration. On this basis, many CYP with CKD 1-3 will appear to have 'stable' or slowly declining CKD for many years.

Indeed, this contentious issue reached consensus in the Delphi survey, with the majority of parents and HCP agreeing in S16 (S16/R1) that only children with actively declining CKD should be offered EPI (see section 7.4.3). However, in those CYP where it is not clear if they are actively declining, HCP concurred with parents in their belief that the EPSM will do no harm, and therefore should be encouraged. Furthermore, even when CKD decline is not clear, the 'renal-healthy-eating' principle of the EPI diet were extolled as establishing good long-term lifestyle habits that should be set within the context of the whole family.

9.3.3 EPI should be encouraged for the whole family

The 'family context' principle is consistent with current paediatric diabetes management (Gilbertson et al. 2010; Smart et al. 2009), which states that 'it is important that the whole family is involved in making appropriate changes based on healthy-eating principles' (Smart et al. 2009). Similarly, in the management of childhood obesity, a family approach, particularly parental involvement, is the factor most likely to determine effectiveness (Moore et al. 2012; Golan, 2006). The key 'leaders' or 'gatekeepers' of diet and lifestyle change can provide an in-road to persuade the entire family to buy-in to the EPI programme. Where the EPI becomes a family habit, that means everyone is healthy together. No one is 'singled' out as having different needs because of CKD, rather it encourages the promotion of a healthy mind-set rather than a 'disease' mind set. Establishing 'little changes' early in the family diet allows for the formation of long-term habits, that 'become the norm', attaining higher sustainability, ownership and success. Whole family buy-in also avoids singling out the CYP with CKD as being different, supporting their freedom to choose and desire to fit in. Feelings expressed by young people in the SONG-Kids focus group study (Gutman et al. 2018) support these findings; they wanted to 'hang out with friends, go out, have fun, be normal kids,' and thought that dietary restrictions imposed by their parents and clinicians limited their 'freedom of choice'.

9.3.4 EPI delivered by the paediatric renal dietitian offers trusted early engagement

In diabetes management, the initial dietary advice by a specialist 'pediatric diabetes dietician…as soon as possible after diagnosis to promote a secure, trusting and supportive relationship' (Smart et al. 2009), encapsulates key primacies outlined by stakeholders in this doctoral study. Parents, CYP and HCP believed early CKD dietary advice should be delivered by the 'expert', the paediatric renal dietitian. An 'expert' in CKD, implies a mandate of authority for the management of the disease (Nielsen et al. 2019). By proposing the delivery of EPSM strategy by the paediatric renal dietitian as part of the clinical diagnosis package, it would offer families the desired timely early engagement to establish the seeds of a long-term therapeutic relationship. This promise of a long-term supportive relationship with the 'expert' would provide

parents with the trusted 'lifeline' many so desperately desired (Weiner et al. 2013; Swallow et al. 2008) (refer to section 9.5.2.1 for implications for clinical practice). In the US study (Ball et al. 2005), for parents, building a good working relationship with the HCP, influenced their concordance and honest communication. It has also been shown to build parents competence to participate in their child's CKD journey (Swallow and Jacob, 2001). Similarly, in the SM challenges and support needs experienced by adult kidney transplant recipients (Been-Dahmen et al. 2018), forming a relationship of trust with HCPs was a prerequisite to an individual's willingness to receive advice. This desire for trust is reciprocal (Lynn-McHale and Deatrick, 2000). Qualitative data exploring family involvement, highlighted parents' uncertainty about their identity in the CKD management process, and their desire to be trusted to manage their own child (Swallow, 2008). This is in keeping with other studies, which found that partnerships between nurses and recipients is a condition of SM support (Bodenheimer et al. 2002; Lorig and Holman, 2003). However, harnessing collaborative partnerships can be a challenge for HCP (Ter Maten-Speksnijder et al. 2015) in traditional healthcare, where SM is viewed as a vehicle to promote compliance (Sadler et al. 2014). Plans are directed with the prescribing of clear clinical goals to the patient. Over recent decades, however, a more collaborative partnership model has emerged, with a recognition of the negotiation of shared-care roles (Swallow et al. 2013). Chronic disease patients and their family have greater access to medical information, share decision-making and expect partnership with HCP (Alt and Schatell, 2008; Holman and Lorig, 2000).

By contrast, for many parents in this doctoral study, the lack of real engagement despite 'seeing' lots of different HCPs, brought feelings of isolation. Isolation, a recognised 'subjective, aversive, unpleasant experience' likely resulting from an individual's reduced connection with others (Florian and Krulik, 1991), is acknowledged in other studies of parents of CYP with CKD. In the qualitative exploration of patients and experiences and perspectives of living with CKD, and a systematic review on 'How do families adjust to having a child with CKD?' social isolation was repeatedly presented as a lifestyle consequence and burden of having CKD (Tong et al. 2009; Aldridge, 2008). Drawing on research on mothers of children with other chronic conditions, isolation and loneliness were identified as concerns

impacting mothers' psychosocial well-being. The value of connecting with other parents and HCPs via support networks and trusted relationships were recognised as a way to support these struggles (Florian and Krulik, 1991) (refer to section 9.5.2.2 for implications for clinical practice).

This need for connection amongst CYP is recognised in the paediatric diabetes literature. Children and young people in the diabetes prevention programme (Coleman et al. 2010) were given opportunity to support each other through social networking and the sharing of stories/experiences. These issues have also been raised before in the CKD adolescent literature. Tong et al. (2013) recognised the need for an integrated support service for young people with CKD, where they can connect with psychosocial and lifestyle management support, as well as accessing school and vocational help.

9.3.5. Theme summary

If the whole family are to embrace the new EPI paradigmatic diet/lifestyle change, offered and delivered by the paediatric renal dietitian during the CKD diagnosis period (refer to section 9.5.2.1 for implications for clinical practice), avoiding the singling out of the CYP with CKD, they must first be offered the promise of a long-term trusted relationship with HCPs.

9.4 MMR Theme 3 – Culinary medicine workshops offer education, support and upskilling

The third centrally defining feature of the EPSM strategy, skills training, was captured in the definition statement (S3) as 'empowered to prepare'.

- The EPI program will encourage a diet for healthy kidneys, with a reduced intake of processed foods (which are high in added phosphate). Parents and carers will be *empowered to prepare* home-cooked meals containing meat, fish and dairy, with an increase in fruit, vegetables, grains and nuts.

Throughout this MMR study culinary workshops have been esteemed for their ability to equip and empower CYP and families to better understand and adhere to medical advice (Coleman et al. 2010; Totadri et al. 2017). Training in practical food preparation teaches families how to use everyday home-based ingredients to promote a healthy

diet, while avoiding expensive, commercially prepared dietary products or unnecessary dietary restrictions (Totadri et al. 2017). Culturally appropriate recipes sought to motivate ongoing habits (Coleman et al. 2010). CYP and parents were able to consolidate their learning at the cookery workshops through social networking and the sharing of stories/experiences with other families and HCPs (Coleman et al. 2010). Similarly, the narrative review identifying strategies to promote adherence to nutritional advice in patients with CKD (Beto et al. 2016), discovered the lack of cooking skills to be a significant factor when influencing CKD patient food choice, convenience and taste. In a focus group study identifying strategies for reducing sodium intake in patients with CKD, HCP and adults with CKD suggested 'creative cooking courses' (Meuleman et al. 2014). Culinary medicine is a vogue approach (Bell, 2019; Hirsch et al. 2019) to treating a patients' health in a holistic way. In the recent review by Hirsch et al. (2019), culinary medicine provides a tool to improve treatment and prevent chronic diseases, using a healthier eating framework to upskill and increase knowledge transfer. Culinary medicine is the 'translational' science that turns information into actions (Hirsch et al. 2019; Hartmann et al. 2013).

> *'Culinary medicine integrates the art of preparing, cooking, and presenting food with the science of medicine to achieve desired health outcomes.'*
>
> *(Hirsch* et al. *2019, p. 1)*

The five elements of the culinary medicine workshops, which make up this third novel MMR theme will now be discussed in turn, namely (1) to translate the complexities of dietary advice into an actionable daily plan; (2) to be inclusive and led by the paediatric renal dietitian; (3) to form part of an individualised patient plan; (4) provide early engagement and relationship building; and (5) the frequency and location of the workshops.

9.4.1 EPI workshops would translate the complexity of the dietary prescription into a simple, realistic and actionable message

Parents in this doctoral study affirmed the complexity of the CKD, in particular during the later stages of CKD, and the need to translate it into a simple, meaningful daily

language. Hartmann and colleagues (2013) affirmed that the complexity of a dietary prescription must involve not only an exchange of nutrition knowledge, but also requires the essential translation into a simple, positive and achievable daily meal preparation plan. In their evaluation of overcoming consumer inertia, Webb and Byrd-Bredbenner (2015) shared a similar philosophy in their four essential components of an effective nutrition education message: realistic, positive, easy to understand, and actionable.

Using age and culturally-appropriate resources and an accessible format (see section 9.5.2.3 for implications for clinical practice), EPI workshops would provide a forum to educate families on the how to 'tackle the root of the phosphate problem'. While being upskilled to prepare foods, they would be provided with a clear rationale for following it through. This desire for a 'local hub' or cookery 'workshop' would help to address the lack of cooking skills and the reliance on processed foods, bothof which are potential concerns of following the EPSM strategy. In the narrative review by Beto and colleagues (2016) identifying strategies to promote adherence to CKD nutritional advice, a lack of cooking skills was found to be a significant factor influencing patient food choice, convenience and taste. Beto et al. commended hands-on interactive nutrition sessions as 'creative action' where patients can see and discuss foods, creating a powerful tool to 'greatly enhance' dietary intake (Beto et al. 2016).

Limited cooking skills is known to be associated with an increased use of convenience foods (Hartmann et al. 2013). The literature elucidates that the rising trend towards a reliance on inexpensive, highly palatable processed foods (Brunner et al. 2010), reflects the increasing number of working women, and a decline in the inter-generational transmission of basic cooking skills and formally taught cooking classes in schools (Hartmann et al. 2013). Additionally, a feeling of 'time scarcity' has driven the adoption of more time-saving food consumption behaviours (Jabs and Devine, 2006). By contrast, families with cooking skills consume more vegetables and less processed foods (Hartmann et al. 2013), which directly support the upskilling rationale of the EPSM strategy.

9.4.2 EPI workshops will be inclusive and led by the paediatric renal dietitian

Mindful that families have a wide variety of "set ups", the fathers, grandparents, guardians and siblings can all represent the 'main leaders' of dietary change. Identifying these food "gatekeepers" is crucial if dietary advice is to take effect (Beto et al. 2016). Involving the whole family, including grandparents and carers, in the workshops would ensure communication, knowledge and skill development extend to the primary food purchasers and preparers.

Parents felt it would be important to include the children, as it would be fun to do it together and would make them feel important. Hartmann et al. (2013) highlighted the value of teaching children and teenagers how to cook, instilling life-long skills to raise awareness of how to prepare healthy foods economically and quickly. Involving CYP, empowers and equips them to be competent participators in the management of their health (Huby et al. 2016). Consistent with these findings Gutman et al. (2018) evidenced how parents appreciated it when clinicians encouraged their child to take some independence and responsibility for SM. Additionally, as parents became more confident in their own knowledge and understanding of CKD, they are more likely to pursue opportunities for greater involvement and control (Gutman et al. 2018).

In line with the delivery of the EPSM diet by the paediatric renal dietitian, during the diagnosis period, as discussed in MMR Theme 2 (see section 9.3.4), likewise the EPSM culinary workshops would be led by the 'nutrition experts'. Support from the multi-professional paediatric renal team would further enhance and endorse the EPSM message during the workshops. As demonstrated in the Stapleton et al. (2001) study, the multi-professional team provided long-term involvement to support knowledge acquisition and retention of SM skills. Furthermore, this multi-professional team approach upholds Lorig *et al's* (2001) holistic model, which targets individuals with long-term chronic diseases, supporting the development of their individual knowledge and skills to share power with the HCPs over the management of their disease. This idea is also consistent with Stepans *et al's* (2002) multi-professional 'early intervention team' approach in the complex management of young children with disabilities. A multi-professional team approach provides opportunity to build

relationship with HCP and other families, while learning new skills and acquiring knowledge.

9.4.3 EPI workshops would be part of an individualised plan

The culinary workshops would be offered as part of an individual's dietetic prescription. Parents who are already preparing home-cooked meals may not need to attend the culinary workshops to upskill, but may want to attend to develop long-term relationships and access social support, whereas busy working mums with 'time scarcity' may choose to access the workshop to seek easy, realistic family meal ideas (Jabs and Devine, 2006). As Havas et al. (2018) argued, CKD SM is about meeting patients where they are by 'tailoring support to improve knowledge, skills, and confidence'. Likewise, Modi et al. (2012) attested SM involved CYP developing a wide range of attitudes, health behaviours and skills, to help minimise the impact of their condition on all aspects of life for themselves and their family/caregivers. In line with previous studies, the delivery of nutritional advice in CKD is most effective when tailored to the specific needs of the CYP and their family (Beto et al. 2016; Been-Dahmen et al. 2017). The provision of face-to-face individualised advice that remains consistent, creative and evolving, is the hallmark of promoting adherence to CKD nutritional advice (Beto et al. 2016). The face-to-face communication desired by parents, has been evidenced to develop the long-term relationship between the family and HCPs (Swallow and Jacob, 2001), building confidence and competence for the CKD journey.

9.4.4 EPI workshops offer early engagement and relationship building

The workshops will further respond to CYP and parents' request for engagement and an early support (see section 8.2.3.3) (also refer to section 9.5.2.2 for implications for clinical practice). Promoting buy-in from the whole family will provide opportunity to meet other families and HCP, addressing issues of isolation. Connecting with HCP and other families in an informal setting will further provide opportunity to build long-term therapeutic and supportive relationship (see section 5.1.2.3 and 8.2.3.3) with

someone who understands the CKD journey. These findings are in line with previous studies where CKD participants desired contact with fellow patients, to share and discuss experiences and lifestyle strategies (Been-Dahmen et al. 2018). Supportive therapeutic relationships with HCP are recognised as a 'vital' lower-level psychosocial need of patients with CKD (Taylor et al. 2016; Watson, 2014). Given that parent functioning and well-being are established predictors of child health outcomes across a range of serious illnesses (Muscara et al. 2015), parents and patients expect their healthcare team to provide psychosocial and strong relational support (Beto et al. 2016), desiring a greater focus on psychosocial input (Tong et al. 2009). Early parental/CYP connection with "others" establishes trust, builds competence to participate in SM, and the confidence to successfully advocate for their child's needs throughout their CKD illness trajectory; it is also recognised as an important determinant of adherence (Beto et al. 2016; Swallow and Jacob, 2001). The workshops would 'harness the power of families mentoring each other' (DEL_HCP06). Those who are motivated and do well could become EPI champions, inspiring and encouraging other to follow suit. The effectiveness of peer support in providing practical, emotional and social assistance to CKD patients may be attributed to the non-hierarchical, reciprocal relationships generated by the sharing of experiences and knowledge with others who have faced or are facing similar challenges (Green and Boulware, 2016).

