Decision making and future planning for children with life-limiting conditions: a longitudinal case study.

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Abstract

Background

The number of children with life-limiting conditions in England is increasing. Families and professionals caring for such children face difficult care and treatment decisions. However, little is known about the factors which influence such decision making and the processes through which decisions are made.

Aim

To explore the process of decision making and future planning for children with life-limiting conditions.

Methods

This study employed a longitudinal, qualitative case study approach, involving eleven cases. Cases were centred around a child and their family, and additional interview participants, including healthcare professionals and extended family members, were nominated by the family to participate in interviews. Data sources included 46 interviews, 72 observations and review of medical notes. Data were analysed using narrative analysis.

Findings

Key findings pertained to the importance of relationships and communication, the nature of decision making and strategies used within the process, and the role of best interests' discussions during disagreements. Good relationships and communication were essential in decision making, as the process often involved multiple stakeholders. Poor inter-professional communication however was common and presented challenges for decision making. Analysis demonstrated that individuals recognise little practical or emotional difference between decision making and future planning, suggesting that they conceptualise both, simply as decision making. Within the process of decision making, numerous strategies were identified which aimed to achieve agreement between stakeholders. These were information framing, persuasion, appeals to evidence and justification. During protracted disagreements, occasionally reference to the child's 'best interests' was made and occurred when professionals were concerned about harm to the child. Consequently the possible limits to parental

decisional involvement through application to the courts, was highlighted, yet never actually implemented. Predominantly collaboration, which was indicative of shared decision making, was observed and reported.

Conclusion

This study revealed the complex and nuanced nature of decision making. This research has highlighted the necessity of good quality relationships between families and professionals, and also communication in the inter-professional relationship. For those involved in decision making for children with life-limiting conditions, an understanding of each individual's holistic wishes and values, alongside their perception of decisional risks to the child are important considerations, particularly during sustained disagreements. This study suggests that despite the policy drive towards shared decision making and the widespread perception that parents are the ultimate decision makers for their children, these assumptions are bounded. It calls for a professional awareness of the moral work undertaken by families in decision making, in order to further promote collaboration and prevent sustained disagreement.

Scholarly outputs from this thesis

Oral papers:

Popejoy, E. Anticipated challenges in researching children's palliative care. Palliative Care Research Society and University of Nottingham PhD Day. 8th December 2015, Nottingham, UK.

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Popejoy, E., Johnston, B., Manning, J Almack, K. and Pollock, K. Exploring communication practices within the healthcare setting: "There's got to be a better system". Defining moments in children's palliative care: Transforming the family experience, 10-11 May 2017. Birmingham, UK.

Poster presentations:

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Popejoy, E., Johnston, B., Manning, J Almack, K. and Pollock, K. Decision making for children with life-limiting illnesses: the different guises of 'convincing' and 'compromising'. Nottingham Paediatric Research Showcase. 14th June 2017, Nottingham, UK.

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List of abbreviations and glossary of terms

ACP	Advance Care Planning.
Case Study	Case study is an approach to research which aims to understand the specifics and complexity of a phenomena within each individual case (or unit of analysis).
CPAP	Continuous Positive Airway Pressure – this is a method of ventilation in which a positive air pressure is delivered, aiming to keep the airway open and ensure adequate gas exchange.
FCC	Family Centred Care.
Fundoplication	This is a surgical procedure which makes the lower oesophageal sphincter (the opening between the oesophagus and stomach) tighter and helps to reduce or prevent gastro-oesophageal reflux and vomiting.
Grommets	These are tiny tubes that are inserted into the child's eardrum when they have a build-up of fluid in the middle ear, which enables the drainage of this fluid away.
HRA	Health Research Authority.
Jejunal feeding	This can be via a naso-jejunal tube which is inserted through the nostril, into the stomach and through into the jejunum, or directly through the abdomen into the jejunum, this is often referred to as a PEG-J. It is used when the child cannot tolerate feeding into their stomach and experiences frequent vomiting or delayed gastric emptying.
Network of Care	This term refers to the stakeholders involved in each case.
NGT	Nasogastric Tube - this is a tube which is passed through the nose, down the oesophagus and into the stomach. It is secured on the face and may be used during an acute illness whilst the child is temporarily unable to feed

	orally, or as a long term solution until a PEG can be inserted surgically.
NHS	National Health Service.
NICU	Neonatal Intensive Care Unit.
NIV	Non-Invasive Ventilation – this is a device which provides breathing support non-invasively and ensures that there is adequate ventilation.
NPA	Naso-Pharyngeal Airway – this is a tube which is inserted into the nostril and reaches the pharyngeal space. It keeps the airway open and is often used for children who have partially obstructed airways, as a way of maintaining airway patency.
PEG	Percutaneous Endoscopic Gastrostomy (often referred to simply as a gastrostomy) - this is a feeding tube which is passed directly through the wall of the abdomen and into the stomach. It is permanent in so far as it can remain in-situ forever, but can be removed (and the hole will close over) if it is no longer needed.
PICU	Paediatric Intensive Care Unit.
Portacath	This is a device which is inserted surgically and provides reliable intravenous access, reducing the need for cannulas and needles to take blood samples.
QoL	Quality of Life.
SDM	Shared Decision Making.
Stakeholder	This term refers to those who have some degree of influence in the child's care and decision making.
TIPS	Trans jugular intrahepatic portosystemic shunt (TIPS) - a procedure that may be used to reduce portal hypertension and its complications, especially variceal bleeding.

Tracheostomy	This is a surgically created airway in which a hole is created through the front of the neck into the trachea and through which a tube is inserted and secured. This assists with breathing in people with anatomical abnormalities or neurological issues meaning that they are unable to protect their own airway.

"Caring for children and young people with life limiting conditions is an embryonic field. Now is the time to remove the barriers, seize the opportunities and move forward with research which will benefit children and families."

(Bluebond-Langner et al., 2017)

Chapter 1. Introduction and Background

1.1. Introduction

The number of children with life-limiting conditions in England is increasing (Fraser et al., 2012). The fluctuating nature of their illnesses mean that they frequently access health services and those caring for them are faced with difficult decisions. Highly publicised cases allude to the possible disagreements between stakeholders and therefore the challenges which exist in making ethically difficult decisions for children with life-limiting conditions. Such cases highlight the disparate viewpoints which can exist within a single case, and indeed within wider society. My experience of nursing children with life-limiting conditions, has enabled me to see first-hand the difficulty which at times exists in decision making for these children. This experience and the lack of research in this field prompted this investigation into decision making for children with life-limiting conditions.

This thesis presents an empirical investigation of the process of decision making for children with life-limiting conditions. The study is novel as a result of its longitudinal design, case study approach and specific focus on children with life-limiting conditions. It explores the network of care, outlining the various individuals involved in decision making, their preferences for involvement, and the communication and negotiation within such networks of care.

This chapter will situate this study in relation to the prevalence of such conditions, relevant national policies and the ethical and legal context of the UK. A discussion of decision making theories and models will follow and the chapter will conclude with an outline of the structure of this thesis.

1.2. Significance of the Field of Study

It is estimated that worldwide the number of children living with life-limiting and life-threatening conditions is at approximately 1.2 million (Worldwide Palliative Care Alliance, 2014). In England, this number is approximately 40,000 and has been shown to be steadily increasing (Fraser et al., 2012), which has, at least partially, resulted from the decreasing mortality in this population (Norman and Fraser, 2014).

"Life-limiting conditions are those for which there is no reasonable hope of cure and from which, ultimately, the child will die. Life-threatening conditions are those for which curative treatment may be feasible but can fail" (Together for Short Lives, 2014, p.1). Figure 1 outlines the four widely recognised categories of life-limiting and life-threatening conditions, as described by ACT and RCPCH (1997).

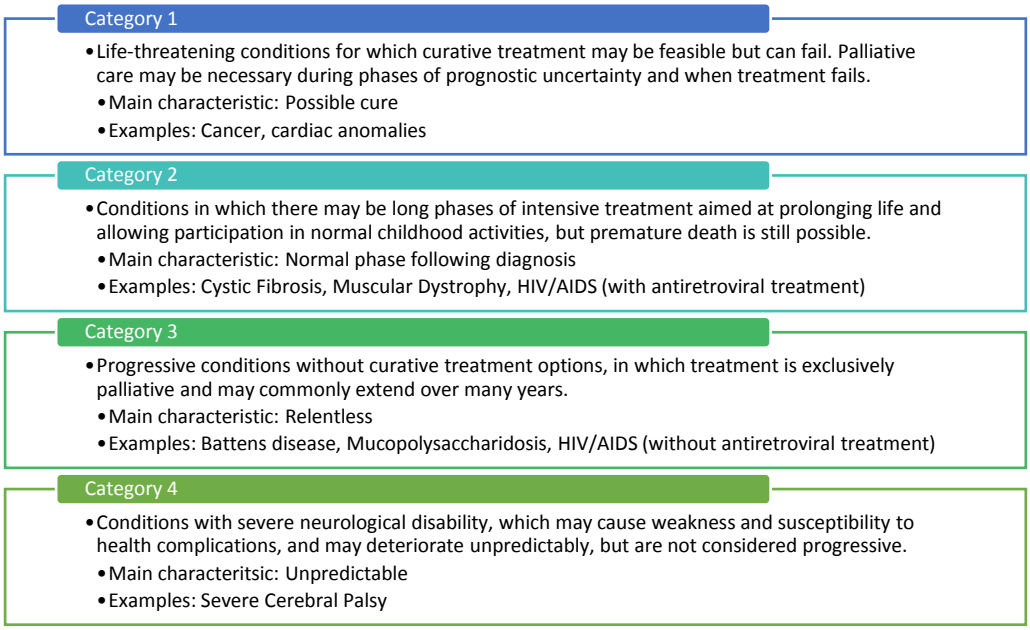


Figure 1: Categories of life-limiting conditions

Category 1 represents life-threatening conditions and categories 2-4 represent life-limiting conditions (Together for Short Lives, 2014). These categories are well accepted and used extensively by the national charity for children with life-limiting and life-threatening conditions, 'Together for Short Lives' (Widdas et al., 2013, Together for Short Lives, 2015c).