9.4.5 Frequency and location of the EPI workshops

Frequency of the EPI workshops was proposed at least annually, but ideally as required, in response to the CYP and families need for education and support. The best location for the workshops was argued as a place that gave the family optimal access to 'timely information', with opportunity to ask questions of the relevant professionals all 'in one place' (DEL_19PAR). The 10-week diabetes prevention program (Coleman et al. 2010) included 2 food demonstrations at local schools. By contrast, in the prospective non-randomised controlled study of a diabetes risk reduction programme (Simmons et al. 2003), four cookery demonstrations were held over the 2-year study period in local church halls. By contrast, a more intensive package was offered to adult CKD patients, as part of an interdisciplinary cooking and

exercise program, delivered at a local community venue (Flesher et al. 2011). The cooking classes ran over 4 weeks for 2 hours per session, with a dietitian-led shopping tour during an additional week. Each cooking session focused on a different topic (self-management, food labels/eating out, phosphate, sodium, potassium, protein), with dietetic-led education, and a chef directing participants in the preparation and tasting of CKD recipes. Home visits were also suggested as a good venue to provide education and support. The burden of travel-related costs was highlighted as a source of anxiety in a recent qualitative study accessing the parental perspective on the financial impact of caring for a child with CKD (Medway et al. 2015). In an effort to significantly reduce travel time and the associated costs, while providing a multi-disciplinary interface, a group in Brisbane, Australia offered videoconferences at the local hospital (Smith et al. 2003). Home visits were suggested as a possible option for education and support. The importance of home visits by paediatric renal team members to impart and update knowledge has long been recognised (Watson, 1995). However, financial pressure on services has eroded the continuity of care between the hospital and community (BMA, 2016) and limited the opportunities for domiciliary care.

9.4.6 Theme summary

Inclusive, accessible and timely EPSM culinary workshops, led by the paediatric renal dietitian, can provide a manageable amount of knowledge exchange, while translating the complexities of the renal-healthy-eating diet into a simple, realistic and actionable message, as part of an individual's dietetic prescription. This will support the translation of information, ameliorating feelings of information overload. The workshops can address the practical prerequisites of the strategy, upskilling those families with 'limited' or a 'lack of cooking skills'. The opportunity to engage with other families and HCPs can address issues of isolation.

9.5 Contributions to Knowledge and Practice

In addition to speaking to the research question, with its aim and seven objectives that delineated the scope of this thesis, the empirical integrative findings and three

novel MMR themes from this study provide several contributions to knowledge and practice, as highlighted next.

9.5.1 Contributions to Knowledge

This is the first MMR study to explore the perspectives of CYP, parents and HCP towards the development of an EPSM strategy. The findings offer a comprehensive, detailed and holistic insight into the multi-stakeholder perspective of whether an EPSM strategy is warranted, and what it could and should look like, which has not been reported previously in the global arena. In this emergent area of research, although a direct correlation exists between FGF-23 and disease progression (Portale et al. 2016; Tsai et al. 2018; Kendrick et al. 2011), and a reduced phosphate intake (organic and inorganic) has demonstrated some positive impacts on reducing FGF-23 (Moe et al. 2011; Di Iorio et al. 2012), there is presently no clinical trial demonstrating the direct relationship between phosphate intake in the early stages (CKD 2-3) and CKD progression. However, this study provides 'real-world' mixed-methods findings to support the feasibility of developing an EPSM strategy. The findings propose that stakeholders believe all CYP with early stage CKD, especially those with an actively progressive diagnosis, would benefit from the EPSM diet, offered as a joint-consultation during the diagnosis period with their consultant paediatric nephrologist and specialist paediatric renal dietitian; allowing them to make an informed choice about early dietary intervention.

Aligned to the Medical Research Council's (MRC) Complex Intervention Framework (2008), this study has addressed the three stages of "Intervention Development": identification of existing evidence, identification and development of knowledge, and some early modelling. Thus, these MMR empirical research findings provide the crucial groundwork and the emergence of some early modelling on the operationalisation of an EPSM strategy for CYP with early stage CKD, namely, the definition, 'how' and 'who' should deliver the strategy, its recipients, and the best time and location to deliver. This study provides the rationale as informed by Hoddinott (2015) and addresses the key decision-making processes that form the early phase of intervention development, something that studies seldom report. These real-world

mixed-methods intervention development findings provide evidence that can be used to present an EPSM strategy argument to commissioners and other decision-makers.

9.5.2 Contributions to Practice

In addition to the contributions this study makes to theory, the integrative MMR findings are relevant to CKD practitioners. Four specific themes constitute the principal practice implications, as described below.

9.5.2.1 Early dietary phosphate intervention as part of the diagnosis package

Following from the second novel integrative MMR theme (see section 9.3) CYP, parents and HCP desire the early 'renal-healthy-eating' strategy to be offered by the paediatric renal dietitian as part of the diagnosis package and adopted by the whole family. This has key implications for practice. Firstly, all paediatric renal healthcare practitioners involved in the early management of CKD need to be educated on the EPSM strategy, including:

- the evidence based rationale underpinning the strategy
- the EPSM dietary approach, and
- the need to offer all CYP diagnosed with early stage CKD, especially those with an underlying progressive diagnosis, an early joint consultation with the doctor and dietitian

By responding to the 'new CKD family's' desire for dietary intervention, the paediatric nephrologist can liaise with the dietitian regarding a joint consultation. As the majority of patients with early stage CKD are seen in the EMEESY shared-care clinics, geographically spread across the region, the consultant paediatric nephrologist leading the individual CYP's management needs to partner with the paediatric renal dietitian when seeing all early stage patients. This requires recognition by the specialised commissioners across the EMEESY region of the need for specialist paediatric renal dietetic input in the diagnosis period; and thus, the funds to commission this service. As this will take specialist dietetic personnel out of the acute tertiary setting, consideration should be given to the appointment of a dedicated specialist early intervention paediatric renal dietitian to address the needs of the

network. Those families that choose not to embrace the EPSM strategy should be given the opportunity to join an early dietary intervention support network.

9.5.2.2 Early dietary intervention support network for all patients/families with early stage CKD

Following the objective 2 response (section 8.2.3.3), it is clear that some parents of CYP with CKD desire an 'early support network'. This reflects their limited interaction with 'experts' between their scheduled hospital/clinic appointments, and the subsequent isolation that many parents experience. The routine of living with CKD threw up many questions, prompting parents to want to reach out to others in the same position as them, or to seek reassurance and expert responses. Additionally, many CYP want to be instrumental in the management of their disease (Healthy Children: Transforming Child Health Information, NHS England, 2016; Lambert et al. 2013; RCPCH Voice Matters, 2020). To meet these needs, face-to-face workshops and/or an online early support network will need to be developed.

However, it is important to recognise the mismatch that can exist between the clinicians drive to best manage each CYP's condition, and the parent/CYP's desire for privacy and choice. Some parents and CYP may not want to connect with a virtual or face-to-face network; preferring to attend their scheduled clinic appointments, and then separate their CKD 'health…and the rest of their lives'. Others may wish to join an online group, but choose not to contribute to posts, but to watch the activities from the sidelines. For CYP who choose to remain anonymous, an early intervention texting hotline or managed chat room, would allow them to voice their opinions or gain access to support from peers and HCPs.

9.5.2.3 Development of a range of tools ranging from simple to complex to support maximum understanding and adherence to early dietary intervention

Following from the response to objective 2 (section 8.2.3.1), CYP and parents desire a range of credible online information, tools and resources to enable them to understand and adhere to the EPSM strategy. The EMEESY Facebook page and the Renal Patient Support Group (Muhammad et al. 2014) provide virtual environments for parents of CYP with CKD, and the CYP themselves, to connect to one another

without the need for a physical presence. Many educational tools and electronic SM resources are available to support patients with CKD (Ong et al. 2013; Huby et al. 2016). These need to be considered, or new tools developed, in order to best provide clear, simple, realistic and positive EPSM educational tools and resources. Other possible resources include hard copies of early intervention recipe booklets, a credit card–sized early dietary intervention guide and multiple platform written and audio support resources.

9.5.2.4 CKD information overload

Following from the first qualitative theme in Phase 2 of this MMR study (see section 5.1.2.1 and 5.1.2.2), CYP and parents described feeling overwhelmed by the content and volume of information they received during the early stages of CKD. In their engagement with 'new CKD families', while recognising the importance of information in the early stage of CKD, clinicians need to be aware of the emotional impact of information exchange during the diagnosis period. Consideration should be given to the potential causes of information overload, such as low education level, health literacy, ethnicity and socioeconomic status (Khaleel et al. 2019). Interventions to address these issues should be considered, such as those mentioned above (see section 9.5.2.3).

A reflexive account of the researcher's positionality will now be discussed.

9.6 Reflexivity and power relations when undertaking research with colleagues/patients

In order to make explicit the researcher's positioning, and its influence throughout the research process, a reflexive journal was maintained on the research journey, capturing personal thoughts and perceptions to maintain objectivity and increase study reliability (Creswell, 2007). The researcher acknowledges that her experiences and worldview influenced her decision to research early dietary phosphate intervention in CYP with CKD. As a parent of three children, early nutrition education and lifestyle play a critical and emotive role influencing adult decision-making structures, and can generate strong power dynamics in family life. Additionally, as a 'foodie' and keen advocate of the plant-based diet, her semi-vegetarian lifestyle

shaped her convictions of the EPSM diet, and its potential to alter the trajectory of disease. Championing an 'unprocessed' semi-vegetarian diet in a culture of 'convenience', gave her some insight to the struggle parents face. However, having worked as a specialist renal dietitian in a renal unit for over twenty years, she was aware that having a CYP with CKD introduced a very unique unfamiliar dynamic.

Throughout the research journey her unique set of values, worldview and professional perspectives (Polit and Beck, 2018) brought to the inquiry aspirations for the children's renal service. As a mother, renal dietitian and health advocate, it became clear that trying to influence dietary change is critical and complex, and does not occur in a vacuum, but involves and is affected by numerous people and structures. She had established a professional relationship with many of the participants over the years, viewing many as friends/colleagues with whom she had shared emotional stories of life's journey. During the research she was mindful of these past experiences/journey and how this had shaped their views. The reflexive journal helped her to identify these moments and determine not to allow such emotive elements of her *a priori* personal and professional knowledge and perspectives to influence the favouring of certain opinions over others. Furthermore, during her interviews with participants, she was constantly mindful of her assumptions and the shared understanding of vague comments, innuendoes and incomplete sentences in response to a particular line of questioning (Kanuha, 2000), and made every effort to avoid "knowing" by asking for clarification and specific detail to be articulated by the participants, even though they knew that she had the knowledge (Breen, 2007). Probing for information that the participant knew the researcher already had, occasionally appeared to aggravate (DeLyser, 2000), tainting the empathic feelings shared from knowing some participants on a deep level (Kanuha, 2000).

The researchers' 'emic' viewpoint as an insider in the renal unit (Kersen, 2016) offered a unique insight and understanding of the group's culture and subject matter (Bonner and Tolhurst, 2002), however it also generated potential power as the holder of knowledge (McDermid et al. 2014). Mindful that this could adversely affect her relationship with participants, in order to mitigate this imbalance of power she sought

to be open, honest and authentic in her experience as a dietitian, and her current role as researcher, wholly committed to accurately representing the participant's perspectives (McDermid et al. 2014). Comfortably articulating this role-duality and the present researcher status took some time for the researcher, a delicate balance with blurred boundaries. Additionally, she was careful to make every effort to employ a suspension of her own insights and views, and treat each contribution in its own right from an open and fresh viewpoint (Shelton and Creghan, 2015). McCracken (1998, p. 32) cautions researchers, by closely drawing on their own views and experiences they can be blinded by familiarity, and fail to see cultural assumptions, thus dulling their 'powers of observation and analysis'. Therefore, in order to restore a sense of mystery and surprise, it was necessary for the researcher to create a critical distance, a recognised challenge for insider-researchers (Breen, 2007), and thus create an ability to be more critically aware of issues emerging from the inquiry (Alvesson and Kärreman, 2011).

Her insider status brought with it an established trust that made recruitment easier with many CYP, parents and colleagues. In reality, the process of recruiting HCP for Phase 2 semi-structured interviews 'snowballed' and seemed to happen very quickly. It was necessary not to exploit this 'power' of familiarity, by avoiding perceptions of coercion (McDermid et al. 2014). This familiarity also meant that rapport was quickly established within many interviews, and she was mindful of the importance of honesty and transparency, while maintaining boundaries, trust and confidentiality.

9.7 Maintaining research quality (integrity and trustworthiness)

Integrity is a critical issue in research, both for the researcher and user of the evidence (Polit and Beck, 2018). There exists controversy among researchers regarding the quality criterion used to appropriately capture integrity, with quantitative purists attesting to the use of rigour and validity, and qualitative researchers preferring trustworthiness (Polit and Beck, 2018). Within mixed methods research, issues of integrity and trustworthiness seek not to find a single truth, rather to provide description of the quality-enhancing activities that were undertaken to demonstrate that the conclusions drawn, and their application are based on a clear audit trail of the evidence (Polit and Beck, 2018).

The following steps were taken to maintain the integrity and trustworthiness of this MM study.

- Multiple methods of data collection and analysis, often referred to as triangulation (Creswell, 2009), strengthened the reliability and internal validity of the findings (Creswell, 2009), supporting the capture of a more complete understanding of the research problem (Polit and Beck, 2018).
- The supervisory team reviewed the analysed data at regular intervals throughout this three-phase MM doctoral study, providing peer review, enhancing the rigour of data analyses and demonstrating the presence of interpretive reliability (Teddlie and Tashakkori, 2009).
- Collecting data from different stakeholder groups provided validation of the data through multiple perspectives (Polit and Beck, 2018).
- The interview guide for CYP and parents/HCP was piloted by a young renal patient and parent of a child with CKD, respectively, for internal validity, suitability and appropriateness (Grove et al. 2013); enhancing the construct validity of the tool.
- Throughout the research journey, the use of a reflexive journal allowed the researcher to recognise her positionality and experience, to maintain objectivity and increase study reliability (Creswell, 2007).
- While recognising the influence of her positionality on the research journey, she actively attempted to employ a suspension of her own insights and views, treating each contribution in its own right from an open and fresh viewpoint (Shelton and Creghan, 2015).
- Intensive listening to gain participant's trust, careful probing during each interview, and verbatim audio recording, all allowed for rich and comprehensive data collection, enhancing the authenticity and quality of the research (Polit and Beck, 2018).
- To enhance the internal validity of the data analysis to inform the Phase 3 round 1 Delphi statements, the researcher mapped Phase 1 and 2 data against the domains from the TIDieR checklist (Polit and Beck, 2018).
- Throughout the Delphi process the researcher maintained an analyses diary to describe and explain the formation of new statements/sub-statements and to

provide a decision making audit trail to enhance procedural auditability and research trustworthiness (Keeney et al. 2011).

- The Delphi Steering Group provided global, national and lay, peer review expertise, enhancing the rigour and credibility of data analyses, limiting subject bias, demonstrating interpretive reliability (Teddlie and Tashakkori, 2009) and increasing the external validity (Polit and Beck, 2018).
- Ongoing iteration and feedback to expert Delphi panellists, which can be viewed as member checking, was a means of enhancing the credibility of the qualitative and quantitative Delphi data analyses (Engles and Kennedy, 2007; Polit and Beck, 2018).

Next, strengths and limitations of this study and suggestions for future research are offered.

9.8 Strengths and limitations of this study and directions for future research

9.8.1 Strengths of this MMR study

This is the first known mixed-methods study exploring the multi-stakeholder perspective on the development of early dietary phosphate SM strategy in CYP with early stage CKD. It also comprises the first mixed studies systematic review with a narrative synthesis describing children's, parents' and other stakeholders' perspectives on early dietary SM to delay disease progression of chronic disease in children. A main strength of this systematic review and MMR study is that it gave CYP with CKD, their parents and a diverse group of HCPs a voice to pragmatically shape and inform the future of early dietary phosphate intervention in clinical practice. Although most of the interviewees worked or lived within easy access to the tertiary centre, a strength of the online Delphi survey meant that participants were recruited from across the EMEESY network, which covers a disparate geographical area.

For the Phase 2 qualitative section it was not necessary to have a large sample, but it was important to have varied respondents that represented differing views on the topic. Therefore, a strength of this study was the sample variation with regard to age,

sociodemographic characteristics and CKD staging, professional/primary roles and membership of specialist groups.

As a practising paediatric renal dietitian, who had an established long-term therapeutic relationships with many of the participants, the researchers' insider 'lens' gave her unique insight and understanding of the group's culture and subject matter (see section 9.6). Interpretive description (Thorne, 2008) esteems the practitioner researcher's role within healthcare.

The Delphi Steering Group, consisting of members from several specialist professional groups, and the parent of a CYP with early stage CKD, providing vital PPI (Patient Public Involvement) perspective, provided expert guidance throughout the Delphi process. Before submitting the age-appropriate interview guide to the Research Ethics Committee (REC), it was piloted by a young renal patient for suitability and appropriateness (Grove et al. 2013). Changes were made in light of any remarks.

9.8.2 Limitations of this MMR study

This study has a number of limitations. Non-English-speaking participants were not included to avoid cultural and linguistic misinterpretation. No funding was available to design the Delphi survey in different languages, or to provide an interpretation services during the interviews. This means that it is not possible to comment on the transferability and ethnocentric value of the findings to these groups. CYP from Indo-Pakistani ethnic groups are over-represented within the CKD population. There may be a specific EPSM strategy perspective among this group that was not captured. Additionally, the study was undertaken across a single managed paediatric renal network in the UK. Therefore, the findings may not be representative of stakeholders involved in the early management of CKD in CYP in other settings.

Some expert Delphi panellists who rated a statement with low agreement, either did so due to their lack of knowledge about an issue and their inability to make a valid judgement, or because of a lack of clarity about what the statement was trying to say. As a result, they seemed to impose on item one of the Likert scale, their own value judgement, not the value described (see section 6.7.8). Additionally, panellists who rated a statement with low or moderate agreement and did not leave a free-text

comment to explain their decision, comprised an important 'outsider' or deviant opinion that was not captured in this study. This group could have contributed fresh insights to the exploration of an EPSM strategy.

The researcher's insider status may have influenced the involvement and responses of the participants. By reflexively accounting for this (see section 9.6), the researcher took advantage of its value and importance in pragmatically championing the perspective of service users and providers to advance the development of the early CKD service.

PPI involvement in the study genesis, Delphi Steering Group and piloting of the interview guide, provided an important service user engagement. The lack of PPI involvement in the data analysis and interpretation process meant this important paediatric renal intervention development study lacked a service user perspective from start to finish.

9.8.3 Directions for future research

This MMR study has identified several potential areas for future research:

1. A replication of the study across multiple paediatric renal units, including non-English speaking participants.
2. Until specialist early intervention paediatric renal dietitians are funded to regularly attend the EMEESY shared-care clinics, an exploration is needed of educational tools available to support virtual early dietary intervention.
3. The development of a web-based, age-appropriate, reusable learning object to support EPSM in shared-care clinics across the network. Any developments of this kind would need to be rigorously evaluated.
4. A feasibility study in preparation for a randomised controlled trial to assess the effectiveness of an early dietary phosphate intervention to delay disease progression in CYP with early stage CKD compared with standard care. The RCT would consist of 3 arms:
    - Intervention group 1: a prescribed and delivered semi-vegetarian diet, comprising one-third animal derived and two-thirds plant-based foods

- Intervention group 2: written early dietary phosphate management information and education
- Control group: current standard care (no dietetic input)

This study is intended for the future development, testing and eventual implementation of an early dietary phosphate intervention strategy, using the current working title of 'The CKD 123 PROTECT study'.

9.9 Main conclusions

This novel exploration of the CYP, parent and HCP's perspective on the development of an EPSM strategy was comprehensively achieved using a three-phase mixed-methods study. An early dietary phosphate self-management strategy, comprising education, skills training, and face-to-face and multi-platform support, is desired by CYP with CKD, their parents and HCP involved in their care. The landscape of CKD dietary management is changing. The EPSM strategy needs to be reframed to capture a new paradigm of early CKD dietary management. The renal-healthy-eating diet, involving an increased consumption of plant-based food and decreased inorganic phosphates from processed foods, is a relevant and necessary tool for CYP with CKD, to promote kidney health, and attenuate the progression of CKD and its associated co-morbidities. In consultation with the paediatric nephrologist, the EPSM strategy should be offered and delivered by the paediatric renal dietitian, during the diagnosis period. Education should be 'drip fed' frequently, over time, in an engaging manner, using accessible multi-platform, age-appropriate resources.

Early parental engagement with the dietitian and consultant would establish the seeds of a trusted long-term therapeutic relationship, influencing concordance and honest communication, building competence for their child's CKD journey. EPSM should target CYP with actively declining early stage CKD, but would do no harm if offered to all CYP with CKD, and their families. Whole family buy-in for the EPSM diet would encourage sustainability, ownership and long-term success. Small dietary changes would lead to long-term habits, where everyone is healthy together, and no-one is singled out as being different.

EPSM culinary workshops, led by the paediatric renal dietitian - supported by the multi-professional team - should be offered to CYP with early stage CKD, their parents and other family leaders of diet and lifestyle change. Workshops would equip and empower CYP and families to better understand and adhere to medical advice, by translating the complexity of the dietary prescription into a simple, realistic and actionable message. The workshops will be part of an individual's dietetic prescription, tailored to the specific needs of the CYP and their family. Engagement with other families and the wider multi-professional team at the workshops, would provide social networking opportunities, and the sharing of stories/experiences to manage the CKD journey. EPSM workshops need to be offered frequently, in response to the changing and evolving needs of the CYP and their family, in a location that best suits the family. This early modelling on the operationalisation of an EPSM strategy provides a framework to guide future research and practice. It is recommended that future researchers investigate the effectiveness of a 'renal-healthy-eating' strategy on the progression of CKD, as part of a randomised controlled trial.

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Appendix 1. Detailed search strategy example on Medline

Identifiers I (Subject headings that relate to the term 'Chronic Disease' **OR** 'Chronic Kidney Disease'):

- Chronic Disease
- Chronic Illness
- Chronicadj2(illness* or disease* or disorder*)

'OR'

- Renal Insufficiency
- Chronic Kidney Failure
- Kidney Disease

'AND'

Identifier II (Subject headings that relate to the term 'child' combined with **'OR'**:

- Child, Preschool
- Infant
- P*ediatric
- Adolescent

'AND'

Identifiers III(Subject headings that relate to the term 'Early Intervention' combined with **'OR'**:

- Secondary prevention
- Early intervention (education)
- Self-care
- Self-management (keyword)

'AND'

Identifiers IV (Subject headings that relate to the term 'stakeholder' combined with **'OR'**:

- Patients
- Parents
- Caregivers
- Nurses
- Physicians
- Consultants
- Nutritionist
- Allied Health Personnel

'AND'

Identifier V (subject headings that relate to the term 'diet' combined with **'OR'**:

- Nutrition therapy
- Diet* therapy
- Diet
- Dietary (keyword)
- Food

'AND'

Identifier VI (Subject headings that relate to the term 'disease progression' combined with **'OR'**:

- Time
- Time factors
- Disease progress (keyword)
- Endpoint determination

'AND'

Phenomena (Subject headings that relate to the term 'perspectives' combined with **'OR'**:

- Attitude
- Attitude to health
- Value of life
- Knowledge
- Perception
- Emotions
- Health knowledge, attitude, practice
- Health behaviour

Appendix 2. Mixed Methods Appraisal Tool

Category of study designs	Methodological quality criteria	Responses			
		Yes	No	Can't tell	Comments
Screening questions (for all types)	S1. Are there clear research questions?				
	S2. Do the collected data allow to address the research questions?				
	Further appraisal may not be feasible or appropriate when the answer is 'No' or 'Can't tell' to one or both screening questions.				
1. Qualitative	1.1. Is the qualitative approach appropriate to answer the research question?				
	1.2. Are the qualitative data collection methods adequate to address the research question?				
	1.3. Are the findings adequately derived from the data?				
	1.4. Is the interpretation of results sufficiently substantiated by data?				
	1.5. Is there coherence between qualitative data sources, collection, analysis and interpretation?				
2. Quantitative randomized controlled trials	2.1. Is randomization appropriately performed?				
	2.2. Are the groups comparable at baseline?				
	2.3. Are there complete outcome data?				
	2.4. Are outcome assessors blinded to the intervention provided?				
	2.5 Did the participants adhere to the assigned intervention?				
3. Quantitative non-randomized	3.1. Are the participants representative of the target population?				
	3.2. Are measurements appropriate regarding both the outcome and intervention (or exposure)?				
	3.3. Are there complete outcome data?				
	3.4. Are the confounders accounted for in the design and analysis?				
	3.5. During the study period, is the intervention administered (or exposure occurred) as intended?				
4. Quantitative descriptive	4.1. Is the sampling strategy relevant to address the research question?				
	4.2. Is the sample representative of the target population?				
	4.3. Are the measurements appropriate?				
	4.4. Is the risk of nonresponse bias low?				
	4.5. Is the statistical analysis appropriate to answer the research question?				
5. Mixed methods	5.1. Is there an adequate rationale for using a mixed methods design to address the research question?				
	5.2. Are the different components of the study effectively integrated to answer the research question?				
	5.3. Are the outputs of the integration of qualitative and quantitative components adequately interpreted?				
	5.4. Are divergences and inconsistencies between quantitative and qualitative results adequately addressed?				
	5.5. Do the different components of the study adhere to the quality criteria of each tradition of the methods involved?				

Taken from Hong et al. (2018) MMAT version 2018

Appendix 3. Tyndall's (2010) AACODS checklist for the evaluation and critical appraisal of grey literature

AACODS		YES	NO	?
Authority	Identifying who is responsible for the intellectual content. **Individual author:** • Associated with a reputable organisation? • Professional qualifications or considerable experience? • Produced/published other work (grey/black) in the field? • Recognised expert, identified in other sources? • Cited by others? (use Google Scholar as a quick check) • Higher degree student under "expert" supervision? **Organisation or group**: • Is the organisation reputable? (e.g. W.H.O) • Is the organisation an authority in the field? **In all cases:** • Does the item have a detailed reference list or bibliography?			
Accuracy	• Does the item have a clearly stated aim or brief? • Is so, is this met? • Does it have a stated methodology? • If so, is it adhered to? • Has it been peer-reviewed? • Has it been edited by a reputable authority? • Supported by authoritative, documented references or credible sources? • Is it representative of work in the field? • If No, is it a valid counterbalance? • Is any data collection explicit and appropriate for the research? • If item is secondary material (e.g. a policy brief of a technical report) refer to • the original. Is it an accurate, unbiased interpretation or analysis?			
Coverage	All items have parameters which define their content coverage. These limits might mean that a work refers to a particular population group, or that it excluded certain types of publication. A report could be designed to answer a particular question, or be based on statistics from a particular survey. • Are any limits clearly stated?			
Objectivity	It is important to identify bias, particularly if it is unstated or unacknowledged. • Opinion, expert or otherwise, is still opinion: is the author's standpoint clear? • Does the work seem to be balanced in presentation?			
Date	For the item to inform your research, it needs to have a date that confirms relevance • Does the item have a clearly stated date related to content? No easily discernible date is a strong concern. • If no date is given, but can be closely ascertained, is there a valid reason for its absence? • Check the bibliography: have key contemporary material been included?			
Significance	This is a value judgment of the item, in the context of the relevant research area • Is the item meaningful? (this incorporates feasibility, utility and relevance) • Does it add context? • Does it enrich or add something unique to the research? • Does it strengthen or refute a current position? • Would the research area be lesser without it? • Is it integral, representative, typical? • Does it have impact? (in the sense of influencing the work or behaviour of others)			

Appendix 4. Example of data extraction variables

Author
Publication year
Study aim(s)
Research design
Methodology
Sampling technique
Sample size
Sample characteristics
Setting
Data collection technique
Intervention
Key outcomes/findings

Appendix 5. Example of calculating the methodological quality of a study

Author, pub year	Types of mixed methods study components or primary studies	Methodological quality criteria	Responses				MMAT 5/11 (45%)	CEBM Quality 7/12 (58%)
			Yes	No	Can't tell	Comments		
Coleman et al. 2010	Screening questions (for all types)	S1. Are there clear research questions?	✓			Although the study follows a sequential exploratory design, the study is not described as a mixed-methods study.	I	
		S2. Do the collected data allow to address the research questions?	✓			The effectiveness of a tailored diabetes intervention program was assessed using a structured quantitative survey and qualitative interviews	I	
	5. Mixed-methods	5.1. Is there an adequate rationale for using a mixed methods design to address the research question?		✓		As there is no clear explanation that the study is following a mixed methods design, there is no clear rationale.	0	
		5.2. Are the different components of the study effectively integrated to answer the research question?	✓			There is some evidence of integration of the different components of the study in the discussion.	I	
		5.3. Are the outputs of the integration of qualitative and quantitative components adequately interpreted?			✓	The findings from the qualitative interviews supported the quantitative survey finding. However, there was no clear evidence of meta- inference of the findings.	0	
		5.4. Are divergences and inconsistencies between quantitative and qualitative results adequately addressed?		✓		This is not addressed	0	
		5.5. Do the different components of the study adhere to the quality of each tradition of the methods involved?				Discussed below		
		Qualitative component 1.1. Is the qualitative approach appropriate to answer the research question?	✓			Qualitative interviews would give access the parents views	I	
		1.2. Are the qualitative data collection methods adequate to address the research question?	✓			Structured qualitative interviews were undertaken with parents	I	
		1.3. Are the findings adequately derived from the data?			✓	The interviews were summarised into general themes, but there was detail regarding the resulting themes. There is no mention of a criterion for data analysis There was limited use of quotes, and no description of the themes.	0	
		1.4. Is the interpretation of results substantiated by data?		✓		This is not clear in the paper.	0	
		1.5. Is there coherence between qualitative data sources, collection, analysis, and interpretation?		✓			0	

Appendix 6. Renal clinic and EMEESY website recruitment poster



Appendix 7. The invitation letter sent to Parents and HCP to take part in an interview

Invitation Letter for Parents and Health Care Professionals Interview
(Final version 1.0 09.10.17)

IRAS Project ID: 215520

Title of Study: An exploration of the perspective of children, parents and key health care professionals towards the development of an early dietary phosphate self-management strategy, to delay disease progression for children with chronic kidney disease stages 1-3, within a managed paediatric nephrology network: A mixed-methods study

Name of Researcher(s): **Pearl Pugh**
Dr Martin Christian
Prof Gina Higginbottom
Dr Pippa Hemingway

Dear Parent/Health Care Professional,

I would like to invite you to get involved in some research that is currently being undertaken by the School of Health Science, at the University of Nottingham. The research is looking at the views of children, parents and health care professionals towards the development of an early dietary phosphate self-management strategy that may delay disease progression for children with chronic kidney disease stage 1-3. The project will be undertaken across the East Midlands, East of England and South Yorkshire (EMEESY) Paediatric Nephrology Network.

You were recommended for participation due to your involvement with the EMEESY Network. The research team would like you to take part in an interview. Your knowledge, experience, vision and insight is required to inform this study. It will provide you with the opportunity to share your views. This may help to improve early dietary phosphate self-management for children with CKD 1-3, which may help to delay disease progression.

The results of the interview will inform the topics that are discussed during the next phase of the study (Delphi survey). I have enclosed an information pack about the research and what is involved. However, if you have any questions, please do not hesitate to contact Pearl Pugh, the lead researcher, and she will call or email you.

I look forward to hearing from you.

Many thanks for your help.

Yours sincerely

Dr Martin Christian
Consultant Paediatric Nephrologist

Appendix 8. Age-appropriate CYP Participant Information Sheet

University of Nottingham
UK | CHINA | MALAYSIA

Participant Information Sheet for Interview with Children and Young People

(Final version 2.0 01.12.17)

IRAS Project ID: 215520

The Early Phosphate Intervention Study

Exploring children and young people's views about phosphate in the diet during early Chronic Kidney Disease

Hello,

My name is Pearl. **My name is Gina.**





We need your help with a project we are doing.

We want to learn about the food that children and young people with early kidney problems like to eat.

We would like you to take part.

What is a research project?
A research project is a way of learning about something.