The World Health Organisation Global Atlas of Palliative Care identifies that globally, progressive non-malignant disease represents the highest proportion, accounting for between 78-91% of children requiring palliative care (World Health Organisation and World Palliative Care Alliance, 2014). Children with cancer represent less than 15% of the global population of children with life-limiting and life-threatening conditions (World Health Organisation and World Palliative Care Alliance, 2014). Similar figures have been reported in the UK, with 13.7% of children with life-limiting conditions having an oncological

diagnosis, and congenital anomalies being the most prevalent (Fraser et al., 2012). This highlights the need to better understand the experiences of children with progressive non-malignant diseases, as they represent the majority of children with life-limiting conditions.

Children with life-limiting and life-threatening conditions have fluctuating disease stages, transitioning frequently between stable, unstable and deteriorating, and finally they will progress to the dying stage. A retrospective five year study showed that in Scotland, between 14-19% of the children with life-limiting conditions enter the stages of unstable, deteriorating or dying[1] each year (Jarvis et al., 2017) during which many decisions are likely to be made. Given the potential instability of children with life-limiting conditions, they may require admission to the paediatric intensive care unit (PICU) for treatment. Fraser and Parslow (2017) found that the majority of admissions to PICU are for children with life-limiting conditions and that these children have significantly longer lengths of stay than children without life-limiting conditions. Children with life-limiting conditions were also significantly more likely to die in the PICU than children without a life-limiting condition (Fraser and Parslow, 2017). This data demonstrates the complexity of the care for children with life-limiting conditions and the likelihood of admission to hospital and/or the PICU. During such periods of instability or critical illness, many different decisions may need to be considered, such as artificial feeding, change to or commencement of medications, and withholding or withdrawing life-sustaining treatment. However, little is known about the specifics of the interactions within which options are communicated and discussed, and decisions negotiated.

Despite malignant diseases affecting only a small proportion of children receiving palliative care, this area has received the greatest attention from researchers (Price et al., 2012). Wood et al. (2010) recognised that different disease milestones are present in each category in varying degrees. For example, gastrostomies were minimally significant for families of children with Category 1 conditions, but much more significant in the other three categories. Similarly Fraser et al. (2014) recognised that differences exist between the needs of

[1] Unstable was defined as an unplanned admission to hospital lasting more than 48 hours.

children and young people with Category 1 conditions and those with progressive conditions. These findings demonstrate that each category is likely to pose different challenges for families and professionals in relation to treatments and decision making. The lack of research into palliative care for children with non-malignant disease demonstrates the need for further research within this population.

1.3. National context

1.3.1 Policy drivers

Guidelines suggest that children with life-limiting and life-threatening conditions should be offered palliative care from diagnosis and that this should extend to their death, and beyond for families (EAPC Task Force, 2007, Together for Short Lives, 2015b). It is also recognised that children's palliative care may be delivered over many years (Together for Short Lives, 2015a). The importance of ensuring equitable access to palliative care has been recognised through the Access to Palliative Care Bill (2015) and national documents have highlighted the need to improve the quality and provision of children's palliative care services (Craft and Killen, 2007, HM Treasury and Department for Education and Skills, 2007, Department of Health, 2008).

The Independent Review of Palliative Care Services for Children and Young People identified that palliative care should be holistic and family centred, aim to enhance quality-of-life, and provide symptom management, respite and bereavement care (Craft and Killen, 2007). Such goals of children's palliative care are echoed by other relevant organisations (Together for Short Lives, 2013b, EAPC Taskforce for Palliative Care in children, 2009). Families of children accessing palliative care should also be offered the opportunity to participate in decision making and planning for their child's future care (Department of Health, 2008, Together for Short Lives, 2013b, East Midlands Children's and Young People's Palliative Care Network, 2011).

Recent national guidance on end of life care for infants, children and young people with life-limiting conditions recommends that professionals involve families in care decisions and work to identify their preferences for future care (National Institute for Health and Care Excellence, 2016). Subsequently quality

standards have been released which identify advance care planning as a priority area for improvement (National Institute for Health and Care Excellence, 2017).

Despite the national policy commitment to improving children's palliative care and ensuring family participation in decision making, most of the existing research has focused on children with cancer (Price et al., 2012), despite the majority of children accessing palliative care having non-malignant conditions. Research more generally into children's palliative care is sparse, as it is still a relatively young discipline (Steele et al., 2008, Knapp, 2009). There exists, therefore, a knowledge deficit regarding how best to improve children's palliative care through involving families in care decisions and this deficit needs to be addressed.

1.3.2. Moral, ethical and legal dilemmas

Controversies in relation to decision making for children with life-limiting conditions have, over the previous two decades, been increasingly discussed within the mainstream and social media. The ease of access to support groups and medical information mean that *"families are much more likely than previously to find themselves at variance with their medical advisers when their assessment of treatment benefits differ from those of the medical team"* (Elias-Jones and Samanta, 2005, p.824).

High profile media cases demonstrate that protracted and significant disagreements between parents and professionals can and do occur, requiring escalation to the courts for resolution. In situations of significant disagreement, professionals must assess whether the parental opinion is in accordance with the child's best interests and if not, they have a professional duty (General Medical Council, 2007, Royal College of Nursing, 2003) to intervene and make an application to the court. The court then becomes the legal arbitrator of what is in the child's best interests (Children Act, 1989).

Despite the widely accepted concept of best interests in the medical and legal field, it has been argued within the bioethical literature to be indeterminate and subjective (Diekema, 2004, Alessandri, 2011, Salter, 2012, Shah et al., 2017, Birchley, 2016b). As outlined by Wilkinson and Savulescu (2017a), when deciding which treatments it is acceptable to offer a child, professionals need to

assess whether a treatment is in the child's best interests and they should not offer it if they do not believe it represents the child's best interests. However, this process takes place from the perspective of the professional and therefore any treatment offers provided will be the ones which the professional believes represent the child's best interests and therefore parents may be unaware of other possible options. Given the subjective nature of the concept of best interests, some treatments, which the parents may perceive to be in the child's best interests, might not be offered. This demonstrates the significant control which professionals have in making best interests decisions and how, despite parents as key players in assessing their child's best interests (Salter, 2012), they may at times be absent from such assessments.

Unresolved disputes necessitating applications to the courts are rare (Wilkinson, 2017), however minor conflict has been shown to be prevalent across paediatric specialities. Reasons include communication breakdown, disagreement about treatment and differing expectations (Forbat et al., 2016). When minor conflicts are not resolved, and escalate to the need to involve the courts, there may be significant implications for trust and ongoing relationships (Forbat et al., 2015).

The media attention surrounding cases of unresolved conflict, most notably the case of Charlie Gard, demonstrated the difficulty which can sometimes occur when making decisions for children with life-limiting conditions. This difficulty results from the often disparate moral viewpoints held by the individuals relevant to the decision (Carnevale, 2005) and the subsequent varied assessments of what constitutes the child's best interests. This consideration highlights the need for further research investigating how decisions for children are made in the context of numerous people with varying degrees of decisional responsibility and how conflicts are avoided (or not) in practice.

1.4. Decision making

An introduction to the available decision making theories and models will be useful in order to understand the existing literature and the findings from this study. The following section considers such theories and models, and their relevance to decision making for children with life-limiting conditions.

1.4.1. Theories of decision making

Traditional theories of decision making evolved from the field of business where decision makers were portrayed as devoid of context and emotion (Simon, 1956, Becker, 1962). These theories are no longer recognised as applicable within the medical setting where decisions carry risk and uncertainty, and emotions influence the decisions made. It is now widely recognised that decision making is not a purely rational process, rather an individuals' rationality is constrained by their access to information and their environment (Simon, 1955). 'Dual-process' theories acknowledge two systems of decision making (Figure 2).

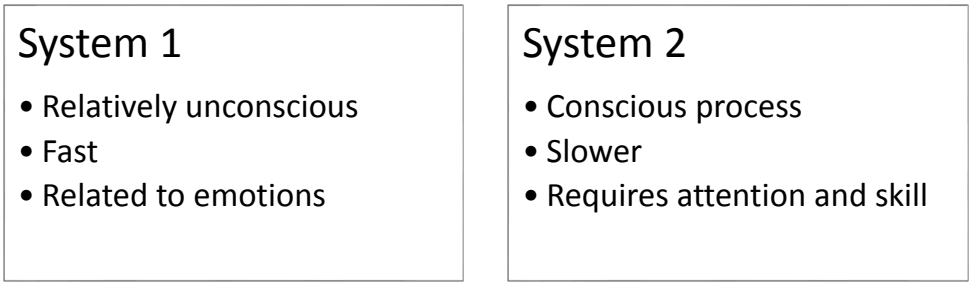


Figure 2: Systems of decision making

Dual process models propose that people make decisions under the 'principle of least effort', that is, once a decision considered good enough has been found, the individual chooses that course of action and ceases to search for alternatives (Chen and Chaiken, 1999). The heuristic, System 1 processes act first and are therefore often the default mode for decision making, especially when resources or motivation are limited (Shiv and Fedorikhin, 1999, Payne and Bettman, 2004). If System 1 fails to make a good enough decision, System 2 processing will be used to find an acceptable alternative course of action (Chen and Chaiken, 1999) or enlisted to confirm a decision. However, dual process models only refer to individual decision making and therefore do not consider how multiple individuals negotiate complex decisions.

Prospect theory has also been developed to explain how people make choices under conditions of risk and uncertainty. It posits that people make a mental model of the decision under consideration, in which they subconsciously frame the problem differently using information about the problem and their own individual context (Beresford and Sloper, 2008) and recognises the heuristic processes involved (Tversky and Kahneman, 1992). Additionally prospect

theory posits that each option presents a 'better' choice with respect to a particular attribute and therefore individuals may sometimes be reluctant to make choices, for fear of experiencing a degree of loss. Loss is experienced in relation to the 'better' attribute of the option that they did not choose, however whilst the decision is in process, they do not experience this loss (Beresford and Sloper, 2008). Prospect theory recognises that different individuals involved in the same decision may frame the decision differently and therefore exploring the mental models that people construct when making decisions will be important in understanding the choice that they make.