Why is this project being done?
Children and young people who have a chronic kidney problem have to eat certain foods. This may help to control their kidney problem. The person at the hospital who teaches you about food and kidney problems is called the dietitian. She would like to know what you think about changing the food you eat during early kidney problems.



Why me?
You have been chosen by your doctor. They know that you have a kidney problem. They have said that you know a lot about your kidney problem and might like to share your thoughts and ideas with us. We are inviting 4-6 other children and young people like you to take part.

What will happen if I take part?
Pearl will arrange to meet with you just once for about 1 hour. She will ask you some questions. You can tell her what you like to eat and how you like to learn. You can tell her what you think by doing drawings, creating with modelling dough, writing or talking. She will ask your permission to take photos of your work.



Pearl will meet with you in your home, at the hospital or university. A place that is easy for you and your parents to get to.

She will tape record our talking. You will not be paid to take part in this project, but your parents will be given money to cover travel costs.

We will ask you to agree to be in this project by signing your name on a piece of paper called a consent form or telling us that you agree to be in the study.

This is to say that you are happy to tell us what you think about diet and learning when you have early kidney problems.

Do I have to say yes to take part?
No, it is completely up to you! Nobody will mind.

If you choose to take part and then change your mind that is OK too. You can stop at any time and you don't have to say why if you don't want to.

Are there any risks to taking part?
We want you to enjoy taking part in this project.

Age-appropriate CYP Participant Information Sheet (continued)

But talking about food and kidney problems may make you feel sad or upset. Your parents or a grown up that looks after you will be there. We can stop our conversation at any time, and start again when you are ready if you want to. We will provide lots of 'tools' like modelling dough, colouring pens, lists and pictures to help you talk about food and kidney problems.

We hope the information and ideas that you give us will help other children and young people with kidney problems.

What should I do now?

Now that you know about the project you need to think about if you want to join in or not.

Who can I ask about this?

Your parents or the grown up who looks after you have information about the project too. If they are not sure how to answer any of your questions, please let us know and we can tell you more.



If you want to ask us anything about this project please contact Pearl on:

Mobile: 07501 931391 (text or call)
Email: ntxpp9@nottingham.ac.uk

Appendix 9. Age-appropriate interview guide for CYP, and parent/HCP

University of Nottingham
UK | CHINA | MALAYSIA

Interview Topic Guide for CYP
(Final version 1.0 09.10.17)

IRAS Project ID: 215520

The topics presented below will serve as a guide to the interviews. The methods employed to facilitate the discussion will vary depending on the individual child or young person's (CYP) age, ability and personal preferences. The interviewer will be flexible, using a variety of different methods in order to help the CYP to express their views. Some CYP may wish to talk, but others may prefer to draw, write messages, lists or create a model with play-dough whilst they share their views. For all interviews, parent's advice will be sought on how best to conduct the interview with the CYP. Parents will be invited to join part or all of the interview, as desired by the parents and CYP.

Materials:

- Digital recorder
- Digital camera (to take pictures of any pictures/drawings/models done by CYP)
- Pens, crayons, paper, play-dough (for CYP to use during the interview)
- Post-it notes for children to write and post messages about the foods they like and dislike
- Graffiti book for older CYP to write messages about the foods they like and dislike
- Stop/pause/start cards to facilitate the interview
- Consent form
- Information sheet

Introduction to interview & consent process

Introductions
Introduce self
Explain what the study is about and the importance of CYP's views
The interviewer will explain that she will be talking to other child who use the same children's renal service as the interviewee
Explain what the interviewer would like to talk about

- You
- Your family
- The foods you like and dislike from a list of foods high in phosphate
- How you would feel if you had to eat less of the foods you like from the list
- How it may affect your family and social life if you had to eat less of the foods you like
- Ideas to improve your learning about diet and phosphate
- Show CYP some of the resources they can use during the interview (e.g. post-its, coloured paper, pens, play-dough etc.)
- Emphasis that there are no right or wrong answers

Assent & Confidentiality

- Explain confidentiality, including when the researcher would have to break it.
- Explain what will happen to the information collected and how researcher will maintain confidentiality
- Emphasise that CYP can skip questions, take a break, or stop the interview whenever s/he wants (where appropriate use traffic light cards to practice stopping/starting, taking a break, skipping a question, don't understand).
- Check CYP is happy to take part and ask the CYP to sign assent/consent form
- Researcher to sign promise of confidentiality on the consent form
- Ask permission to use audio recorder and set it up with CYP
- Discuss with parent and CYP what parent is going to do during the interview.

They may wish to stay in, or sit outside during the interview.

Warm up questions

Your family

- People the CYP lives with
- Ages of brothers and sisters
- Pets
- Draw map of house/family members

A bit about you

- About the child
- Likes and dislikes
- School

Questions about early dietary phosphate

Your Mum/Dad tell me you have Chronic Kidney Disease, and that you go to …… to get help with that. There are many ways of looking after your kidneys e.g. controlling your blood pressure, taking special medicine and eating the right foods. Certain foods are good for the kidneys, and other foods may not be very good. I don't know what you and your family like to eat, so today I would really like to find out what you all like.

Could you tell me what your favourite foods are?
Which foods do you dislike?
Do you like any of these foods? (List of good kidney foods and a list of bad kidney foods)
What it is like having CKD?
The impact on the family and social life

Do you think we could talk for a bit about what it might feel like if you were told to eat less of certain foods because they are not very good for your kidneys?
Would it affect the meals you eat as a family?
What would other people in your family think about this?
Would it make a difference to the places you go to eat?
Would it affect the things you do and the places you go with your friends?

Age-appropriate interview guide for CYP (continued)

Do you think we could talk a bit about how you would like to learn more about the good and bad kidney foods?

What is your preferred method of receiving new information?

- Booklet with written information
- Booklet with written information and pictures
- Computer or mobile phone game
- Mobile phone app
- Talking with someone face-to-face
- Facebook group where you can ask questions
- Phone a dietitian or renal specialist when you have questions

Other things you feel would be affected by eating less of certain foods

- School meals
- Birthday parties or special occasions
- Holidays

How you would feel when the changes you make in your diet don't make any obvious difference to your kidney disease?
Would you like to have some feedback in the form of a blood test or reassurance from the HCP that the changes you have made in your diet are making a difference?
Is there any other information you would like to know about reducing dietary phosphate and the possible delay in disease progression?
What other information would you like to have received during your early visits to the children's kidney clinic?

So you have told me all about the foods you like and dislike and how it would make you feel if you had to eat less of certain foods, now I would like to talk about what other diet stuff you would like more information on when you visit the kidney clinic.

Cool down
OK so that's all the questions I have, is there is anything else you wanted to tell me that we haven't talked about so far?

End of interview

Thank CYP for taking part in the project.
Remind CYP what you are going to do with the audio-recording. Ask the CYP if they would like to receive information about the results of the study when it has finished, and how they would prefer to receive this information (letter, photograph, email or leaflet).
Ask CYP what they would like to do with anything they have made during the interview (leave it with the CYP/have the researcher take it with them/have the researcher take photographs of what they have made).

Interview guide for parent/HCP

University of Nottingham
UK | CHINA | MALAYSIA

Interview Topic Guide for Parents and HCPs

(Final version 1.0 09.10.17)

IRAS Project ID: 215520

The topics presented below will serve as a guide to the interviews.

Materials:

- Digital recorder
- Consent form
- Information sheet

NOTE: As it is likely that interview participants will have varying levels of involvement with the children's kidney service, questions asked will be tailored to their service involvement/experience. For example, a parent with a child with CKD stage 3 may already be following a renal diet, but a child with CKD stage 1 may have no current dietary restrictions. Also, a junior doctor may have had very little involvement with children with CKD stage 1-3, but a senior consultant may have extended experience.

Consent process & introduction

Introductions

- Introduce self and explain purpose of the project
- Explain what topics the interview will cover
- Explain confidentiality
- Go through information sheet
- Answer any questions
- Explain how long the interview is anticipated to last

Consent & Confidentiality

- Explain confidentiality, including when the researcher would have to break it.
- Explain what will happen to the information collected and how researcher will maintain confidentiality
- Emphasise that interviewees can skip questions, take a break, or stop the interview whenever s/he wants
- Check the interviewee is happy to take part and ask the interviewee to sign the consent form
- Researcher to sign promise of confidentiality on the consent form
- Ask permission to use audio recorder and set it up with the interviewee

Warm up questions

Parent

- People the interviewee lives with
- Ages of children

- Job

HCP

- Job
- How long they have worked with children with CKD

Family background (parents)

- Who is in the family/house?
- Age of other children
- Family members nearby
- Involvement of family members with the child's diet
- A typical mealtime

Background to child's condition (parents)

- Confirm child's diagnosis and CKD stage
- Length of diagnosis
- Duration of involvement with the kidney clinic
- Impact of child's illness on daily life

About early dietary phosphate (parents)

- Does your child currently follow a special renal diet?
- What do you know about early dietary phosphate self-management in children with CKD stage 1-3?
- What do you understand may be the benefits of starting a dietary phosphate restriction during CKD stage 1-3?
- What do you know about a phosphate restricted diet?
- If you were offered dietary phosphate advice for your child at this stage, how would you feel?
- How would starting your child on a special phosphate restricted diet affect the rest of the family? i.e. denying one child certain foods, eating out, special occasions
- Do you think the whole family should be encouraged to follow the EPI diet?
- How would you feel about following a phosphate restricted diet when you can't see any immediate benefit (i.e. no change in the blood phosphate level)?
- Now that you know something about the potential benefits to starting a phosphate restriction at CKD stage 1-3, do you feel a diet would be empowering or imposing for the management of your child's kidney problem?
- If you were given dietary phosphate advice, how would you like to receive it? i.e. written handout or booklet, mobile phone app, website, workshops, recipe books etc.
- When do you think the EPI diet should be offered/introduced?

Background to HPCs involvement

- Current position in the renal service
- How long have they held this position?
- Level of involvement with children in the early stages of CKD (1-3)

About early dietary phosphate (HCPs)

- Do you talk to children/parents about the renal diet?
- What do you know about early dietary phosphate management in children with CKD 1-3?
- What do you understand to be the potential benefits of starting a dietary phosphate restriction during the early stages CKD 1-3?
- If your patient was encouraged to follow a dietary phosphate restriction during the early stage of CKD (1-3), would you be supportive of this?
- Should the whole family be encouraged to follow the EPI diet?

Interview guide for parent/HCP (continued)

- Do you feel there are any concerns to CYP/family/hospital in starting a phosphate restriction during the early stages of CKD (1-3)? (barriers to accessing and implementing change)
- Who should provide this early dietary phosphate advice?
- Do you feel a dietary phosphate restriction should only be started when there is evidence of its benefit?
- When should the EPI diet be offered/introduced?
- Have you heard of FGF-23?
- Do you think that we should be measuring FGF-23 levels in children with CKD 1-3?
- Now that you know something about the potential benefits to starting a dietary phosphate restriction during the early stage of CKD (1-3), do you feel this diet would be empowering or imposing to your child's/family management of CKD?
- How should dietary phosphate advice be delivered? i.e. written handout or booklet, mobile phone app, website, workshops, recipe books etc.

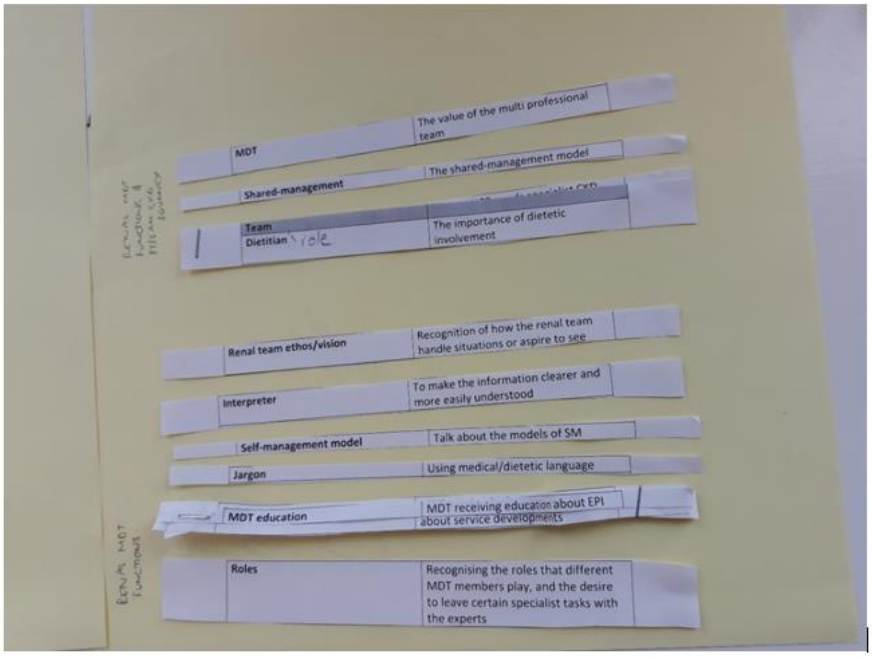
The information you have provided has been very helpful. Once all of the interviews have been undertaken and the data analysed, the next phase of the study will involve a Delphi survey. This will seek to gain consensus on the key factors that need to be considered when developing an early dietary phosphate self-management strategy for children with CKD 1-3. The Delphi will involve a series of 3 online consensus surveys that can be completed at home/work or a convenient time and place for you. Would you be interested in taking part in this part of the study?

If I haven't already got their email, ask if they would be willing to give it.

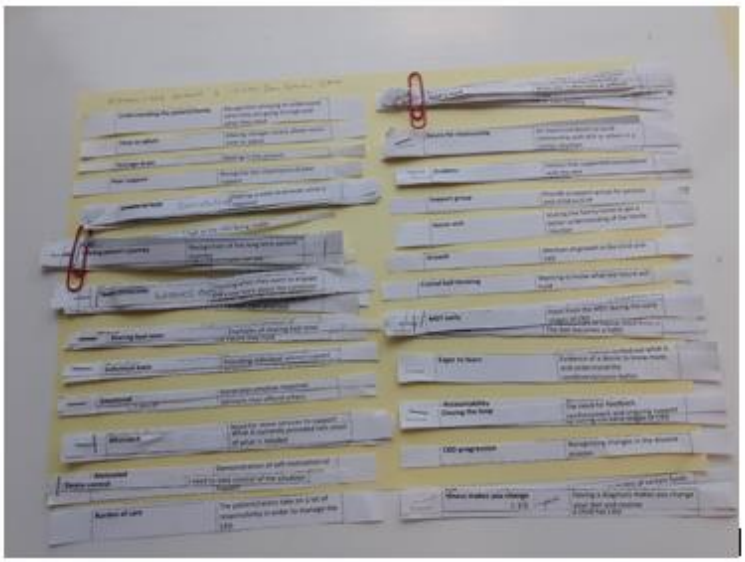
Cool down
OK so that's all the questions I have, is there is anything else you wanted to tell me that we haven't talked about so far?

End of interview
Thank parent/HCP for taking part in the project.
Remind the interviewee what you are going to do with audio-recording. Ask the interviewee if they would like to receive information about the results of the study when it has finished.

Appendix 10. Photographs of the developing data analysis process (steps 2 and 3)



- Renal multi-disciplinary team function
- Renal MDT function & child and family CKD journey



- child and family CKD journey
- knowledge
- parental role
- ways of working or making sense of the journey

Appendix 11. A section of the Word matrix showing documentation of analytical thinking

Theme 1 (HCP) - The need for support through timely early engagement and relationship-building

HCPs recognise the importance of first building a relationship with the individual and their family, before dietary lifestyle change can be encouraged. This will give understanding of the family's priorities, what they are able to handle, how it should be given, and how much information to give. The will feel valued and informed, sufficient to make an informed choice. This can be supported by telephone reviews to encourage, and motivate. Support groups, renal community role models and buddying networks would further build this early engagement.

	Sub-theme	Participant		Explanation	Quote
15	**Therapeutic relationship**	AJL_08HCP	Face to face support for parents of school aged children	HCP feel that it is important for parents of school aged children to have face to face contact, to support lifestyle change	"...it tends to be the over 5s where actually having that face to face contact I think makes a huge difference in terms of helping people to make the lifestyle changes."
16	**Therapeutic relationship/ Time is important**	AJL_08HCP	Early advice needs to be given after initially meeting the individual face to face, and having started a therapeutic relationship.	For advice to be effective it needs to be built upon a therapeutic relationship with the individual/family. Asking a HCP to call and give advice to an individual over the telephone, without having previously met the family is hard.	"...asking someone here [tertiary centre] to have a telephone consultation with someone, which I don't think – and I've not been on the receiving end of it or had any feedback, but my perception would be that that's going to be harder than having a face to face discussion with someone. And because a lot of it is on the therapeutic relationship you build with someone and it's a lot harder to do to do that if you're calling out of the blue and giving that advice over the telephone."
17	**Therapeutic relationship**	AJL_08HCP	A relationship must be established before dietary lifestyle can be encouraged	Dietary change involves lifestyle change, and that requires motivation and support. This is best given once you have built a relationship with someone; where trust and respect has been established. The specific advice and support can be tailored to their individual needs, and what works for them and also what they would find difficult to follow.	"I think it's you know diet changes are lifestyle changes and that is something that is needs motivation and support, and that is hard to give if you don't have that relationship that you've built with someone where they know you and you have that element of trust and respect for you and then you can get to know them and find out what works for them and give them specific advise and support in the areas they find difficult or you know the areas that are new to them."
18	**Therapeutic relationship**	AJL_08HCP	Telephone support can follow on from an initial face to face contact	HCP need to take the time to connect with individuals on a face to face level. Once the seeds of a relationship have been sown, then telephone consultations can help, providing valuable support and advice. If there is a local link in the shared care hospital who shares the same ethos, this relationship can be further supported.	"...it has to be done on an individual basis because some people will have that initial contact...that's where once you've build that relationship actually that's where telephone consultations can help, because they can build on that, and then be that little, you know reminder support "you're doing well keep it going". And then the other aspect is having that local person that can be the link, and if their coming with the same ethos and same input then they can support that."
	Therapeutic relationship	FH_01HCP		A therapeutic relationship allows for the provision of timely feedback to establish whether they have understood the information correctly, and answer any questions. Feedback can't just be a one off, it needs to be ongoing.	"...things like the EMEESY website are really helpful as resources, cos' we live in a day and age where people just want to look it all up on the web, don't they? So, knowing where to signpost to, for accurate information. But then, it's having that feedback loop closed, isn't it? They've got to have someone they can contact back, to say 'well actually, I didn't quite understand this, can I clarify this'. You know, you need to close that loop and make sure you're following it through and not just saying 'Here it is, get on with it'. You've got to keep revisiting it."
20	**Therapeutic relationship**	ARW_07HCP	CKD changes the family prescription forever. Relationship is key	Once an individual receives the diagnosis of CKD, their life is changed forever. The family need time and support as they come to terms with the diagnosis. Understanding the nature of their condition and whether it is progressive is important. All of this is about building relationship as you share information.	"...if you sit across the table from parents and say, 'your child has chronic kidney disease' that changes the family prescription for ever! ...So then you have to kind of get over that kind of shock, and support them with the advice. But you are in for the long haul. You know, you have to know how each child is likely to progress... it's all about building relationship, and sharing that information..."

Appendix 12. Supporting research documents

EMEESY Facebook advert



Renal clinic advert for children and parents



Letter of invitation for HCP and parent (Phase 2 - interviews)

University of Nottingham
UK | CHINA | MALAYSIA

Invitation Letter for Parents and Health Care Professionals Interview
(Final version 1.0 09.10.17)

IRAS Project ID: 215520

Title of Study: An exploration of the perspective of children, parents and key health care professionals towards the development of an early dietary phosphate self-management strategy, to delay disease progression for children with chronic kidney disease stages 1-3, within a managed paediatric nephrology network: A mixed-methods study

Name of Researcher(s): **Pearl Pugh**
Dr Martin Christian
Prof Gina Higginbottom
Dr Pippa Hemingway

Dear Parent/Health Care Professional,

I would like to invite you to get involved in some research that is currently being undertaken by the School of Health Science, at the University of Nottingham. The research is looking at the views of children, parents and health care professionals towards the development of an early dietary phosphate self-management strategy that may delay disease progression for children with chronic kidney disease stage 1-3. The project will be undertaken across the East Midlands, East of England and South Yorkshire (EMEESY) Paediatric Nephrology Network.

You were recommended for participation due to your involvement with the EMEESY Network. The research team would like you to take part in an interview. Your knowledge, experience, vision and insight is required to inform this study. It will provide you with the opportunity to share your views. This may help to improve early dietary phosphate self-management for children with CKD 1-3, which may help to delay disease progression.

The results of the interview will inform the topics that are discussed during the next phase of the study (Delphi survey). I have enclosed an information pack about the research and what is involved. However, if you have any questions, please do not hesitate to contact Pearl Pugh, the lead researcher, and she will call or email you.

I look forward to hearing from you.

Many thanks for your help.

Yours sincerely

Dr Martin Christian
Consultant Paediatric Nephrologist

Email: martin.christian@nuh.nhs.uk

Lead researcher:
Pearl Pugh, Health Sciences PhD student
Email: ntxpp9@nottingham.ac.uk
Study mobile (text or call): 07501 931391

Enc. Participant Information Sheet



Participant Information Sheet for HCP and parent (Phase 2 - interviews)

Interview Participant Information Sheet for Parents and HCPs
(Final version 2.0 01.12.17)

IRAS Project ID: 215520

Title of Study: An exploration of the perspective of children, parents and key health care professionals towards the development of an early dietary phosphate self-management strategy, to delay disease progression for children with chronic kidney disease stages 1-3, within a managed paediatric nephrology network: A mixed-methods study

Name of Researcher(s): Pearl Pugh
Professor Gina Higginbottom

We would like to invite you to take part in our research study. Before you decide we would like you to understand why the research is being done and what it would involve for you. One of our team will go through the information sheet with you and answer any questions you have. Talk to others about the study if you wish. Ask us if there is anything that is not clear.

What is the purpose of the study?

The purpose of the research is to seek the views of parents, health care professionals and children regarding the development of an early dietary phosphate self-management strategy for children and young people with early stage (1-3) chronic kidney disease (CKD). Your knowledge, experience, vision and insight is required. We would like to know what you understand about this topic, how it should be delivered, and the potential benefits and challenges of implementing early dietary phosphate self-management in children. The aim of the study is to provide information for staff working with the NHS who are in a position to improve the early dietary phosphate management for children with CKD, with the potential to delay disease progression.

What do you mean by early dietary phosphate intervention or self-management?

Early dietary phosphate intervention or self-management refers to the provision of dietary phosphate education and support for the child/young person and their family, which may improve the management of chronic kidney disease. This education and support may be delivered at the Queens Medical Centre or at your local hospital.

Why have I been invited?

You are being asked to take part in this research because you are involved with the East Midlands, East of England and South Yorkshire (EMEESY) Paediatric Nephrology Network. We would like to know more about your knowledge, experience, vision and insight towards the development of an early dietary phosphate self-management strategy for children with CKD 1-3.

Do I have to take part?

It is up to you to decide whether or not to take part. Participation in this project is completely voluntary. Whether or not you take part will not influence the current care children receive from

health services in any way. If you do decide to take part, you will be given this information sheet to keep and be asked to sign a consent form. If you decide to take part, you are still free to withdraw at any time and without giving a reason. This would not affect your legal rights.

What will happen to me if I take part?

Taking part in the project would involve you meeting with a researcher for an interview. The interview will take place at a time and place that is convenient for you. For example, the researcher can visit you at home, meet in a quiet place in the hospital, or book an interview room at the University. The interview will last approximately an hour and will be about the provision of an early dietary phosphate self-management strategy for children with CKD 1-3. All questions are optional and you do not need to answer any questions you do not want to. A digital audio recording device will be used to record the interview and written notes will be taken. Once uploaded, the audio recording of the interview will be deleted. Anonymous direct quotes from the interview may be used in the study reports.

Expenses and payments

Participants will not be paid to participate in the study. If participants incur any costs e.g. travel costs, these will be reimbursed.

What are the possible disadvantages and risks of taking part?

Sharing your views and experience of managing CKD 1-3, and discussing early dietary phosphate self-management may bring up issues of a sensitive nature. Participants can be assured that all information shared during the interview will be handled in a confidential and sensitive manner, and any future reference to information they share will be anonymised. Researchers will make every effort to ensure that the participants feel secure and are put at ease during the interview process. If you become upset or show signs of distress during the interview, you will be offered the opportunity to continue at another time or to leave the study. The participant will be signposted to the team social worker or the specialist renal nurse for support.

What are the possible benefits of taking part?

By participating in this study, you will have the opportunity to share your insights and express your views on the delivery of phosphate self-management for children with early CKD. We cannot promise the study will improve early dietetic services for children with CKD, but the information we get from this study may help us identify the benefits and challenges of developing an early dietary phosphate self-management strategy, which in turn will be fed back to individuals who manage and provide services for children, and to national policy makers.

What happens when the research study stops?

Throughout the study and once it has stopped the information and demographic data you provide will be kept confidential and will only be seen by the research team. The information collected during the interview may be used to write reports on service developments in the area of early dietary phosphate self-management. The people who take part in the research will not be named in these reports. The information collected will only be used for the purpose of this study and will not be used for any other project.

What if there is a problem?

If you have a concern about any aspect of this study, you should ask to speak to the researchers who will do their best to answer your questions. The researcher's contact details are given at the end of this information sheet. If you remain unhappy and wish to complain formally, health care professionals can contact Joanne Lymn, Head of School, Faculty of Medicine & Health Sciences, Medical School, Queen's Medical Centre, and Nottingham, NG7 2UH.

Participant Information Sheet for HCP and parent (Phase 2 - interviews) (continued)

Email:joanne.lymn@nottingham.ac.uk. Parents can contact the Patient Advice and Liaison Service (PALS), NUH NHS Trust, c/o PALS, Freepost NEA 14614, Nottingham NG7 1BR. E-mail: pals@nuh.nhs.uk.

In the event that something does go wrong and you are harmed during the research and this is due to someone's negligence then you may have grounds for a legal action for compensation against the University of Nottingham but you may have to pay your legal costs. The normal National Health Service complaints mechanisms will still be available to you.

Will my taking part in the study be kept confidential?

We will follow ethical and legal practice and all information about you will be handled in confidence.

If you join the study, the data collected during the study will be looked at by authorised persons from the University of Nottingham who are organising the research. They may also be looked at by authorised people to check that the study is being carried out correctly. All will have a duty of confidentiality to you as a research participant and we will do our best to meet this duty.

All information which is collected about you during the course of the research will be kept strictly confidential, stored in a secure and locked office, and on a password protected database. Any information about you which leaves the hospital will have your name and address removed (anonymised) and a unique code will be used so that you cannot be recognised from it.

Your personal data (address, telephone number) will be kept for 12 months after the end of the study so that we are able to contact you about the findings of the study (unless you advise us that you do not wish to be contacted). All other data (research data) will be kept securely for 7 years. After this time your data will be disposed of securely. During this time all precautions will be taken by all those involved to maintain your confidentiality, only members of the research team will have access to your personal data.

Although what you say in the interview is confidential, should you disclose anything to us which we feel puts you or anyone else at any risk, we may feel it necessary to discuss this matter with the named safeguarding representative (Dr Farida Hussain).

What will happen if I don't want to carry on with the study?

Your participation is voluntary and you are free to withdraw at any time, without giving any reason, and without your legal rights being affected. If you withdraw then the information collected so far cannot be erased and this information may still be used in the project analysis.

What will happen to the results of the research study?

The information and demographic data you provide will be kept confidential and will only be seen by the research team. The information collected during interviews will be used to inform the topics during the next phase of the study (Delphi survey – an online structured group interaction process, where experts in the field answer questions anonymously) and may be used to write reports on service developments in the area of early dietary phosphate self-management. The people who take part in the research will not be named in these reports. The information collected will only be used for the purpose of this study and will not be used for any other project. The only time confidentiality would be broken is if you tell the researcher something that suggests that you are in danger of being harmed by others. The matter will be discussed with the named safeguarding representative detailed below.

Who is organising and funding the research?

This research is being organised by the University of Nottingham and is being funded by Collaboration for Leadership in Applied Health Research and Care, National Institute for Health Research/Health Education East Midlands.

Who has reviewed the study?

All research in the NHS is looked at by an independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed and given a favourable opinion by Health Research Authority Research Ethics Committee (HRA REC Ref: 17/EM/0418) and The Nottingham 1 Research Ethics Committee.

Further information and contact details

If you would like further information on this research or have any questions before you decide whether you would like to participate, please do not hesitate to contact us using the details below. Please use Pearl Pugh as your primary contact point.

The research team

Lead researcher
Pearl Pugh's email: ntxpp9@nottingham.ac.uk
Pearl's study mobile (text or call):
07501 9311391

Researchers
Prof Gina Higginbottom
gina.higginbottom@nottingham.ac.uk
Dr Pippa Hemingway
pippa.hemingway@nottingham.ac.uk
Dr Martin Christian
martin.christian@nuh.nhs.uk

Named representative
Dr Farida Hussain
farida.hussain@nuh.nhs.uk



Consent form for HCP and parent (Phase 2 - interviews)

CONSENT FORM FOR PARENTS/HCPs
(Final version 2.0 01.12.17)

Title of Study: **Early Phosphate Intervention. An exploration of the perspective of children, parents and key health care professionals towards the development of an early dietary phosphate self-management strategy, to delay disease progression for children with chronic kidney disease stages 1-3, within a managed paediatric renal network: A mixed-methods study**

IRAS Project ID: 215520

Name of Researcher: Pearl Pugh

Name of Participant: **Please initial box**

1. I confirm that I have read and understand the information sheet version number dated for the above study and have had the opportunity to ask questions. ☐
2. I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason. I understand that should I withdraw then the information collected so far cannot be erased and that this information may still be used in the project analysis. ☐
3. I understand that relevant sections of my data collected in the study may be looked at by authorised individuals from the University of Nottingham, the research group and regulatory authorities where it is relevant to my taking part in this study. I give permission for these individuals to have access to these records and to collect, store, analyse and publish information obtained from my participation in this study. I understand that my personal details will be kept confidential. ☐
4. I understand that the interview will be audio recorded and that anonymous direct quotes from the interview may be used in the study reports. ☐
5. I agree to take part in the above study. ☐

Name of Participant	Date	Signature
Name of Person taking consent	Date	Signature

2 copies: 1 for participant and 1 for the project notes

Letter of invitation for parents r egarding interviews with CYP (Phase 2 - interviews)

Invitation Letter for Parents of Children and Young Peoples Interview
(Final version 1.0 09.10.17)

IRAS Project ID: 215520

Title of Study: An exploration of the perspective of children, parents and key health care professionals towards the development of an early dietary phosphate self-management strategy, to delay disease progression for children with chronic kidney disease stages 1-3, within a managed paediatric nephrology network: A mixed-methods study

Name of Researcher(s): **Pearl Pugh**
Dr Martin Christian
Prof Gina Higginbottom
Dr Pippa Hemingway

I would like to invite your child to get involved in some research that is currently being undertaken by the School of Health Science, at the University of Nottingham. The research is looking at the views of children, parents and health care professionals towards the development of an early dietary phosphate self-management strategy that may delay disease progression for children with chronic kidney disease (CKD) stage 1-3. The project will be undertaken across the East Midlands, East of England and South Yorkshire (EMEESY) Paediatric Nephrology Network.

Your child has been recommended for participation due to their involvement with the EMEESY Network. Their knowledge and experience of CKD are required to inform this study. I have enclosed an information pack for you and your child about the research and what is involved. However, if you have any questions, please do not hesitate to contact Pearl Pugh, the lead researcher, and she will call or email you.