1.4.2. Clinical Decision Making

Many authors have conducted concept analyses of decision making in relation to clinical decision making and health care, but these have predominantly focused on decision making by an individual. Noone (2002) conducted a concept analysis of client decision making in healthcare, identifying that the process begins with recognition of a stimulus and results in a commitment to a particular course of action. Concept analyses from a healthcare professional perspective have also been conducted with the specific foci on clinical reasoning (Simmons, 2010) and decision making in nursing practice (Johansen and O'Brien, 2016). These concept analyses acknowledged that decision making and clinical reasoning are greatly complex endeavours, and that contextual factors such as values and stress (Johansen and O'Brien, 2016, Simmons, 2010) have an impact on the process. All these concept analyses identified the consequences of the concept as the decision made and suggested that the consequences of one decision may become the antecedents of the next decision, *"making it cyclical and recursive"* (Simmons, 2010, p.1155). These models represent decision making as a process carried out internally by one stakeholder and resulting in an action, rather than made through a process of interaction and discussion with others. Decision making in the setting of child health however, involves multiple individuals, such as parents, professionals and the child and therefore these models are unlikely to be directly applicable to this type of decision making.

1.4.3. Shared Decision Making

The Shared Decision Making (SDM) approach within health care has been gaining prominence since the late 1990's, when it was recognised that patient

involvement in their care and treatment was necessary within a health care system with finite resources (Da Silva, 2012). In recent years SDM has been promoted in UK health care policy. This approach of patient participation is outlined in 'Equity and Excellence: Liberating the NHS' (Department of Health, 2010) in which a commitment to SDM is made through the slogan *"no decision about me, without me"* (p.3). The implementation of SDM in clinical practice has been identified as challenging, with identified barriers including lack of time, lack of training, perceived knowledge of the patient and attitudes of professionals (Da Silva, 2012). 'Equity and Excellence: Liberating the NHS' (Department of Health, 2010) states that when reference is made to involving patients in decisions, this applies to children and their families also, demonstrating a commitment to SDM within paediatrics.

The concepts of participation and collaboration in care, alongside family-centred care (FCC)[2], have also grown in prominence over the past two decades to account for the multiple perspectives involved in the care of children. These concepts were developed as a way of promoting family involvement in the provision of care whilst children were hospitalised (Coyne, 1996, Hutchfield, 1999). Despite SDM and FCC being promoted in recent years, the challenges specific to practically implementing them in relation to child health have thus far not been addressed.

Recently Park and Cho (2017) conducted a concept analysis of SDM in paediatrics and identified its attributes as: active participation of parents, children and the health professional, collaborative partnership, compromise, and shared goals for the child's health. Similar attributes were identified in relation to FCC (Smith et al., 2015b). The literature has identified that FCC is not implemented in accordance with the attributes upon which it is based (Paliadelis et al., 2005, Coyne and Cowley, 2007, McKean et al., 2012). Similarly some literature suggests that the implementation of SDM is not commensurate with its defining attributes (Lipstein et al., 2014, Brinkman et al., 2011) and is variable according to the child's condition, with those with developmental and

[2] The concepts of participation, collaboration and family centred care are often used interchangeably and have similar attributes. For brevity, these concepts will be discussed together and simply referred to family centred care.

behavioural disorders experiencing less SDM than those with physical conditions (Smalley et al., 2014, Lipstein et al., 2016). However, such claims are usually followed by suggestions for how this situation could be improved, rather than critiquing the applicability of SDM in this field. Given the nature as parents and professionals as proxy decision makers (Wyatt et al., 2015, Park and Cho, 2017) in the field of child health, and the ultimate role of the court as the final arbitrator of a child's best interests (Children Act, 1989), there are limits to SDM in this field. These limits have thus far not been appropriately explored within health care policy or the literature.

1.4.4. Relational Models of Decision Making

With the increased attention to patient choice and policy drivers to practice SDM (Department of Health, 2010), relational models of decision making have been created. Hinds and Kelly (2010) developed a relational model of decision making for children with cancer which recognised the roles of the child, parents and professionals and a number of factors, such as, parental trust in professionals and information provision, in decision making. However, despite acknowledgement of some relational features of the process of decision making, the resultant model was incredibly simplified. It was developed through reference to research from the parental perspective and published recommendations, and therefore may have failed to fully account for the specific factors which influenced the professionals' decisions. This model overlooks the role of multiple professionals in decision making and the integration of their potentially different perspectives in the process of decision making. Nicholson (2012) also developed a model of decision making from her research in children's palliative care, which was much more comprehensive and involved a consideration of how system factors can impact on decision making. Despite its increased relevance, this model was also based on research from the parent perspective only and is therefore likely to have failed to fully account for the relational issues in decision making, which involve multiple professionals, each with their own influences and beliefs.

Most recently Carnevale et al. (2017) has proposed a relational ethics framework for children with complex health care needs. This framework is underpinned by the understanding that when making decisions about the child's care, the parent

has a moral obligation to consider the impact on the wider family and themselves, rather than just the child and that this poses moral tensions for them. This framework recognises the potential moral conflicts which may be present when parents make decisions for their child. It proposes that professionals should strive to understand, and facilitate understanding within the child's family and wider network, of the moral experiences of all those involved in the decision, including themselves. These relational models of decision making are useful in order to understand decision making in the complex relational contexts of children with life-limiting conditions.

1.5. Personal reflections and impetus for the study

I embarked upon this study as a Staff Nurse with 5 years clinical experience and had cared for a number of families of children with life-limiting conditions. One particular family of a boy with a degenerative neurological condition stands out in my mind as the first time I considered the process of decision making in this population. I had known the family for many years since I was a student nurse and during one admission I watched as the child deteriorated and the family struggled to make decisions regarding his care and treatment. This child had had numerous previous admissions which all followed the same pattern of decision making, where the child's Emergency Care Plan (which stated what the ceilings of care should be and had been agreed to by his mother) was put aside at her request and the treatment necessary to keep the child alive was provided. This particular admission signalled the final time that this pattern occurred, as the child's mother realised that he had become so uncomfortable and decided to take him to the hospice, where he died a few days later. Following this incident, I decided to search for the available literature on decision making for children with life-limiting conditions and realised that this was sparse. Having observed the anguish experienced by this child's family in decision making about his care, the identified research deficit in this area and my desire to undertake further academic study, I embarked on this PhD.

1.6. Thesis structure

The structure of this thesis is briefly outlined below, in order to guide the reader in relation to its contents.

Chapter 2 presents a thorough review of the literature and is formed of two sections. The first section provides a narrative review of the literature pertaining to paediatric chronic illness and palliative care, in order to illuminate aspects of decision making more broadly within paediatric long term conditions. The second section provides a systematic review and thematic synthesis of the literature specific to decision making for children with life-limiting conditions. The chapter concludes with a consideration of the gaps in the literature.

Chapter 3 presents the aim and objectives of this empirical study. It provides an account of the philosophical and methodological assumptions which underpin this research, followed by the specifics of the study design, data collection methods and study procedures. The chapter then discusses the relevant ethical considerations of this study, details the process of data analysis employed and finally considers issues related to the research credibility.

Chapter 4 provides an introduction to the cases and seeks to acquaint the reader with the participants involved in data collection, the specifics of each case and the types of decisions under consideration. A timeline is provided for each case in order to contextualise the study period within the lives of the families and detail how the relevant decisions for them occurred over time.

Chapters 5 provides a brief introduction to the findings chapters and how these chapters inter-relate. It then discusses the communication required for decision making and the relationships which it takes place within. It highlights the importance of good communication in the development of relationships and the concerns which arise when communication is sub-optimal. It then provides an account of the presence of trust as an important feature of decision making and the need to seek second opinions at times when it does not exist.

Chapter 6 presents the findings which illuminate the process of decision making. It details different categories of decisions in relation to their characteristics and the significant moral accountability felt by those involved in decision making. The chapter then outlines the relational components of decision making through consideration of how options are communicated and the strategies which are employed in the quest for agreement between all stakeholders.

Chapter 7 presents the findings in relation to protracted disagreements and the resultant appeal to the child's best interests when negotiating a decision. It outlines the differences in different stakeholders' perceptions regarding best interests and who is best placed to assess best interests'. It then considers the implications for relationships when the concept is invoked with reference to its associated legal connotations. Finally it provides an understanding of professional justification, which is a key component of the moral work undertaken when best interests are appealed to.

Chapter 8 presents a discussion of the study findings and the methodological insights from this study. It outlines the novel contribution which this study has made and presents a conceptual framework of decision making for children with life-limiting conditions. The chapter then discusses these findings in relation to the wider literature. It considers my own impact on the research, through a reflexive account and then outlines the strengths and limitations of the research, and the implications for research and practice.

Chapter 9 provides a conclusion for this thesis, summarising the findings and the overall contribution that this thesis makes to our understandings of decision making for children with life-limiting conditions.

1.7. Conclusion

This chapter has oriented the reader to this thesis. It has provided an introduction to the population of children with life-limiting conditions, outlining the significance of the current study, an overview of decision making theory, my own personal motivations for undertaking this research and an outline of the thesis structure.

Chapter 2. Literature Review

2.1. Introduction

This chapter aims to provide a better understanding of the process of treatment decision making for children with life-limiting conditions. It will explore the decision making literature within the field of children's chronic illness and palliative care[3], in order to grasp a wider understanding of the factors and individuals involved in the process of decision making more generally. The findings of a systematic review and thematic synthesis of qualitative literature specific to decision making for life-limited children will then be presented. Finally the chapter will culminate in the identification of gaps in the literature and the justification for this empirical research.

2.2. Decision making in paediatric chronic illness and palliative care

Decision making research within paediatric palliative care has been primarily conducted in the field of paediatric oncology (Price et al., 2012). Although this, and the research within the context of paediatric chronic illness, may not be directly applicable to children with life-limiting conditions, it may highlight additional factors worthy of consideration and help in developing an understanding of the factors involved in the process, and the appropriateness and acceptability of various methods of study. The types of decisions that are relevant to children within these contexts include: parental decisions around termination of the pregnancy, commencement of medications, artificial nutrition and hydration, oxygen therapy, non-invasive ventilation, tracheostomy placement, surgical intervention, research participation, acceptance of respite care and end of life care (Toly et al., 2012, Allen, 2014, Ling et al., 2016, Beecham et al., 2016). Those caring for the child may be faced with many of these decisions over their life and many factors may influence the decisions made. Such factors will now be discussed.

[3] Some reference to ethical and wider healthcare literature is made in a few places where it provides relevant additional understanding of the process of decision making.

2.2.1. Family factors

Perception of the evidence

Situational awareness is the primary antecedent of decision making as outlined in the aforementioned concept analyses (Noone, 2002, Johansen and O'Brien, 2016, Simmons, 2010). Therefore some awareness of the need for action is required for decision making and the results of clinical tests may often provide this awareness. However, previous research has shown that parents may interpret these results differently from professionals, resulting in a situation where they lack the awareness of the need for action (Craig, 2004, Nicholson, 2012, Thorne et al., 1997). Such situations can result in data conflict, where individuals have different interpretations of the data or information provided to them (Moore and Kordick, 2006) and therefore different perceptions of the need for a decision.

Values

Family values, spirituality and feelings of support have been identified as factors influencing decision making for medically complex infants (Allen, 2014) and this has similarly been identified for children with cerebral palsy (Mahant et al., 2011) and palliative care needs (Carroll et al., 2012). In order for families to make difficult decisions, Renjilian et al. (2013) found that parents of children accessing palliative care services often utilised heuristics, defined as *"decision making aids or shortcuts expressed verbally as terse rules of thumb"* (p.e567), to help make and communicate decisions. Such heuristics included a consideration of the degree of suffering and quality of life, suggesting that these are key factors and values by which parents make decisions.

Maintaining normality

Families of children with chronic illnesses desire to maintain normality and resist the intrusion of the illness into family life. Normalisation has been identified as an important part of families living in the midst of chronic illnesses, in which over time following diagnosis, the illness becomes part of the new normal and individuals incorporate selected pieces of information into their life story to support their accounts of living life as normal (Robinson, 1993). In her secondary analysis of multiple datasets, Robinson (1993) found that the main way in which normality was demonstrated to others was through covering up, a strategy aimed at reducing the visibility of any problems associated with the

child's condition. Furthermore, it has been suggested that attempts at preserving normality are protective for the family and demonstrate successful adaptation to the child's condition (Knafl et al., 2010).

Research has shown that leading a normal life is valued by and is a significant goal for parents of children with a variety of chronic or life-threatening conditions (Earle et al., 2007, Nelson, 2002, Nelson et al., 2012, Smith et al., 2015a, Smith, 2010, Carnevale et al., 2006a). Furthermore, parents feel a moral obligation to do the right thing for their children and for children with cleft palate, they saw this as being to opt for 'normalizing' surgery (Nelson et al., 2012). This demonstrates that maintaining normality may be a key goal of parents' decision making and therefore, the moral work that they undertake in ensuring that visible signs of the child's condition are minimised. Such parental desire to protect their child and family's normality was similarly reported by Bluebond-Langner (1996) for children with cystic fibrosis, which is a less 'visible' condition. The desire to maintain normality been shown to result in avoidance of decision making (Bluebond-Langner et al., 2016). Particularly for the decision regarding gastrostomy placement for children with neurological disability, the need for families to maintain a sense of normality has been identified as a significant factor in parental decision making (Guerriere et al., 2003, Craig, 2004, Mahant et al., 2011). However, Smith (2010) described that alongside the value that families placed on normality, they integrated this with watchfulness and vigilance.

Expertise

During the course of a child's illness, parents develop expertise regarding their child's care. This expertise develops through their specific and ongoing experience caring for their child and the acquisition of technical skill and knowledge, which enables them to detect small changes in the child's condition (Kirk et al., 2005, Nicholson, 2012, Reeves et al., 2006, Smith et al., 2015a). Parents of children with rare conditions may also seek to educate themselves about the condition and treatment, subsequently feeling a sense of expertise about the condition itself (Dewan, 2017), and in some cases perceive that they possess superior expertise to professionals (Wood et al., 2010, Dewan, 2017). Feelings of expertise and confidence sometimes results in parental participation

in, or challenge to, professional's decision making (Wood et al., 2010, Gibson, 1999). When parents develop this confidence and sense of expertise, they may begin to trust their own opinion and have at times been shown to make complex clinical decisions with or without reference to professionals (Thorne et al., 1997, Kirk et al., 2005).

Moral work

It is a socially accepted norm that parents take responsibility for their children and make decisions which put their needs first (Pilnick et al., 2011). This social expectation may be internalised by parents, resulting in a moral responsibility to accept the role of ultimate decision maker. Moral work has also been recognised in relation to gastrostomy decisions, where mothers use justification as a mechanism of maintaining their identities as good mothers (Craig, 2004).

For children with cancer and serious illnesses, evidence suggests that parents perceive that being a good parent entails trying to do the best thing for their child, being present with them (Hinds et al., 2009), making sure their child feels loved, making informed decisions, focussing on the child's health (Feudtner et al., 2015) and putting the child's needs above their own (October et al., 2014). Furthermore, some research suggests that despite the difficulty that it causes them personally, mothers take on an advocacy role for their child and consider this to be the role of a good mother (Todd and Jones, 2003, Ryan and Cole, 2009). Carnevale et al. (2006b) investigated parents' moral experiences in making life-support decisions for their child and found that they often include significant feelings of guilt, even regarding aspects of the child's illness which they have no control over. Guilt was also identified by Atout et al. (2017) who found that the fear of guilt meant that decision making was difficult and that mothers were prepared to defer to professional guidance or to seek out more information in order to be better informed and to avoid these feelings in the future. These findings demonstrate the overall moral factors which influence the way in which parents make decisions, yet moral work has not been explicitly researched or discussed in relation to children with chronic or palliative care needs.

2.2.2. Professional factors

Perception of the evidence

As with the family factors, professionals' situation awareness alerts them to the need for action and this may present in the form of clinical test results for the individual child. When making decisions professionals privilege research evidence and the results of clinical investigations (De Vos et al., 2015, Lotz et al., 2016, Heath et al., 2016). In contrast to parents' interpretation of these, the research suggests that professionals take such results at face value and prioritise them because they can be "*verified by objective 'factual' evidence"* (Heath et al., 2016, p.189). Such evidence will influence the option which professionals feel is appropriate and therefore the way in which they engage families in decision making.

Culture

Culture has been demonstrated to be an influential factor in professional decision making. Empirical evidence has demonstrated that variation exists in neonatal end of life care decision making between different countries (Cuttini et al., 2000, Rebagliato et al., 2000) suggesting that the cultural background of the professional may be influential in their decision making. Carnevale and Bibeau (2007) recognise that France practices a significant degree of medical paternalism, stating that *"physicians are the recognised authorities in France for judging appropriate medical treatment for critically ill children"* (p.129). Different strategies for making treatment decisions exist in different countries, particularly in regard to neonatal intensive care decisions (van Zuuren and van Manen, 2006). The UK often employs an individual prognostic strategy where intensive care is initiated and the likely prognosis re-evaluated on a regular basis to determine whether treatment should be withdrawn. Other countries opt for a strategy relying on statistical prognoses to guide the initiation of life sustaining treatment, or near certainty of a poor prognosis to withdraw life sustaining treatment (van Zuuren and van Manen, 2006). These studies which demonstrate cultural variation were conducted within the NICU setting and therefore may not be transferable to the wider paediatric population. Differences in decision making for neonates, and children or adults, have been found, suggesting that the status of the neonate, from a cultural perspective, may influence the decision

making (Dupont-Thibodeau et al., 2017) and may be responsible for the different findings from the neonatal literature. However, given the presence of some cultural difference about the status of the neonate, it could be supposed that other cultural variation exists and may therefore impact on decision making.

Values and moral work

Alongside cultural context as a professional factor in decision making, individual professionals' perceptions are influential. Orfali (2004) found in her study of French and Canadian neonatal ICU's that despite the different context in terms of parental autonomy, in both contexts, medical staff controlled the decision making through their use and conceptualisation of uncertainty in relation to prognosis. Randolph et al. (1997) similarly found that even within the same cultural context professionals prioritise different factors in order to make an end of life care decision for the child. This variability in practice has been termed the 'roster lottery' and in an ethical analysis of the literature it has been argued that such individual professional factors are irrelevant to patient care and should not exist within decision making (Wilkinson and Truog, 2013). The presence of physician related variability within a single cultural context signals to the role of their personal values in decision making. Such variability was reported by Richards et al. (2017) who found that within one unit, four different approaches to communicating the options to the families were used by physicians, further suggesting that individual factors relevant to the physician influence their decision making.

Within the literature professionals have narrated the need to act as moral beings in their practice. Research suggests that nurses feel that they are perceived as being good nurses when they conform to the norms of their practice area and when they agree with the doctors' recommendations (Sørlie et al., 2003). Research with doctors has revealed that tensions exist in their decision making, representing internal moral conflict and distress which occurs when professionals carry out care at the parental request, which they do not feel is ethically appropriate (Burns et al., 2001, Palda et al., 2005, Austin et al., 2009). Moral conflict sometimes resulted from pressure from parents, previous experience of some children surviving and the moral difficulty of withdrawing treatment (Sørlie et al., 2001, Sørlie et al., 2000, Pye, 2013). When

disagreements between individuals occur and decisions are questioned, the moral integrity of the other individuals is brought into question (Sørlie et al., 2000).

The desire to avoid moral distress may explain why professionals may use directive communication or persuasion, in order to ensure that the child receives the care that they assess as being ethically appropriate (Gibson, 2001, Kinali et al., 2006) and further outlines the moral work being undertaken in the process of decision making.

2.2.3. Relational factors

Information framing and persuasion

An integrated review of the literature conducted by Allen (2014) revealed that parental decision making is influenced by the information provided to them and how this information is communicated. As described above, professionals' values influence their decision making and the way in which they are likely to communicate the options to families. Although Richards et al. (2017) did not investigate the reasons for professionals' option framing, their study about end of life decision making in the PICU demonstrated variation in the way in which they presented options to families. Land et al. (2017) presented findings which also suggested professionals have different approaches to communicating and framing options, in their review of paediatric and adult conversation and discourse analytic studies. Decision making studies using vignettes have also demonstrated that subtle mechanisms of communication, such as positive or negative framing, or communication of the hospital's default policy, influence neonatal decision making (Marteau, 1989, Haward et al., 2008, Haward et al., 2012).