If you are interested in your child taking part in this study, please call or email me or Pearl Pugh using the contact details below. We will then call or email you to answer any questions you might have.

I look forward to hearing from you.

Many thanks for your help.

Yours sincerely

Dr Martin Christian
Consultant Paediatric Nephrologist

Email: martin.christian@nuh.nhs.uk

Lead researcher:
Pearl Pugh, Health Sciences PhD student
Email: ntxpp9@nottingham.ac.uk
Study mobile (text or call):

Enc. Participant Information Sheet for
(1) Parent and (2) Child/Young Person



Participant information sheet for interviews with CYP (Phase 2)

University of Nottingham
UK | CHINA | MALAYSIA

Participant Information Sheet for Interview with Children and Young People

(Final version 2.0 01.12.17)

IRAS Project ID: 215520

The Early Phosphate Intervention Study

Exploring children and young people's views about phosphate in the diet during early Chronic Kidney Disease

Hello,

My name is Pearl. **My name is Gina.**





We need your help with a project we are doing.

We want to learn about the food that children and young people with early kidney problems like to eat.

We would like you to take part.

What is a research project?
A research project is a way of learning about something.

Why is this project being done?
Children and young people who have a chronic kidney problem have to eat certain foods. This may help to control their kidney problem. The person at the hospital who teaches you about food and kidney problems is called the dietitian. She would like to know what you think about changing the food you eat during early kidney problems.



Why me?
You have been chosen by your doctor. They know that you have a kidney problem. They have said that you know a lot about your kidney problem and might like to share your thoughts and ideas with us. We are inviting 4-6 other children and young people like you to take part.

What will happen if I take part?
Pearl will arrange to meet with you just once for about 1 hour. She will ask you some questions. You can tell her what you like to eat and how you like to learn. You can tell her what you think by doing drawings, creating with modelling dough, writing or talking. She will ask your permission to take photos of your work.



Pearl will meet with you in your home, at the hospital or university. A place that is easy for you and your parents to get to.

She will tape record our talking. You will not be paid to take part in this project, but your parents will be given money to cover travel costs.

We will ask you to agree to be in this project by signing your name on a piece of paper called a consent form or telling us that you agree to be in the study.

This is to say that you are happy to tell us what you think about diet and learning when you have early kidney problems.

Do I have to say yes to take part?
No, it is completely up to you! Nobody will mind.

If you choose to take part and then change your mind that is OK too. You can stop at any time and you don't have to say why if you don't want to.

Are there any risks to taking part?
We want you to enjoy taking part in this project.

Participant information sheet for interviews with CYP (continued)

But talking about food and kidney problems may make you feel sad or upset. Your parents or a grown up that looks after you will be there. We can stop our conversation at any time, and start again when you are ready if you want to. We will provide lots of 'tools' like modelling dough, colouring pens, lists and pictures to help you talk about food and kidney problems.

We hope the information and ideas that you give us will help other children and young people with kidney problems.

What should I do now?

Now that you know about the project, you need to think about if you want to join in or not.

Whom can I ask about this?

Your parents or the grown up who looks after you have information about the project too. If they are not sure how to answer any of your questions, please let us know and we can tell you more.



If you want to ask us anything about this project please contact Pearl on:

Mobile: 07501 931391 (text or call)
Email: ntxpp9@nottingham.ac.uk

Assent for CYP to take part in an interview (Phase 2)

ASSENT FORM FOR CHILD/YOUNG PERSON
(Final version 2.0 01.12.17)

Title of Study: **Early Phosphate Intervention. An exploration of the perspective of children, parents and key health care professionals towards the development of an early dietary phosphate self-management strategy, to delay disease progression for children with chronic kidney disease stages 1-3, within a managed paediatric renal network: A mixed-methods study**

IRAS Project ID: 215520

Name of Researcher: Pearl Pugh

Name of Parent:

Name of Participant (Child/Young Person):

	Please initial box
1. I have read the project information sheet version number dated and have had the opportunity to ask questions.	☐
2. I understand what taking part in the study involves.	☐
3. I understand that I can change my mind about taking part in the study at any time, without explaining why. This will not affect my medical care at any time in any way.	☐
4. If I choose to change my mind then the information that has been collected so far cannot be removed and may be used in the final report.	☐
5. The person who I talk with will ONLY tell someone else what I say if she thinks I am unsafe or being hurt. Otherwise, everything will be private. I understand what this means.	☐
6. The researcher, Pearl Pugh, can record what I tell her using a digital recorder.	☐
7. I understand that she may use what I say in her final study report.	☐
8. The researcher, Pearl Pugh can take photos of any work that I undertake during the interview.	☐
9. I want to take part in this study.	☐

Letter of invitation for parents and HCPs to participate in the Delphi survey (Phase 3)

Invitation Letter for Parents and Health Care Professionals Delphi Survey
(NOT previously interviewed)
(Final version 1.0 09.10.17)

IRAS Project ID: 215520

Title of Study: An exploration of the perspective of children, parents and key health care professionals towards the development of an early dietary phosphate self-management strategy, to delay disease progression for children with chronic kidney disease stages 1-3, within a managed paediatric nephrology network: A mixed-methods study

Name of Researcher(s): **Pearl Pugh**
Dr Martin Christian
Prof Gina Higginbottom
Dr Pippa Hemingway

Dear Parent/Health Care Professional,

I would like to invite you to get involved in some research that is currently being undertaken by the School of Health Science, at the University of Nottingham. The research is looking at the views of children, parents and health care professionals towards the development of an early dietary phosphate self-management strategy that may delay disease progression for children, during chronic kidney disease (CKD) stage 1-3. The project will be undertaken across the East Midlands, East of England and South Yorkshire (EMEESY) Paediatric Nephrology Network.

You were recommended for participation due to your involvement with the EMEESY Network. Your knowledge, experience, vision and insight is required to inform this study. It will provide you with the opportunity to interact with other parents and health care professionals involved in early CKD management; this may help to improve early self-management for children with CKD, which may help to delay disease progression.

The research team would like you to contribute to an online Delphi survey. A Delphi survey is a structured communication technique where experts in the field answer two or more rounds of questionnaires anonymously, in order to build consensus on the topic. The survey will be conducted over a period of several months. The survey can be completed at a time and place convenient to you, such as home, workplace or a café.

The results of the Delphi survey will be used to write a report, and articles for publication, about the development of an early dietary phosphate self-management strategy for children with CKD 1-3. I have enclosed an information pack about the research and what it involves. However, if you have any questions, please do not hesitate to contact Pearl Pugh, the lead researcher, and she will call

or email you.

If you are interested in your child taking part in this study, please call or email me or Pearl Pugh using the contact details below. We will then call or email you to answer any questions you might have.

I look forward to hearing from you.

Many thanks for your help.

Yours sincerely



Dr Martin Christian
Consultant Paediatric Nephrologist

Email: martin.christian@nuh.nhs.uk

Lead researcher:
Pearl Pugh, Health Sciences PhD student
Email: ntxpp9@nottingham.ac.uk
Study mobile (text or call): 07501 931391

Enc. Participant Information Sheet

Delphi participant information sheet for parents and HCPs (Phase 3)

Delphi Participant Information Sheet for Parents and HCPs
(Final version 2.0: 01.12.17)

IRAS Project ID: 215520

Title of Study: An exploration of the perspective of children, parents and key health care professionals towards the development of an early dietary phosphate self-management strategy, to delay disease progression for children with chronic kidney disease stages 1-3, within a managed paediatric nephrology network: A mixed-methods study

Name of Researcher(s): Pearl Pugh
Professor Gina Higginbottom

We would like to invite you to take part in our research study. Before you decide we would like you to understand why the research is being done and what it would involve for you. One of our team will go through the information sheet with you and answer any questions you have. Talk to others about the study if you wish. Ask us if there is anything that is not clear.

What is the purpose of the study?

The purpose of the research is to seek the views of parents, health care professionals and children regarding the development of an early dietary phosphate self-management strategy for children and young people with early stage (1-3) chronic kidney disease (CKD). Your knowledge, experience, vision and insight are required. We would like to know what you understand about this topic, how it should be delivered, and the potential benefits and challenges of implementing early dietary phosphate self-management in children. The aim of the study is to provide information for staff working with the NHS who are in a position to improve the early dietary phosphate self-management for children with CKD; this may help to delay disease progression.

What do you mean by early dietary phosphate intervention or self-management?

Early dietary phosphate intervention or self-management refers to the provision of dietary phosphate education and support for the child and their family, which may improve the management of chronic kidney disease. This education and support may be delivered at the Queens Medical Centre or at your local hospital.

Why have I been invited?

You are being invited to take part in this research because you are involved with the East Midlands, East of England and South Yorkshire (EMEESY) Paediatric Nephrology Network. As you are involved in providing care for children and young people with CKD 1-3, you will have 'expert' knowledge about managing the disease. Therefore, we would like to know your views, knowledge, experience, vision and insight towards on the development of an early dietary phosphate self-management strategy. We are inviting 20-30 participants like you to take part.

Do I have to take part?

It is up to you to decide whether or not to take part. If you do decide to take part you will be given this information sheet to keep and be asked to sign a consent form. If you decide to take part you are still free to withdraw at any time and without giving a reason. This would not affect your legal rights.

What will happen to me if I take part?

Taking part in this research will involve you completing 2-3 rounds of an online questionnaire, called a Delphi survey. A Delphi survey is a structured group interaction process, where experts in the field answer questions anonymously. After each round, the researcher provides an anonymous summary of the experts' views, to provide a focus for the next round. Thus, experts are encouraged to revise their earlier answers in light of the replies of other participants. The process seeks to achieve consensus on the given topic.

The survey will be emailed to all participants at the same time. It will take 30-45 minutes to complete - depending on the amount of detail you would like to contribute. As the survey will be available on a mobile platform, i.e. mobile devices, it can be completed at a time and place convenient to you, during a 2-3 week period (e.g. your office, home or a public place). The survey will be about your views on the development of an early dietary phosphate self-management strategy for children with CKD 1-3.

Expenses and payments

Participants will not be paid to participate in the study. If participants incur any costs e.g. travel costs, these will be reimbursed.

What are the possible disadvantages and risks of taking part?

Sharing your views and experience of managing CKD 1-3, and discussing early dietary phosphate self-management may bring up issues of a sensitive nature. Participants can be assured that all information shared online during the Delphi survey will be anonymised, therefore others participants will not know who else is taking part. All information shared during the Delphi survey will only be accessible to the other participants and the research team. If a participant feels unhappy to continue their involvement in the Delphi survey, they can withdraw from the study at any time, without giving a reason. The participant will be signposted to the team social worker or the specialist renal nurse for support.

What are the possible benefits of taking part?

By participating in this study, you will have the opportunity to share your insights and express your views on the delivery of phosphate self-management for children with early CKD. We cannot promise the study will improve early dietetic services for children with CKD, but the information we get from this study may help us identify the benefits and challenges of developing an early dietary phosphate self-management strategy, which in turn will be fed back to individuals who manage and provide services for children, and to national policy makers.

What happens when the research study stops?

Throughout the study and once it has stopped the information and demographic data you provide will be kept confidential and will only be seen by the research team. The information collected during the Delphi survey may be used to write reports on service developments in the area of early dietary phosphate self-management. The people who take part in the research will not be named in these reports. The information collected will only be used for the purpose of this study and will not be used for any other project.

Delphi participant information sheet for parents and HCPs (Phase 3) (continued)

What if there is a problem?

If you have a concern about any aspect of this study, you should ask to speak to the researchers who will do their best to answer your questions. The researcher's contact details are given at the end of this information sheet. If you remain unhappy and wish to complain formally, health care professionals can contact Joanne Lymn, Head of School, Faculty of Medicine & Health Sciences, Medical School, Queen's Medical Centre, Nottingham, NG7 2UH. Email:joanne.lymn@nottingham.ac.uk. Parents can contact the Patient Advice and Liaison Service (PALS), NUH NHS Trust, c/o PALS, Freepost NEA 14614, Nottingham NG7 1BR. E-mail: pals@nuh.nhs.uk In the event that something does go wrong and you are harmed during the research and this is due to someone's negligence then you may have grounds for a legal action for compensation against the University of Nottingham but you may have to pay your legal costs. The normal National Health Service complaints mechanisms will still be available to you.

Will my taking part in the study be kept confidential?

We will follow ethical and legal practice and all information about you will be handled in confidence.

If you join the study, the data collected during the study will be looked at by authorised persons from the University of Nottingham who are organising the research. They may also be looked at by authorised people to check that the study is being carried out correctly. All will have a duty of confidentiality to you as a research participant and we will do our best to meet this duty.

All information which is collected about you during the course of the research will be kept **strictly confidential**, stored in a secure and locked office, and on a password protected database. Any information about you which leaves the hospital will have your name and address removed (anonymised) and a unique code will be used so that you cannot be recognised from it.

Your personal data (address, telephone number) will be kept for one year after the end of the study so that we are able to contact you about the findings of the study (unless you advise us that you do not wish to be contacted). All other data (research data) will be kept securely for 7 years. After this time your data will be disposed of securely. During this time all precautions will be taken by all those involved to maintain your confidentiality, only members of the research team will have access to your personal data.

What will happen if I don't want to carry on with the study?

Your participation is voluntary and you are free to withdraw at any time, without giving any reason, and without your legal rights being affected. If you withdraw then the information collected so far cannot be erased and this information may still be used in the project analysis.

Involvement of the General Practitioner/Family doctor (GP)

It will not be necessary for the GP to be informed of your participation in this study.

What will happen to the results of the research study?

The information collected during the Delphi survey will be used to build consensus on the topics which you the 'experts' believe are important in the development of a strategy for early dietary phosphate self-management. The findings of the study will be compiled into a final report and submitted to the University of Nottingham as part of a doctoral research qualification. The findings will also be published in accessible paediatric nephrology and health science journals which will make a contribution to academic theory and practice. The people who take part in the research will not be named in these reports. The information collected will only be used for the purpose of this study and will not be used for any other project.

Who is organising and funding the research?

This research is being organised by the University of Nottingham and is being funded by National Institute for Health Research/Health Education East Midlands

Who has reviewed the study?

All research in the NHS is looked at by an independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed and given a favourable opinion by The Faculty of Medicine and Health Sciences Research Ethics Committee (FMHS REC ref: 17/EM/0418), and The Nottingham 1 Research Ethics Committee.

Further information and contact details

If you would like further information on this research or have any questions before you decide whether you would like to participate, please do not hesitate to contact us using the details below. Please use Pearl as your primary contact point.

The research team

Lead researcher
Pearl Pugh's email:
ntxpp9@nottingham.ac.uk
Pearl's study mobile (text or call):
07501 931391

Researchers
Prof Gina Higginbottom
gina.higginbottom@nottingham.ac.uk
Dr Pippa Hemingway
pippa.hemingway@nottingham.ac.uk
Dr Martin Christian
martin.christian@nuh.nhs.uk

Named representative
Dr Farida Hussain farida.hussain@nuh.nhs.uk



Appendix 13. A section of Delphi round 1 survey



Exploring Parents' and Healthcare Professionals' Views towards the Development of an Early Dietary Phosphate Self-management Strategy for Children and Young People with Early Stage Chronic Kidney Disease

Early Phosphate Intervention study (EPI)

Delphi Consensus Survey
Round 1

Researchers: Pearl Pugh, Professor Gina Higginbottom, Dr Pippa Hemingway and Dr Martin Christian.

We are conducting a survey across the East Midlands, East of England and South Yorkshire Children's Renal Network (also known as the EMEESY Network). The survey seeks to find out the views of parents and healthcare professionals towards the development of a dietary education programme. The programme (known as Early Phosphate Intervention or EPI) aims to teach children and parents about the management of dietary phosphate during the early stages of Chronic Kidney Disease (CKD).