The way choice is communicated and understood also influences decision making. For many decisions in paediatric oncology (Hinds et al., 2000, Coyne et al., 2014, Day et al., 2016, de Vries et al., 2013, Pentz et al., 2012, Overbay, 1996), NICU (Orfali and Gordon, 2004) and neurology (Smith et al., 2013, Heath et al., 2016), parents perceive there to be only one choice. In some situations there may only be one viable option, but in other cases families' perception of choice may be influenced by the way in which it is communicated. In addition to

the more passive strategy of information framing, persuasion has been identified as an active strategy which is used in many care decisions (Radford et al., 1997, Thorne et al., 1997, Craig, 2004, Linnard-Palmer and Kools, 2005, Verhagen et al., 2009, Power, 2012, Brotherton and Abbott, 2012). It has been recognised that persuasion may be perceived as a moral duty for professionals (Barilan and Weintraub, 2001, Maclean, 2006, Swindell et al., 2010, Smith and Pettegrew, 1986), thereby explaining its presence in decision making. Power (2012) highlighted that persuasion is a significant feature of the work that professionals undertake when they encounter ethically complex decisions in the PICU. This occurs through 'preparing the ground' by sharing information in advance of the decision and framing the communication towards the decision which the professionals feel is appropriate. Radford et al. (1997) and Verhagen et al. (2009) identified that in addition to this, professionals also sometimes enlisted the help of others in their attempts to persuade families of a decision.

Fighting and advocacy were identified as the key strategies employed by families in decision making disagreements with professionals. Price et al. (2012) recognised the metaphor of fighting for children with life-limiting conditions, identifying it as a means for parents to overcome service constraints and barriers to accessing care. This was in contrast to families of children with cancer, who used it only as a metaphor against the disease (Price et al., 2012), thus demonstrating the possible differences in decision making for children with life-limiting and life-threatening conditions. The need for families to fight to overcome barriers to care and ensure that parental expertise is considered in the decision making process has been acknowledged across the health and education settings (Duncan, 2003, Lindblad et al., 2005, Hess et al., 2006, Ryan and Cole, 2009, Nicholson, 2012). The presence of persuasion and fighting highlights the disagreements which can occur during decision making and the employment of strategies to ensure agreement is reached.

Trust

Trust has been identified as an important factor in the relationships within which decision making takes place. Primarily this has been in relation to the family-professional relationship (Nicholson, 2012, Forbat et al., 2015, Davies et al., 2017), but has also been highlighted as an important consideration when children

and young people are engaged in decision making, as they refer to their parents for advice (Nicholson, 2012, Mitchell, 2012, Broome and Richards, 2003). Other research has alluded to the presence of trust as an essential factor when making difficult decisions about the child's medical care (Jackson et al., 2008, Zaal-Schuller et al., 2016b) and the implications for decision making when this trust is damaged (Jackson et al., 2008). Nevertheless, Davies et al. (2017), in their qualitative multicentre longitudinal study, found that even when trust is damaged, parents utilise mechanisms of containment in order to ensure that they maintain at least one trusting relationship within which care decisions can be made. In their study, no situations of significant conflict were observed (Davies et al., 2017), suggesting that containment may prevent the escalation of overt conflict.

Conflict

Conflict has been identified as a frequent and anticipated feature of family-professional interactions for children with intellectual disability, and one which impacts on decision making (Todd and Jones, 2003). This results from parents' perception of a power imbalance, a feeling of not having their concerns listened to and through encountering barriers to accessing services (Todd and Jones, 2003). Conflict has been identified to occur particularly in relation to end of life care decision making (Verhagen et al., 2009, Archambault-Grenier et al., 2018) and occurred as a result of interaction with different physicians, differences in values and beliefs and the emotional burden of the decision (Archambault-Grenier et al., 2018). Moore and Kordick (2006) investigated conflict in the relationship between families of children with cancer and their healthcare providers, resulting from divergent perspectives between families and professionals. They identified the presence of five types of conflict: data, interest, structural, relationship and value conflict (Moore and Kordick, 2006).

Different parental reactions to conflict have been identified, with some parents employing passive tactics, such as avoidance (Moore and Beckwitt, 2003). Assertive and confrontational reactions to conflict also occur and have frequently been described in the literature using military metaphors such as 'resistance' or 'fighting' (Ryan and Cole, 2009, Duncan, 2003, Hess et al., 2006, Nicholson, 2012, Lindblad et al., 2005).

McKeever and Miller (2004) discuss the need for parents to 'fight' in relation to Bourdieu's concept of symbolic capital (power in relation to role, legitimacy and authority). They assert that professionals possess more symbolic capital and thereby claim authority, which results in symbolic violence (practices which create and maintain social inequalities). Although the description of fighting suggests antagonism, some parents of disabled children learn how to negotiate power through the employment of intermediaries with greater symbolic capital, such as nurses, to advocate on their behalf (McKeever and Miller, 2004). When conflicts remain unresolved and occur in the presence of poor communication and relationships, it can result in a breakdown to trust (Forbat et al., 2015).

Communication

A quantitative questionnaire study investigating the experiences of parents of children with Cerebral Palsy in relation to communication, demonstrated that overall, parents are satisfied with the communication between themselves and the various professionals involved in their child's care (Gulmans et al., 2009). However, other, predominantly qualitative, research has identified that parents have some concerns about the communication between themselves and professionals. Such concerns related to the sensitivity with which diagnoses are made (Moore and Kordick, 2006, Lindblad et al., 2005, Graungaard and Skov, 2007, Murrell et al., 2017), the extent to which their expertise is considered (Moore and Kordick, 2006, Graungaard and Skov, 2007, Kirk and Glendinning, 2002, Murrell et al., 2017) and their perception that sometimes information is not communicated, or is intentionally withheld (Kirk and Glendinning, 2002, Davies et al., 2017, Smith et al., 2015a).

The literature in relation to inter-professional communication, however, demonstrates a much more widespread concern. Parents in the study by Gulmans et al. (2009) reported in the quantitative survey that the majority of professionals communicated well with each other. However, despite this finding, their free-text responses demonstrated many concerns about information not being communicated between different professionals. These findings were mirrored in other studies in which parents described a lack of inter-professional communication (Golden and Nageswaran, 2012, Dewan, 2017, Lindblad et al., 2005, Kirk and Glendinning, 2002, Gulmans et al., 2009). Findings demonstrate

that parents and professionals believe that parents have a very important role in inter-professional communication. Despite this, neither group would feel comfortable with parents acting as the primary mechanism of communication (Stille et al., 2007). Nevertheless, parents' perception of poor inter-professional communication has resulted in many parents feeling a responsibility to act as the primary communicator and care co-ordinator (Kirk and Glendinning, 2002, Gulmans et al., 2009, Golden and Nageswaran, 2012). Key-workers have been promoted as a way of removing this co-ordination burden from families (Liabo et al., 2001, Together for Short Lives, 2013a, Rodriguez and King, 2014). However some research has highlighted that this role may not be meeting families' needs, as it is variable in terms of provision (Greco and Sloper, 2004) and lack of confidence in the key worker means that families are unable to reap the benefit of the role (Rodriguez and King, 2014, Greco et al., 2007).

Alongside the family perception of lack of inter-professional communication, research amongst physicians has also confirmed a perception that their communication with their colleagues is sub-standard. In a survey of children's hospitalist and community physicians, Harlan et al. (2009) found that only 50% of their respondents rated the communication with each other as good, very good or excellent. Similar results indicating poor inter-professional communication were found by Stille et al. (2003b) and Ruth et al. (2011). Research in this area has demonstrated that physicians perceive that lack of inter-professional communication poses a barrier to optimal care and may pose risks to the child's care (Stille et al., 2003a, Stille et al., 2006).

Decisional responsibility

Individuals involved in the process of decision making are reported to perceive varying degrees of moral responsibility for their participation. Cross-sectional research with parents has highlighted their commitment, in principle, to engaging their child in decision making (Maurer et al., 2010, Hinds et al., 2000), although these studies were conducted in oncology, where choices are limited due to disease specific protocols. Children in such settings are aware of the limited choice and trust their parents and professionals to make decisions for them (Coyne et al., 2014). Such findings, however, may not be transferable to the life-limited population, where equipoise may be more common and choice

more readily available. The provision of choice to the child regarding timing of treatments and non-critical decisions has been described as a way for parents to engage children in decision making and assist them to develop autonomy (de Vries et al., 2013, Coyne et al., 2014, Miller, 2009). However parents may use information framing in their communication with the child (Miller, 2009). Austin et al. (2017) found that in engaging their children in decision making, parents framed the information positively, so as to alleviate anxiety and concerns. Parental involvement for children and young people who are competent to make their own decisions however, has been shown to be requested and valued (Mitchell, 2012, Skyrme, 2016) suggesting that children do not desire ultimate decisional responsibility.

The existing research suggests that desired parental decisional responsibility is individual to the family (Pyke-Grimm et al., 2006, Mack et al., 2011, Carroll et al., 2012, Sullivan et al., 2014). This body of literature however, suggests that many parents feel a moral responsibility to make decisions for their child (Carroll et al., 2012, Sullivan et al., 2014), although other research has conversely indicated that parents prefer to make shared decisions, or delegate responsibility to the professionals (Pyke-Grimm et al., 2006, Mack et al., 2011). The difference in context between these studies may account for the differences in preferred parental decisional responsibility. Carroll et al. (2012) and Sullivan et al. (2014) investigated children with a range of life-limiting and life-threatening diagnoses, whereas Pyke-Grimm et al. (2006) and Mack et al. (2011) investigated children with cancer, with the latter being newly diagnosed. Feelings of expertise have been linked to parental decisional involvement (Wood et al., 2010, Gibson, 1999) and the differences in parental decisional responsibilities in these studies may be explained by differences in parental expertise between those established in palliative care and those with a new cancer diagnosis.

Within the PICU and NICU setting, similarly diverse perceptions of parental decisional responsibility have been identified (McHaffie et al., 2001, Madrigal et al., 2012, Zaal-Schuller et al., 2016a). Professionals in this context often perceive making such end of life decisions to be too burdensome for parents and advocate for a SDM approach (McHaffie et al., 2001) or adopt the responsibility

for making end-of-life care decisions, in consultation with the family (Carnevale et al., 2011, Carnevale et al., 2012, Kahveci et al., 2014). Within the UK a non-dissent approach has been described to occur for ethical dilemmas in the PICU. Power (2012) identified that for withdrawal or withholding life-sustaining treatment decisions, professionals feel that they do not need agreement from parents, but that they need to ensure that parents do not disagree. He described that often parental disagreement is overcome by persuasion and negotiation, suggesting that parental involvement may be a gesture aimed at obtaining their agreement, rather than active participation in decision making.