You will be given a list of 48 statements. We would like you to tell us how important you think each statement is when thinking about developing a dietary education programme. You will also be asked to suggest other factors that matter to you. This type of survey is called a Delphi survey and it is used to get everyone's opinion, with the aim of reaching an agreement on the factors that you feel are most important.

Your opinions will inform the preferred future development of an early dietary phosphate education programme for children and young people with early stage CKD.

Before starting Delphi Round 1 I invite you to complete the following consent and demographic form, which will take 2 minutes.

In submitting my response (ticking the box below) I am giving my consent to participate in the study and state that:

1. I confirm that I have read and understand the Delphi Participant Information Sheet (attached with the email) version number 2.0 dated 01.12.17 for the above study and have had the opportunity to ask questions.

2. I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason. I understand that should I withdraw then the information collected so far cannot be erased and that this information may still be used in the project analysis.

3. I understand that relevant sections of my data collected in the study may be looked at by authorised individuals from the University of Nottingham, the research group and regulatory authorities where it is relevant to my taking part in this study. I give permission for these individuals to have access to these records and to collect, store, analyse and publish information obtained from my participation in this study. I understand that my personal details will be kept confidential.

4. I understand that the Delphi survey is anonymous and anonymised direct quotes from the survey may be used in the study reports.

5. I agree to take part in the above study. ☐

Thank you for agreeing to take part in the Early Phosphate Intervention Delphi Survey

Demographic Form

1. Gender

- ☐ Man
- ☐ Woman
- ☐ Prefer not to say

2. Your age group (years)

- ☐ 18-29
- ☐ 30-39
- ☐ 40-49
- ☐ 50-59
- ☐ 60 and over

3. Regarding your involvement with children and young people with CKD, what is your primary role?

- ☐ Parent/carer
- ☐ Paediatric nephrologist
- ☐ Children's nurse
- ☐ Dietitian
- ☐ Social worker
- ☐ Teacher
- ☐ Other

If you selected Other, please specify:

4. If you are a healthcare professional, where do you work? If you are a parent/carer, in which area(s) is the hospital(s) you attend?

- ☐ East Midlands
- ☐ East of England
- ☐ South Yorkshire

5. As a parent/healthcare professional, how many years' experience do you have caring for | or working with children and/or young people with CKD?

- ☐ <1 year
- ☐ 1-3 years
- ☐ 4-6 years
- ☐ 7-10 years
- ☐ > 10 years

6. As a parent/carer or healthcare professional, are you a member of a specialist group? Please tick as many as apply.

- ☐ Parent support group
- ☐ Royal College of Nursing
- ☐ The British Dietetic Association
- ☐ Paediatric Renal Interest Group
- ☐ European Society for Paediatric Nephrology
- ☐ The British Association of Paediatric Nephrology
- ☐ National Union of Teachers
- ☐ British Association of Social Workers
- ☐ Other
- ☐ None of the above

If you selected Other, please specific:

Thank you for completing the consent and demographic form. Please read the following instructions carefully before completing Delphi Round 1 as fully as you can.

Instructions on how to complete Delphi Round 1

The Delphi will comprise up to **three short surveys** (each round will take 15-20 minutes to complete, depending on how much information you would like to contribute), in Round 2 and 3 you will see a summary of your own and the group results.

The aim of the Delphi survey is to reach agreement on what you feel is important in the development of an Early Phosphate Intervention programme for children and young people with early stage CKD. As you complete the survey keep in mind what you would like to see in an Early Phosphate Intervention programme, and what you think needs to be considered to enable it to be put into practice.

We would also welcome written feedback on the wording of the statements provided, in terms of readability, word choice, or anything else you feel is important.

In this Delphi Round 1 we would like you to tell us how important you think each statement is on a scale of 1 to 4. We have called this your 'level of agreement'. These numbers correspond to a response as below:

4	**Very high agreement –** I completely agree that the statement should be used to support the development of an Early Phosphate Intervention
3	**High agreement –** I agree, but the statement requires some ***small*** changes before it should be used to support the development of an Early Phosphate Intervention
2	**Moderate agreement –** I partly agree, but the statement requires ***significant changes*** before it should be used to support the development of an Early Phosphate Intervention
1	**Low agreement –** I feel that this statement ***should not*** to be used to support the development of an Early Phosphate Intervention

Agreement is reached when a statement is rated 'Very high agreement' by 70% of the expert panel.

If you would like to provide a comment or clarify the reason behind your rating, please do so in the box provided at the end of each section. **Providing a reason or comment is optional.**

Please enter a rating for each statement. There are no right or wrong responses. We are interested in your opinion.

There may be factors not listed that you think should be included. If you would like to add any issues or factors, please add them at the end of the survey in the space provided, and you can rate these factors. Each statement is based on:

- a comprehensive review of the research literature; and
- issues that were identified by children and young people, parents and healthcare professionals who were interviewed during an earlier phase of this study.

Your opinions will inform the preferred future in the development of an Early Phosphate Intervention for children and young people with early stage chronic kidney disease.

Delphi round 1 – Statements that will inform the development of an Early Phosphate Intervention

In your opinion, how much do you agree with each statement to inform the development of an Early Phosphate Intervention for children and young people with CKD?

Think about what you would like to see, and what needs to be considered when developing an Early Phosphate Intervention for children and young people with early stage CKD, that will enable it to be put into practice.

Domain	Statement (Select your response to each statement by clicking inside one box)	Very high agreement	High agreement	Moderate agreement	Low agreement
	This first section is an example of how to complete the Delphi survey. First read the statement: *Statement A - 'I think chocolate should be banned from vending machines in any school where there is a child with Chronic Kidney Disease'*				
	If you completely agree with this statement and feel, it is very important in the development of an Early Phosphate Intervention, then click inside the box **'Very high agreement'**. If you feel it is good, but the statement would benefit from some small changes, click **'High agreement'**. If the statement needs significant change then click **'Moderate agreement'**. If you feel the statement is not good and should not be included, click **'Low agreement'**.	☐	☐	☒	☐
	If you would like to leave a comment about a statement, **please include the statement number in the comments box below, followed by your comment.** Move on to the next statement.				
	Please add your comments: **A – the reason I think chocolate should be ...**				

Delphi round 1 - please complete as fully as you can.

Domain	Statement (Select your response to each statement by clicking inside one box)		Very high agreement	High agreement	Moderate agreement	Low agreement
Definition	1	Early phosphate intervention (EPI) is a program of dietary education, engaging with children, young people and their families during the early stages of chronic kidney disease (CKD 2-3), to support diet and lifestyle change towards early dietary phosphate self-management	☐	☐	☐	☐
	2	Early phosphate intervention (EPI) should be labelled 'early intervention healthy eating'	☐	☐	☐	☐
	3	The EPI diet primarily encourages a healthy eating diet, with a reduced intake of processed food, promotion of home-cooked meals containing meat, fish and dairy, and an increase in plant-based foods such as fruit, vegetables, grains and nuts.	☐	☐	☐	☐
	Please add your comments:					
Rationale for the development of an early dietary phosphate intervention.	WHY design an EPI? These statements relate to chronic disease (Select your response to each statement by clicking inside one box)		Very high agreement	High agreement	Moderate agreement	Low agreement
	4	Diet has a crucial role to play in the management of a chronic disease	☐	☐	☐	☐

Space was provided at the end of the survey for additional comments and suggestions.

Are there factors not listed that you think are important and should be included in the development of an EPI? If you would like to add any factors or make any further comments please add them to this table, and rate these factors.

Additional factors	Detail your factors or comments below: (Select your response to each statement by clicking inside one box)	Very high agreement	High agreement	Moderate agreement	Low agreement
		☐	☐	☐	☐
		☐	☐	☐	☐
		☐	☐	☐	☐
		☐	☐	☐	☐
		☐	☐	☐	☐

Appendix 14. Mapping Phase 1 and 2 data against the domains from the TIDieR checklist (2014)

Category	Statement	Phase 1 Systematic Review	Phase 2 Interviews
Definition	Early phosphate intervention (EPI) is a program of dietary education, engaging with children, young people and their families during the early stages of chronic kidney disease (CKD 2-3), to support lifestyle change towards early dietary phosphate SM	√	√
	Early phosphate intervention (EPI) should be labelled 'eaRly intErVention hEALthy-eating' (REVEAL)		
	The EPI program primarily encourages a healthy-eating diet, with a reduced intake of processed food, promotion of home-cooked meals, quality meat, fish and dairy, and an increase in plant-based foods such as fruit, vegetable, grains and nuts.		
Rationale for the development of an early dietary phosphate intervention	Diet has a crucial role to play in the management of a chronic disease	√	
	'Junk foods' and processed foodstuffs are viewed as problem foods to be limited in the management of a chronic disease	√	
	A plant-based rich diet containing vegetables, fruit, grains, pulses, nuts and eggs, meat and fish promotes health, and actively supports the management of a chronic disease diagnosis	√	
	Dietary education can increase confidence in the early management of a chronic disease	√	
	When a child is diagnosed with a chronic disease, a family are more willing to consider dietary/lifestyle changes	√	√
	An excessive intake of dietary phosphate in the early stages of CKD is one of the major triggers for long-term bone and heart problems.		√
	Children and young people with early stage CKD are at risk of consuming large amounts of inorganic (man-made) phosphates due to their addition to		

	processed foods in the form of food additives and preservatives		
	Diet is one of the hardest parts of CKD, introducing the diet early would help to make it the norm for the whole family		√
	You are going to have medicine lifelong; you are going to need to be careful about your diet lifelong.		√
	From diagnosis, parents, children and young people are actively seeking credible information about diet and CKD	√	
	Addressing dietary phosphate early is really tackling the root of the problem		√
	Children and parents need to be told the impact of diet from the early stage, and then they can make an informed choice.		√
	Introducing dietary change early, reduces the dietary burden of care in the later stages of CKD		√
	Dietary phosphate education is a long-term process and should begin in the early stages of CKD		√
	Early dietary advice should be viewed as important as the medical treatment		√
Intended users	Early phosphate intervention should be offered to all children and young people with early stage CKD (2-3) who are likely to progress to the later stages of CKD (4-5)		√
	EPI should be made available across the East Midlands, East of England and South Yorkshire (EMEESY) children's renal network		√
	The EPI program should be directed at the whole family		√
	Mothers are often the main leaders of diet and lifestyle change in a family		√
Delivery of an early dietary phosphate intervention (What/Who/How/Where/When)	The best place to deliver the EPI program is in the children's renal out-patient clinic		√
	The EPI program should be offered at, or close to the diagnosis of CKD		√

	Dietary information/education always carries much more authority when it comes from people who have expertise in nutrition.		
	A children's dietitian (ideally with training in the management of CKD) is the best person to deliver the EPI program	√	
	Written dietary phosphate information is helpful, but it is no substitute for a face-to-face conversation with an expert in children's renal nutrition.		√
	Social media should be better utilised as an educational tool during the early stages of CKD		√
	The EPI program needs to be delivered using a clear, simple, realistic and positive message	√	√
	The EPI program needs to fit within a budget		√
	The EPI program needs to be flexible so that it can be tailored to meet the changing needs of the child or young person and their family		√
	Ongoing feedback and support from healthcare professionals is essential to encourage adherence to dietary/lifestyle change	√	√
	An EPI support group should be set up for parents across the shared-care renal network e.g. online or face to face in the clinic		√
	The healthcare professional should solely direct the delivery of the EPI program		√
	The healthcare professional should work in partnership with the child or young person and their family in order to tailor the EPI program to the changing needs of the individual		√
	The child or young person and their family should direct the delivery of the EPI program		√
	Blood results can act as powerful reinforcements to support adherence to dietary/lifestyle change		√
	Currently available markers of disease progression such as parathyroid hormone (PTH), blood pressure and protein in the urine, should be used to identify		√

	those children with CKD 2-3 who may benefit from receiving the EPI program.		
	Fibroblast growth factor-23 (FGF-23) is a blood test that provides information about phosphate control and how quickly CKD will progress (not currently used in clinical practice)		√
	If a blood test is used in the early stage to inform progress it needs to be 'something that is plain speaking, that directly relates to practice'		√
	In the absence of an early marker of phosphate management, early dietary phosphate education is essential for all children with progressive CKD.		√
Considerations when planning an early dietary phosphate intervention	It is important to build a trusted relationship with a healthcare professional before lifestyle dietary change can take place.		√
	Time is crucial for trust and respect to be built in a relationship		√
	The healthcare professional need time to understand the individual and family needs.		√
	Parents of children with early stage CKD need support from other parents who are in or have been in their situation e.g. support groups, social media		√
	Patient stories and shared experiences are vital; they provide reassurance and make a difference to how a person feels they are able to manage their diet/lifestyle change.		√
	Early dietary advice should be given the same emphasis as the medical treatment		√
Potential concerns	EPI may be of no benefit to the child or young person and their family		√
	It is important to recognise that choosing to follow an EPI diet may be a burden to some families		√
	EPI may create anxiety for the child or young person and their family		√
	EPI may 'label' the child or young person, limiting family expectations of their child or young person		√
	EPI may be intrusive to family life		√

	Due to a lack of cooking skills, the EPI plan may be difficult for some families to follow		√
	The children's (renal) dietitian needs to have more of a presence in the shared-care clinics		√
	More opportunity needs to be made available to get to know the child or young person and their family during the early stages of CKD		√
	Due to infrequent hospital/clinic visits parents and children or young people with early stage CKD often feel isolated		√

Statements shaded in grey were revised or collapsed.

Appendix 15. Delphi results tables

Box 32. Definition S3

Domain:	Statement		Very high agreement	High agreement	Moderate agreement	Low agreement
Definition	3.	The EPI diet primarily encourages a healthy eating diet, with a reduced intake of processed food, promotion of home-cooked meals containing meat, fish and dairy, and an increase in plant-based foods such as fruit, vegetables, grains and nuts.				
How **the panel** responded in round 1			50%	28.6%	17.8%	3.6%
Your round 2 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after round 1.						
Definition	3.	The EPI program will encourage a diet for healthy kidneys, with a reduced intake of processed foods (which are high in added phosphate), parents and carers will be empowered to prepare home-cooked meals containing meat, fish and dairy, with an increase in fruit, vegetables, grains and nuts.	☐	☐	☐	☐

Box 33. Median LOA rating, % LOA and participant group weighting for the 12 rationale statements in round 1

Category rationale: (S4-15)	Median LOA rating (1-4)	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S4	Diet has a crucial role to play in the management of a chronic disease				
	4	89.3 (H93.75/P83.3)	10.7 (H6.25/P16.6)		
S5	'Junk foods' and processed foodstuffs are viewed as problem foods to be limited in the management of a chronic disease				
	3	46.5 (H37.5/P58.3)	35.7 (H43.75/P25)	17.8 (H18.75/H16.6)	
S6	The provision of early dietary education gives children, young people and families a tool to more confidently manage their own diet during the early stages of a chronic disease				
	4	75 (H75/P75)	21.4 (H25/P16.6)	3.6 (H0/H8.3)	
S7	When a child is diagnosed with a chronic disease families are more willing to consider dietary/lifestyle changes				
	3.5	50 (H37.5/P66.6)	28.6 (H31.25/P25)	21.4 (H31.25/H8.3)	
*S8	Diet is one of the hardest parts of CKD, introducing the diet early would help to make it the norm for the whole family				
	4	67.9 (H60/P83.3)	28.6 (H40/P16.6)		
S9	You are going to have medicine lifelong; you are going to need to be careful about your diet lifelong.				
	3.5	50 (H43.75/P58.3)	42.9 (H50/P33.3)	7.1 (H6.25/P8.3)	
S10	Dietary phosphate education is a long term process and should begin in the early stages of CKD				
	4	89.3 (H81.25/P100)	10.7 (H18.74/P0)		
S11	Children and parents need to be told the impact of diet from an early stage, and then they can make an informed choice.				
	4	82.2 (H75/P91.6)	17.8 (H254/P8.3)		
S12	Introducing dietary change early, reduces the dietary 'burden of care' in the later stages of CKD				
	3.5	50 (H37.5/P66.6)	39.3 (H50/P25)	10.7 (H12.5/P8.3)	
S13	From diagnosis, parents, children and young people are actively seeking credible information about diet and CKD				
	4	57.1 (H50/P66.6)	28.6 (H25/P33.3)	14.3 (H25/P0)	
S14	Children and young people with early stage CKD are at risk of eating large amounts of phosphates as these are added to processed foods in the form of food additives and preservatives				
	4	64.3 (H62.5/P66.6)	28.6 (H31.25/P25)	7.1 (H6.25/P8.3)	
S15	Early dietary advice should be viewed as important as the medical treatment				
	4	78.6 (H81.25/P75)	17.9 (H18.75/P16.6)	3.6 (H0/P8.3)	