Differences in parental decisional involvement appears somewhat associated with the cultural context. Carnevale et al. (2012) and Orfali and Gordon (2004) found that differences exist between North American and French contexts, with parents in the French context having lesser decisional autonomy. Such association between decisional role and culture can also be inferred from individual studies, such as Kilicarslan-Toruner and Akgun-Citak (2013) and Atout et al. (2017) who interviewed parents of children with cancer in Turkey and Jordan respectively, and Mack et al. (2011) studied parents of children with cancer in the USA, with the former having a less active role than the latter. However, despite the stated preference for decisional responsibility, Orfali and Gordon (2004) found that in cases where decisional responsibility was perceived to be shared, parents were more satisfied with the outcome. The relational factors involved in decision making are complex and this may be particularly evident in decisions which are ethically or emotionally difficult.

2.2.4. Ethical and legal issues

Ethically and emotionally difficult decisions

Decisions related to tube feeding have been demonstrated to be challenging for families to make, due to the psychosocial issues surrounding this choice (Craig, 2013). Gastrostomy feeding may be viewed by parents as giving up hope and accepting failure, alongside presenting feelings that normality had disappeared (Morrow et al., 2008, Brotherton and Abbott, 2012, Thorne et al., 1997). Although professionals often recognise early on that tube feeding may be in the best interest of the child, parents may at times experience emotional resistance, but in hindsight often express the decision as the best thing for the child (Radford

et al., 1997). The professional perception that parents are pleased with their decision in retrospect, may result in more active persuasion and pressure towards families to agree to the decision.

Some research with professionals has demonstrated their awareness of the emotional difficulty of these decisions for parents (Rouse et al., 2002, Morrow et al., 2008). However professionals prioritise the child's physical health in decision making, seeing this as primary over other concerns, whereas other psychosocial factors are considered equally as relevant by parents (Morrow et al., 2008). This signals to the possibly conflicting moral work being undertaken by families and professionals, and professionals may struggle to fully understand parents' hesitation of a treatment which will improve their child's physical health.

For the emotionally and ethically challenging decisions around end of life care, parents and professionals may preference different factors (Streiner et al., 2001, Tomlinson et al., 2011). To date, however, there is a lack of research which has investigated the basis for these different preferences, or developed strategies to arrive at an agreeable solution. Lotz et al. (2016) elicited doctors' views of end of life care decision making in relation to two vignettes presenting options for life-sustaining treatment and found that doctors utilise a range of factors for these decisions, including treatment related and prognostic factors, a consideration of the family, the child's current health status, their own clinical experience, and legal and economic considerations. However there are likely to be limitations to the transferability of vignette studies into clinical practice, as the emotional and relational factors which influence decision making cannot be easily recreated in a vignette.

Despite the policy drive towards advance care planning (ACP) (National Institute for Health and Care Excellence, 2016), barriers to having such discussions with families, for example, time, professional worry about communication and a mismatch between professional and family belief about prognosis, remain (Durall et al., 2012, Heckford and Beringer, 2014, Mitchell and Dale, 2015). There has been little empirical research conducted regarding the implementation of ACP in paediatrics and the current literature has largely excluded the voice of professionals (Lotz et al., 2013). Alongside ACP decisions, parents of critically

ill children are required to make other decisions which they perceive as significant, such as, whether to inform the child about their care, trust the medical team, or accept hospice or respite care (Michelson et al., 2013). ACP, including end of life care planning, may begin many years before the child's death and when making such decisions parents prioritise the ability to change their mind in light of the child's changing condition or factors relevant at the time (Beecham et al., 2016).

Best interests

The literature suggests that when ethically or emotionally challenging decisions take place, disagreement may occur and individuals' culture and experience influences their views about what is morally appropriate (Carnevale, 2005). The concept of best interests has been widely used in health care in order to overcome disagreement through reference to an objective assessment of what is best for the child. Although McHaffie et al. (2001) argue that *"both in law and in ethics, parents do, in most cases, have the authority to decide about their children's welfare"* (p.108), the legal framework and professional codes within the UK place limits upon this. The General Medical Council guidance (2007) states that doctors must act in the child's best interests[4] to prevent harm, even in situations where parents disagree, and the law places the ultimate responsibility for such decisions in the hands of the court (Children Act, 1989). The need for the courts to intervene is rare (Wilkinson and Savulescu, 2017b), but when disputes about a child's medical treatment cannot be resolved informally, an application to the courts may be necessary (General Medical Council, 2007).

Despite reference to the 'best interest' principle in law and codes of practice, it is argued that it is flawed as a means for assessing the limit at which the state should intervene, as it is subjective and value-laden (Alessandri, 2011, Shah et al., 2017). Other authors have argued that it is indeterminate and fails to respect the family (Salter, 2012, Diekema, 2004, Birchley, 2016b, Shah et al., 2017) by *"pit[ting] the state's determination of "best interest" against that of the parents"* (Diekema, 2004, p.247), with the harm principle being proposed as a

[4] The term 'welfare' is predominantly used in the legal context and the term 'best interests' is used in the medical context, but refer to the same principle. The term 'best interests' will be used in this thesis, as it is the terminology most frequently used in relation to medical decisions.

solution to this indeterminacy. However, the harm principle is not without its flaws, as judgements about harm can be as indeterminate as those about best interests and using the terminology of harm implies condemnation, whereas best interests does not, and therefore may be less antagonistic in conflicted decisions (Birchley, 2016a). McDougall and Notini (2014) undertook a systematic review of the normative literature and concluded that none of the principles outlined for decision making in conflicted cases provide sufficient ethical guidance for professionals and that such guidance is required.

There is medical and ethical consensus that courts ought to intervene when the child is being put at risk. However, further consideration is needed of what constitutes a considerable harm to the child and what constitutes 'reasonable disagreement' to justify overruling the parents' request (Wilkinson and Savulescu, 2017b). Given the indeterminate nature of the current ethical principles which guide decision making in conflicted cases and place it in the hands of the law, the concept of parental responsibility is problematized. As such parental responsibility can be argued to be *"little more than a façade"* (Gillett, 2017) which requires legal and/or medical agreement. A capacity based assessment for parents (similar to the Mental Capacity Act, 2005), alongside some criteria to assess that the decisions were not made out of malice, as a way to value parental responsibility in situations of moral conflict, has been suggested to overcome these issues (Gillett, 2017), but this area requires further work.

In conflicted cases, moral conflict occurs as a result of disparate moral viewpoints. Carnevale (2005) argues for an individual approach to resolving moral conflict through mediational work in order to *"reconcile conflicts among the multiple voices in the clinical encounter by seeking to 'fuse' the disparate moral views"* (p.249). Such a style of decision making is akin to the shared mind approach advocated by Epstein and Street (2011), which recognises the benefits of relational autonomy in which complex decisions can be processed using the cognitive capacity of multiple persons, rather than overwhelming the capacity of one person. Such an approach assists in the distributing of emotional and cognitive effort in making the decision.

2.3. Systematic Review and Thematic Synthesis: Decision making for children with life-limiting conditions

The previous section has presented a broad overview of the literature regarding decision making for children with chronic illnesses, life-limiting and life-threatening conditions. Given the differences in these populations and the population of children with life-limiting conditions, the foregoing section may have highlighted some factors which are not applicable to children with life-limiting conditions. Alternatively some of the more relevant factors to this population may have been overshadowed. An investigation into decision making with the specific focus being on children with life-limiting conditions is therefore warranted in order to illuminate any similarities or differences in the process for this group.

This section will present a systematic review and thematic synthesis of the specific literature for decision making for children with life-limiting conditions[5]. The objective of this section was to provide a review and synthesis of the qualitative research with the specific focus of decision making and future planning, solely for children with life-limiting conditions.

2.3.1. Methodology

Thematic synthesis of qualitative research aims to provide a re-interpretation of previous findings, such that the conclusions go beyond the conclusions developed within the individual research studies (Thomas and Harden, 2008, Sandelowski and Barroso, 2006). It is a data driven process and involves extracting and coding the text, developing descriptive themes and sub-themes, and generating analytical themes (Thomas and Harden, 2008). Inferences are made regarding the relationships between the descriptive themes and research questions, resulting in abstract analytical themes.

Eligibility Criteria

The eligibility criteria for inclusion in the review are outlined in Table 1. Studies were included if they investigated decision making for people with life-limiting

[5] This provides an updated version of the previously published review: POPEJOY, E., POLLOCK, K., ALMACK, K., MANNING, J. C. & JOHNSTON, B. 2017. Decision-making and future planning for children with life-limiting conditions: a qualitative systematic review and thematic synthesis. *Child: Care, Health and Development,* 43**,** 627-644.

conditions, as long as the study involved some life-limited children (28 days-18 years old) in the sample. Due to the lack of research in the area and the need to better understand the process of decision making, it was decided to include studies which featured adults in their sample, although it is acknowledged that this has implications for the results of the synthesis.

Inclusion Criteria
• The study must focus on children (>28 days old) and young people with a life-limiting conditions within categories 2-4 (see Figure 1). • The study must involve health care professionals, family members, 'significant others' or the child, who make decisions about the child's care. • The study must be qualitative in design or have a qualitative component. It will consider studies that focus on qualitative data including, but not limited to, designs such as phenomenology, grounded theory, ethnography and action research. Descriptive qualitative studies that describe the experience or describe the effects of the experience will also be considered. • Studies recruiting professionals from ICU's if the focus of investigation is children with pre-existing life-limiting conditions.
Exclusion criteria Any studies: • including children or young people with life-threatening conditions. • where families or professionals are recruited from ICU's, unless the focus is specifically on decision making for children with life-limiting conditions. • recruiting families or professionals from neonatal ICU as many of the participants will be discussing children under the age of 28 days and therefore outside the criteria of the study • using purely quantitative methods.

Table 1: Thematic synthesis eligibility criteria

Searching for literature

A systematic approach to literature searching was taken and various bibliographic databases, covering the fields of health care and psychology, were searched; these were CINAHL, Medline, Embase and PSYCHInfo. Indexed terms and keywords were used to find papers relevant to the review question. In order to find any relevant literature outside of the health field, a free text search was conducted in ProQuest at the advice of a librarian. Additionally, EThOS (E-

Theses Online Service) was searched using the words “life limiting” AND “child”, in order to identify unpublished doctoral theses. The initial electronic search was conducted in December 2014 and was updated periodically until May 2017. Authors of relevant conference abstracts were also contacted to identify if they had articles presenting the findings from their research or for unpublished work.

Screening of Studies

Searching and screening of studies is summarised in Figure 3. The title and abstracts of the papers retrieved from the searches were read and seventy-two papers retained following the initial screening and removal of duplicates. Sixty-four of these studies were excluded based on the sample population, focus of the study, use of purely quantitative methods or because they were unable to be obtained despite exhaustive efforts. Eight studies met the inclusion criteria. The reference lists of the included studies were screened and one further eligible study was identified. On searching for this study, two other papers presenting different findings from this study were found and included. A total of eleven studies were included in the review.

Appraisal

Appraisal of included studies was conducted in order to consider crucial aspects of the research, such as whether the methods and sample are appropriate to investigate the aim (Dixon-Woods et al., 2004) and ensures that the value of the research is balanced against any methodological weaknesses (Edwards et al., 2000). Walsh and Downe (2006) collated a number of appraisal checklists into one appraisal guide and this was used in this synthesis to provide an overview of the limitations of individual studies and guide me in identifying the quality of included studies. Following appraisal, all studies were included within the synthesis.

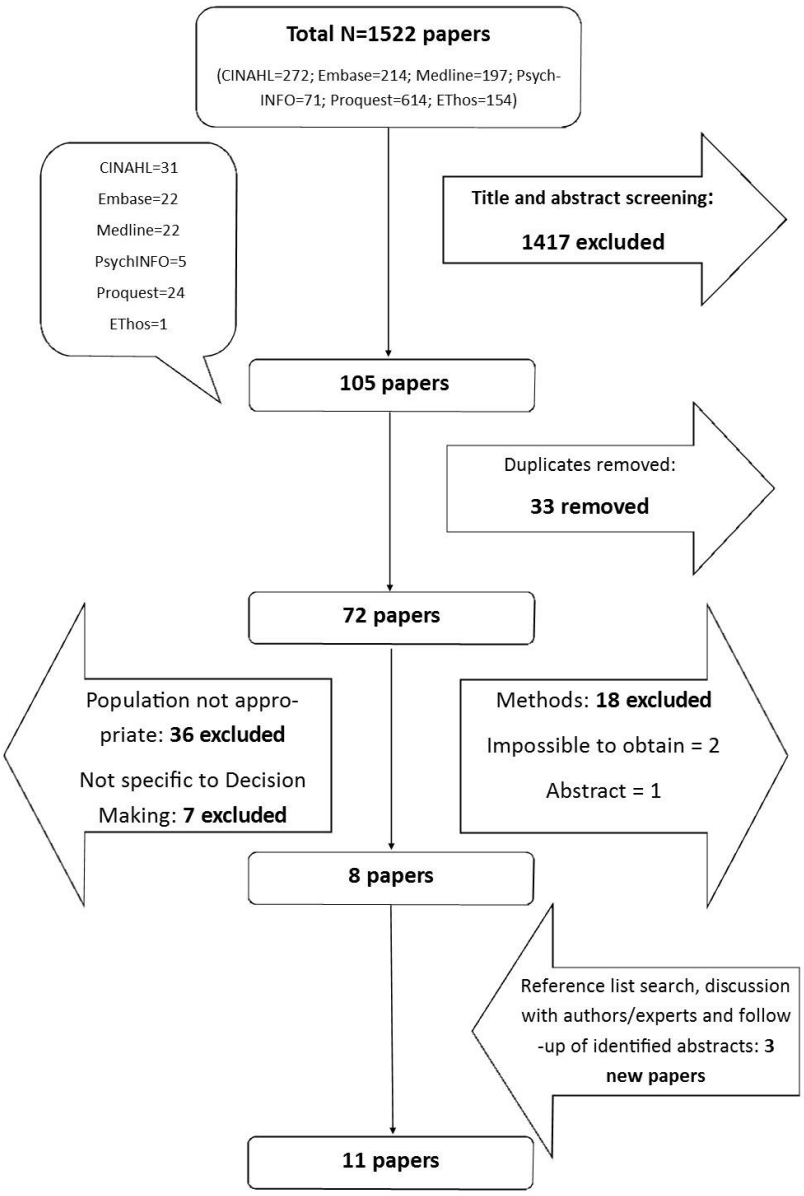


Figure 3: Detail of literature search

Description of papers

Included papers focussed on a range of stakeholders and the included studies collectively had a sample of 129 parents or guardians (henceforth referred to as parents for brevity), 45 children and young people and 43 professionals. Three papers included only parents, one paper included only the child/young person and two papers included only health professionals. Five papers sought more than one perspective, three included parents and the child/young person, in the fourth paper, cases were comprised of a mixture of the child/young person, the parent/carer and other family members, and the fifth paper included both parents

and professionals and was the only study to do so. Five papers included young people (up to the age of 37 years) and/or their parents. A summary of the papers is provided in Table 2.

Study, country	Purpose	Methodology and methods	Study Participants	Context	Main themes and sub-themes identified
Erby et al (2006), USA	Experiences of and attitudes to advance care planning for parents of DMD patients.	Exploratory, cross-sectional, semi-structured interviews.	Convenience sampling. 17 parents from 16 families of children and young people with DMD. Ages 8-37 yrs.	Recruited from Muscular Dystrophy groups.	1) Advance Care Planning (ACP) Communication 2) Factors that influence ACP Communication: a)Competing demands b)Distancing from diagnosis c)Absence of communication guidance d)Stages of receptivity 3) Navigating the unspeakable
Mitchell et al. (2011) UK	The realities of choice-making processes within families, exploring the respective roles and experiences of young people and their parents	Exploratory, longitudinal, semi-structured interviews.	Purposive sampling. 27 young people from 24 families and 44 parents from 33 families. Ages 13-22 yrs.	From 2 children's hospices.	Themed under the interview topics: 1) Young people's perspectives on making choices 2) Young people's experiences 3) Factors considered by parents regarding involving the young person 4) Moving on from full time education – experiences of parents 5) Factors and processes supporting satisfaction
Mitchell (2012) UK	Perspectives of disabled young people with degenerative	Exploratory, longitudinal,	Purposive sampling. 27 young people from	From 2 children's hospices.	1) Parents' role in choice making a)Nature of the choice

	conditions on making choices with parents and peers.	semi-structured interviews.	24 families and 44 parents from 33 families. Ages 13-22 yrs.		b)Which parent helped c)Being listened to d)Value of shared decision making e)Parents' changing and evolving role 2) Role of peers
Mitchell (2014), UK	To explore the perspective and experiences of disabled young people with degenerative conditions as they face significant medical interventions and engage in decision-making processes.	Exploratory, longitudinal, semi-structured interviews.	Purposive sampling. 10 young people. Ages 13-22 yrs.	From 2 children's hospices.	1) The decision a)Why was the decision raised b)Decisional Roles c)How were the decisions perceived? 2) The options 3) Making the choice a)Invasiveness of option b)Uncertainty and risks c)Quality of Life d)Maintaining Health e)Retaining Independence f)Parents' wishes g)The actual decision 4) Reflection on choice
Mitchell and Dale (2015), UK	Experiences of PICU staff in advance care planning for children with LLI.	Exploratory, cross-sectional, semi-structured interviews.	Purposive sampling. 14 professionals (8 consultants and 6 senior nurses).	One PICU in Leeds.	1) Recognition of a LLI trajectory 2) ACP as a MDT structured process 3) Value of ACP 4) Inverse consequences of inadequate ACP
Nicholson (2012),	Decision processes in use of	Exploratory,	Purposive sampling. 9	From one children's	1) Decision Factors

UK	technological support in children with LLI.	cross-sectional, unstructured interviews.	bereaved parents, 10 parents of children with limited/no decisional capacity, 6 parents of children with capacity and five young people. Ages 3-32 yrs.	hospice in England.	a)Weighing up the need for technological support b)Weighing up the potential outcomes 2) Decision Features a)Uncertainty b)The individual child c)Parents role as decision makers d)Beliefs and Values e)Gut instincts 3) Process Factors a)Background to decision b)Time c)Professional involvement d)Information for decision making e)The Healthcare system
Popejoy (2015) UK	To understand the 'lived experience' of parents throughout the process of making and revising end of life care decisions for their child.	Exploratory, cross-sectional, semi-structured interviews.	Purposive sampling. 3 bereaved parents from 3 families. Ages 3-15 yrs.	From one Children's Hospital in England.	1) Making decisions a)Timing b)Decision Makers c)Values 2) Revising and Implementing Plans a)Barriers and facilitators to implementation of plans b)Revising plans
Rapoport et al	Explore experiences of	Exploratory,	Purposive sampling.	From one hospital.	1) Introduction of FANH to parents

(2013), Canada	bereaved parents regarding foregoing artificial nutrition and hydration (FANH) decisions for their child.	cross-sectional, semi-structured interviews.	11 parents of 7 children from 6 families. Ages 1 month-15 yrs.	Identified from the Advanced Care Team.	a)Being open to option in relation to perceived poor Quality of Life (QoL) b)Positive aspects of physician who introduced option 2) Decision making process a)Parental agreement b)Importance of perceived family QoL c)Childs perceived QoL d)Importance of consistent message from professionals e)Concerns about being judged
Wissow et al (2001), USA	Test acceptability and feasibility of undertaking advance directive development using a values history.	Exploratory, cross-sectional, semi-structured interviews	Convenience sample within one HIV service. At least 12 parents or guardians (reporting is unclear). 3 children participated. Ages 3-16 yrs.	Took place within normal clinic consultation, just somewhat structured interview to elicit values. Majority of decision makers were not biological parents.	1) Disabilities that would make life not worth living 2) Tolerance of Pain and Risk in Hope of Recovery 3) Past experience 4) Preferences for End of Life Care 5) Preserving life as long as possible 6) Spirituality
Delany et al	Explore acceptability and	Exploratory,	Purposive and	Recruited from one	1)Family empowerment