*missing data for S8 value 3.5 (H6.25%/P 0%)

Participant group (H = HCPs, P = parents) % weighting for each LOA is given in brackets

Box 34. Rationale S7

Domain:	Statement – why design an EPI?		Very high agreement	High agreement	Moderate agreement	Low agreement
Rationale	7.	When a child is diagnosed with a chronic disease families are more willing to consider dietary/lifestyle changes				
How **the panel** responded in round 1			50%	28.6%	21.4%	
Your round 2 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after round 1 into 2 new statements.						
Rationale	7a.	When a child is diagnosed with a chronic disease, knowing and understanding the impact of diet on the disease may motivate families to make diet/lifestyle changes	☐	☐	☐	☐
Rationale	7b.	During the early stages of a chronic disease, diet/lifestyle change can provide a positive focus for the family, knowing that they are making an important contribution to disease management.	☐	☐	☐	☐

Box 35. Rationale S13

Domain:	Statement – why design an EPI?		Very high agreement	High agreement	Moderate agreement	Low agreement
Rationale	13.	From diagnosis, parents, children and young people are actively seeking credible information about diet and CKD				
How the panel responded in round 1			57.1%	28.6%	14.3%	
Your round 2 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after round 1.						
Rationale	13.	From diagnosis, parents, children and young people should have access to helpful and credible information about diet and CKD	☐	☐	☐	☐

Box 36. Rationale S14

Domain:	Statement – why design an EPI?		Very high agreement	High agreement	Moderate agreement	Low agreement
Rationale	14.	Children and young people with early stage CKD are at risk of eating large amounts of phosphates as these are added to processed foods in the form of food additives and preservatives				
How **the panel** responded in round 1			64.3%	28.6%	7.1%	
Your round 2 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after round 1.						
Rationale	14.	Due to the availability of processed foods, children and young people with early stage CKD are at risk of eating large amounts of phosphates as these are added to processed foods in the form of food additives and preservatives.	☐	☐	☐	☐

Box 37. Intended user's S18

Domain:	Statement – who should receive EPI?		Very high agreement	High agreement	Moderate agreement	Low agreement
Intended users	18.	Mothers are often the main leaders of diet and lifestyle change in a family				
How the panel responded in round 1			32.1%	50%	14.3%	3.6%
Your round 2 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after round 1.						
	18.	The EPI program should target the main leaders of diet and lifestyle change in a family	☐	☐	☐	☐

Box 38. Median LOA rating, % LOA and participant group weighting for the 3 intended user's statements in round 1

Category: intended users (S16-18)	Median LOA rating (1-4)	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S16	Early phosphate intervention should be offered to all children and young people with early stage CKD (2-3) who are likely to progress to the later stages of CKD (4-5)				
	4	78.6 (H75/P83.3)	17.9 (H25/P8.3)		3.6 (H0/P8.3)
S17	The EPI program should be directed at the whole family				
	4	78.6 (H75/P83.3)	21.4 (H25/P16.6)		
S18	Mothers are often the main leaders of diet and lifestyle change in a family				
	3	32.1 (H25/P41.6)	50 (H62.5/P33.3)	14.3 (H12.5/P16.6)	3.6 (H0/P8.3)

Box 39. Delivery: where to deliver the EPI, S20

Domain:	Statement – where should we deliver EPI?		Very high agreement	High agreement	Moderate agreement	Low agreement
Delivery	20.	The best place to deliver the EPI program is in the children's renal out-patient clinics, across the East Midlands, East of England and South Yorkshire (EMEESY) children's renal network				
How the panel responded in round 1			53.6%	39.3%	7.1%	
Your round 2 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after Round 1 into 2 new statements.						
Delivery	20a.	The EPI program should be delivered in a location that best suites the family, this may be the home, a shared-care renal clinic close to home, hospital ward or another suitable venue.	☐	☐	☐	☐
Delivery	20b	During the early stages of CKD, parents, children and young people could be invited to attend a group session with other families close to their home, to learn about the EPI program.	☐	☐	☐	☐

Box 40. Delivery: how should the EPI be delivered, S23

Domain:	Statement – how should the EPI be delivered?		Very high agreement	High agreement	Moderate agreement	Low agreement
Delivery	23.	Written dietary phosphate information is helpful, but it is no substitute for a face to face conversation with an expert in children's renal nutrition.				
How **the panel** responded in round 1			64.3%	32.1%	3.5%	
Your round 2 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after round 1 into 2 new statements.						
Delivery	23a	The EPI program should ideally be delivered initially during a face to face consultation with an expert in children's renal nutrition. This would affirm understanding of the advice, and provide an opportunity for the family to ask any questions.	☐	☐	☐	☐
Delivery	23b.	After a face to face conversation with an expert in children's renal nutrition, written dietary phosphate information (including recipe information) is vital, and provides a useful reference that can be frequently accessed at home.	☐	☐	☐	☐

Box 41. Other considerations: support, S34

Domain:	Statement - support		Very high agreement	High agreement	Moderate agreement	Low agreement
Other considerations	34.	Patient stories and shared experiences are vital, they provide reassurance and make a difference to how a person feels they are able to manage their diet/lifestyle change.				
How the panel responded in round 1			64.2%	17.9%	17.9%	
Your round 2 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has remained the same.			☐	☐	☐	☐

Box 42. Other considerations: SM S35

Domain:	Statement – self-management		Very high agreement	High agreement	Moderate agreement	Low agreement
Other considerations	35.	The healthcare professional should solely direct the delivery of the EPI program.				
How the panel responded in round 1			10.7%	28.6%	28.6%	32.1%
Your round 2 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after round 1.						
Other considerations	35.	The healthcare professional should not solely direct the delivery of the EPI program.	☐	☐	☐	☐

Box 43. Other considerations: SM S37

Domain:	Statement - self-management model		Very high agreement	High agreement	Moderate agreement	Low agreement
Other considerations	37.	The child or young person and their family should direct the delivery of the EPI program				
How the panel responded in round 1			14.3%	39.3%	14.3%	32.1%
Your round 2 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after round 1.						
Other considerations	37.	In partnership with the healthcare professional, the parent, child or young person, should direct the delivery of the EPI program	☐	☐	☐	☐

Box 44. Other considerations: early dietary blood test, S41

Domain:	Statement - early dietary blood test		Very high agreement	High agreement	Moderate agreement	Low agreement
Other considerations	41.	If a blood test becomes available for use in the early stage, it needs to be something that clearly tells the healthcare professional how to manage the diet				
How the panel responded in round 1			57.1%	21.4%	14.3%	7.2%
Your round 2 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after round 1.						
Other considerations	41.	If a blood test which gives an understanding of phosphate control, becomes available for use in the early stages of CKD, it needs to be something that clearly informs the healthcare professional and family of the impact of dietary change.	☐	☐	☐	☐

Box 45. Other considerations: potential concerns, S42

Domain:	Statement - potential concerns		Very high agreement	High agreement	Moderate agreement	Low agreement
Other considerations	42.	It is important to recognise that choosing to follow an EPI diet may be a burden to some families				
How the panel responded in round 1			42.9%	39.3%	7.1%	10.7%
Your round 2 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after round 1.						
Other considerations	42.	With the right level of education and support, choosing to follow the EPI diet should not become a burden to the family	☐	☐	☐	☐

Box 46. Other considerations: potential concerns, S45

Domain:	Statement - potential concerns		Very high agreement	High agreement	Moderate agreement	Low agreement
Other considerations	45.	It is important to recognise that following the EPI diet may be intrusive to family life				
How the panel responded in round 1			32.2%	35.7%	21.4%	10.7%
Your round 2 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after round 1.						
Other considerations	45.	Following the EPI program will require the child or young person and their family to adapt to the necessary dietary changes. This may take time, but with support it does not need to be intrusive to family life.	☐	☐	☐	☐

Box 47. Other considerations: potential concerns, S47

Domain:	Statement – potential concerns		Very high agreement	High agreement	Moderate agreement	Low agreement
Other considerations	47.	In order to deliver the EPI diet, children's dietitians needs to have more of a presence in the shared-care clinics				
How the panel responded in round 1			60.7%	28.6%	10.7%	
Your round 2 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has remained the same.			☐	☐	☐	☐

Box 48. Median LOA rating, % LOA and participant group weighting for the intended users statements in round 1 and 2

Category: intended users (S16-18)	Median LOA rating	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S16	4	78.6 (H75/P83.3)	17.9 (H25/P8.3)		3.6 (H0/P8.3)
S17	4	78.6 (H75/P83.3)	21.4 (H25/P16.6)		
S18/R2	4	86.4 (H84.6/P88.8)	4.5 (H7.7/P0)	9.1 (H7.7/P11.1)	

Box 49. Delivery S24 round 1-3

Domain:	Statement 24 – how should the EPI be delivered?		Very high agreement	High agreement	Moderate agreement	Low agreement
Delivery	Round 1.	Social media is not used to its potential as a dietary educational tool during the early stages of CKD				
How **the panel** responded in round 1			28.6%	39.3%	17.9%	14.2%
Delivery	Round 2.	For those that access social media, regular posts on the EMEESY website would provide information to support early dietary phosphate education.	Very high agreement	High agreement	Moderate agreement	Low agreement
How **the panel** responded in round 2			54.5%	31.8%	13.7%	
Your round 3 response (to select your response please click inside one box. Please note you do not have to change your response). Please note this statement has been revised after round 2.						
Delivery	Round 3.	The use of social media (e.g. EMEESY, Twitter or Facebook) in the EPI program would provide users with a useful additional tool and source of information or support.				

Box 50. Median LOA rating and % LOA and participant group weighting for the delivery S24 in round 1-3

Category: delivery (S24)	Median LOA rating	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S24 (how should the EPI be delivered)	3	28.6 (H43.7/P8.3)	39.3 (H37.5/P41.6)	17.9 (H12.5/P25)	14.2 (H6.25/P25)
S24/R2	4	54.5 (H69.3/P33.3)	31.8 (H23/P44.4)	13.6 (H7.7/P22.2)	
S24/R3	4	90.5 (H92.3/P87.5)	9.5 (H7.6/P12.5)		

Box 51. Median LOA rating, % LOA and participant group weighting for the delivery S33b in round 3

Category: other considerations (33)	Median LOA rating	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S33 (support)	4	57.1 (H62.5/P50)	25 (H25/P25)	14.3 (H12.5/P16.6)	3.6 (H0/P8.3)
S33b/R2	4	68.2 (H53.8/P88.8)	27.3 (H38.4/P11.1)	4.5 (H7.7/P0)	
S33b/R3	4	85.7 (H77/P100)	14.3 (H23/P0)		

Box 52. Median LOA rating, % LOA and participant group weighting for the other considerations: potential concerns S42 in round 1-3

Category: other considerations (S42)	Median LOA rating	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S42 (potential concerns)	3	42.9 (H56.25/P25)	39.3 (H31.25/P50)	7.1 (H6.25/P8.3)	10.7 (H6.25/P16.6)
S42/R2	3	45.4 (H31.25/P55.5)	36.4 (H37.5/P22.2)	18.2 (H12.5/P22.2)	
S42/R3	4	71.4 (H61.5/P87.5)	23.8 (H30.7/P12.5)		4.8 (H7.7/P0)

Box 53. Median LOA rating, % LOA and participant group weighting for the other considerations: potential concerns S45a and b in round 1-3

Category: other considerations (45)	Median LOA rating	Very high agreement (4)	High agreement (3)	Moderate agreement (2)	Low agreement (1)
S45 (potential concerns)	3	32.2 (H43.7/P16.6)	35.7 (H37.5/P33.3)	21.4 (H12.5/P33.3)	10.7 (H6.25/P16.6)
S45/R2	4	59.1 (H46.1/P77.7)	27.3 (H38.4/P11.1)	13.6 (H15.3/P11.1)	
S45a/R3	4	85.7 (H84.6/P87.5)	14.3 (H15.3/P12.5)		
S45b/R3	4	71.4 (H77/P62.5)	14.3 (H15.3/P12.5)	9.5 (H0/P25)	4.8 (H7.7/P0)

Appendix 16. Cross tabulation of frequencies for Cohen's Kappa

S8R1 and S8R2

S8R1	S8R2*				
		Low Agreement	Moderate Agreement	High Agreement	Very high agreement	
	Low agreement	0	0	0	0	0
	Moderate agreement	0	0	0	0	0
	High agreement	0	0	5	2	7
	Very high agreement	0	0	0	14	14
		0	0	5	16	21

*N=1 missing data

S12R1 and S12R2

S12R1	S12R2					
		Low agreement	Moderate agreement	High agreement	Very high agreement	
	Low agreement	0	0	0	0	0
	Moderate agreement	0	0	1	0	1
	High agreement	0	0	6	3	9
	Very high agreement	0	0	0	12	12
		0	0	7	15	22

S34R1 and S34R2

S34R1	S34R2				
		Low agreement	Moderate agreement	High agreement	Very high agreement	
	Low agreement	0	0	0	0	0
	Moderate agreement	0	3	0	0	3
	High agreement	0	0	2	3	5
	Very high agreement	0	0	0	14	14
		0	3	2	17	22

S40R1 and S40R2

S40R1	S402R2				
		Low agreement	Moderate agreement	High agreement	Very high agreement	
	Low agreement	0	0	1	0	1
	Moderate agreement	0	1	0	2	3
	High agreement	0	0	0	5	5
	Very high agreement	0	0	0	13	13
		0	1	1	20	22

S47R1 and S47R2

S47R1	S472R2				
		Low agreement	Moderate agreement	High agreement	Very high agreement	
	Low agreement	0	0	0	0	0
	Moderate agreement	0	1	0	1	2
	High agreement	0	0	3	3	6
	Very high agreement	1	0	0	13	14
		1	1	3	17	22

Appendix 17. Letter of HRA Approval

NHS
Health Research Authority

Professor Gina Higginbottom
The Mary Seacole Professor of Ethnicity and Community Health
University of Nottingham
Faculty of Medicine & Health Sciences
University of Nottingham
Derby Road
NG7 2UH

Email: hra.approval@nhs.net

17 January 2018

Dear Professor Higginbottom

Letter of HRA Approval

Study title:	**Early Phosphate Intervention Mixed Methods Study: An exploration of the perspective of children, parents and key health care professionals towards the development of an early dietary phosphate self-management strategy to delay disease progression in children with Chronic Kidney Disease stage 1-3, within a managed paediatric renal network**
IRAS project ID:	**215520**
Protocol number:	**17090**
REC reference:	**17/EM/0418**
Sponsor	**University of Nottingham**

I am pleased to confirm that HRA Approval has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications noted in this letter.

Participation of NHS Organisations in England

The sponsor should now provide a copy of this letter to all participating NHS organisations in England.

Appendix B provides important information for sponsors and participating NHS organisations in England for arranging and confirming capacity and capability. **Please read *Appendix B* carefully**, in particular the following sections:

- *Participating NHS organisations in England* – this clarifies the types of participating organisations in the study and whether or not all organisations will be undertaking the same activities
- *Confirmation of capacity and capability* - this confirms whether or not each type of participating NHS organisation in England is expected to give formal confirmation of capacity and capability. Where formal confirmation is not expected, the section also provides details on the time limit given to participating organisations to opt out of the study, or request additional time, before their participation is assumed.