(2017), Australia	value of a written resource to assist in making end-of-life decisions for children with life-limiting illnesses.	cross-sectional, semi-structured interviews	snowball sampling of professionals involved in provision of care to parents of children with life-limiting illnesses. 18 professionals (doctors, nurses, play therapists, chaplain and social worker).	hospital.	a)Giving words and clarity b)Conversation starter c)'I'm not alone in this' d)A resource to take away 2)Not just for families a)A guide for staff b)Processes of communicating – when to give the resource? c)Processes of communicating – who should be involved? d)Processes of communicating – how to give the resource?
Zaal-Schuller et al (2016) The Netherlands	To compare the experiences of parents and doctors in end of life decision making for children with profound intellectual and multiple disabilities.	Exploratory, cross-sectional, semi-structured interviews.	Purposive sampling of families and snowball sampling of professionals. 17 parents and 11 doctors. Ages 0-19 yrs.	Recruited from a number of organisations in the Netherlands.	1)The influence of previous healthcare encounters 2)Anticipation and timing of the end of life decision making process 3)Provision of information and advice 4)Disagreement 5)Contributions to decision-making 6)The final decision maker

Table 2: Study characteristics

2.3.2. Synthesis

Data contained within a clearly defined results section were extracted for coding and all direct quotes were also extracted. Extracted data were coded in NVivo Pro using a data driven approach and refined through an interactive process of constant comparison. Codes were grouped into descriptive themes and then subsequently synthesised further into analytical themes (see Figure 4). The synthesis has provided a more comprehensive understanding of how decision making works in a wider context, considering the various factors that influence and constrain it, thus providing an understanding at the more analytical and conceptual level. Predominantly the individual studies had a much narrower focus in relation to decision making and the synthesis provided an opportunity to gain a broader overview of decision making. One study (Nicholson, 2012) identified many of the issues included within the synthesis and enabled the identification of these themes in the other studies, providing an opportunity for them to be unpicked and expanded on.

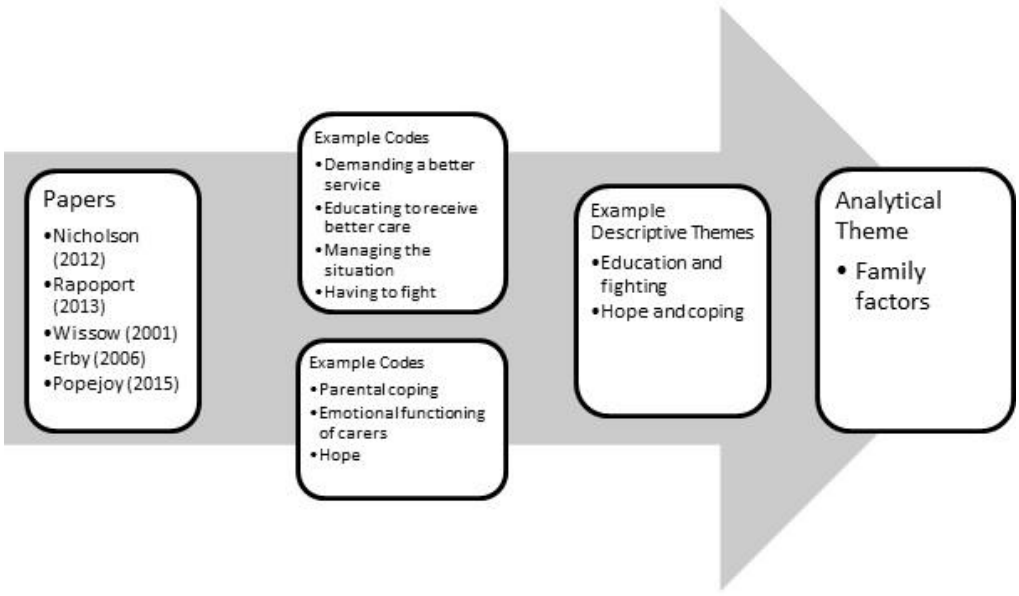


Figure 4: Example of the development of an analytical theme

2.3.3. Results

Nineteen descriptive themes were identified and four analytical themes developed from these. The four analytical themes appeared to represent four different types of factors which influenced the decisions made by families and

professionals: decisional factors, relational factors, family factors and system factors (Table 3). These four factors will be discussed below and quotations to demonstrate the themes are provided in Appendix 1.

Theme	Description
Decisional Factors	Decisional factors were related to the specifics of the decision being made, including the consequences of a decision and the time available to make the decision. Underpinning descriptive themes: Timing of decisions, consequences, decisional considerations, difficulty in decision making, future planning.
Family Factors	Family factors impacted on decision making and depended on their background, culture and experience, awareness of the child's condition and their coping mechanisms. Underpinning descriptive themes: Coping mechanisms, awareness about illness, educating and fighting, family life.
Relational Factors	Relational factors involved individuals' perception of their involvement or choice in decision making, communication within the network of care and trust Underpinning descriptive themes: Gathering opinions, involvement in decisions, trust, agreement in decisions, communication and information exchange, support for decisions.
System Factors	System factors impacted on decision making through bureaucratic constraints and lack of access to support. Underpinning descriptive themes: Problems with the system, factors related to professionals, medicalisation, access to support.

Table 3: Description of Analytical Themes

Decisional Factors

Decisional factors encompassed a range of considerations specific to the decision being made. The extent to which families and children were given

sufficient time to consider the decision and whether the decision could be predicted in advance, appeared to be important (Nicholson, 2012, Erby et al., 2006, Zaal-Schuller et al., 2016b). Individuals held conflicting opinions about whether future possibilities ought to be introduced to them at an early stage or whether they should be considered only when clinically necessary, as this impacted on different families' abilities to cope (Erby et al., 2006, Nicholson, 2012, Mitchell and Dale, 2015, Delany et al., 2017).

Decisions appeared to be considered in relation to previous decisions, interventions and the child's prognosis (Mitchell and Dale, 2015, Rapoport et al., 2013, Nicholson, 2012), rather than occurring as isolated incidents. The possible consequences of a decision, such as, aesthetic implications, the long or short term nature or reversibility of the decision were often considered (Nicholson, 2012, Mitchell, 2014, Mitchell et al., 2011), although individuals had differing perceptions about whether a decision was reversible. One young person identified having a gastrostomy as a decision with lifelong consequences (Mitchell, 2014), whereas other families considered this same decision as reversible (Nicholson, 2012). The child's level of comfort or quality of life had the biggest influence on the decision (Mitchell, 2014, Nicholson, 2012, Rapoport et al., 2013), but parents found the process of analysing the child's quality of life difficult (Rapoport et al., 2013). The difficulty in decision making was often made worse by the emotional and physical exhaustion of parents' caregiving responsibilities (Nicholson, 2012, Popejoy, 2015b).

Advance discussions and planning regarding limitation of treatment offered families time to consider what they want for their child (Mitchell and Dale, 2015, Zaal-Schuller et al., 2016b), although it was acknowledged that such plans can and do change (Popejoy, 2015b, Mitchell and Dale, 2015). Professionals viewed such discussions and plans as a communicative mechanism to ensure that care provided is in the best interests of the child and commensurate with the preferences of the family (Mitchell and Dale, 2015), but expressed that having such conversations was often difficult (Delany et al., 2017, Zaal-Schuller et al., 2016b). The perspective of families regarding the purpose and benefit of such discussions was not identified within the reviewed studies.

Family Factors

Family factors impacted on the decision making process and the decision made. The child's character, views and their disease trajectory acted to influence the decisional process (Erby et al., 2006, Nicholson, 2012). Approaching specific stages of a known disease trajectory often precipitated decisional events. Nevertheless, a great deal of uncertainty existed, making planning for an uncertain future difficult (Erby et al., 2006, Zaal-Schuller et al., 2016b). Family background was also important; those who considered themselves to have a good education and more life experience, both in terms of age and exposure to the health care system, felt that this helped them challenge professionals in decision making. There was discussion of the need to search for information and fight for the appropriate care for their child (Erby et al., 2006, Nicholson, 2012, Zaal-Schuller et al., 2016b) which appeared to be more apparent where distrust in professionals was expressed. Although family background was noted as important, it appeared that parental confidence in making decisions naturally increased over time (Nicholson, 2012).

The family's coping abilities and strategies were also influential on decision making, with some families feeling that they could not cope with certain decisional outcomes (Nicholson, 2012, Wissow et al., 2001). Discussing difficult decisions resulted in a period of decreased coping for some parents (Popejoy, 2015b), which may lead to families resisting planning for the future in order to maintain their ability to cope with daily life (Erby et al., 2006). Other families used spirituality to help them cope (Wissow et al., 2001). Families identified that decision making is part of their life; requiring a delicate balance alongside managing the day to day care of their child and the practicalities of 'normal life' (Nicholson, 2012, Erby et al., 2006, Mitchell et al., 2011). Decisions were also considered in relation to their impact upon the wider family and other children (Rapoport et al., 2013).

Family awareness of the child's condition, prognosis and future possibilities was important in decision making. Families may not be aware of the illness trajectory (Wissow et al., 2001) or what intensive treatment involves or what the consequences would be (Mitchell and Dale, 2015). Families' developing awareness of the seriousness of their child's condition and the implications of

their symptoms over time made decision making easier and allowed them to recognise the need for a particular course of action (Nicholson, 2012). Awareness was developed either by witnessing other children's illnesses or seeking advice from families who had previously made similar decisions (Rapoport et al., 2013, Popejoy, 2015b, Mitchell, 2012, Mitchell, 2014, Wissow et al., 2001, Nicholson, 2012, Zaal-Schuller et al., 2016b). However, some families tried to distance themselves from the diagnosis and associated community (Erby et al., 2006) so may not have had access to this type of support or developed awareness in this way. Professionals felt that they have an important role in facilitating the family's awareness and likely treatment outcomes by providing them with appropriate information (Mitchell and Dale, 2015).

Relational Factors

Families mostly felt involved in the decision making process (Nicholson, 2012, Popejoy, 2015b, Zaal-Schuller et al., 2016b), but sometimes felt that they were not listened to (Popejoy, 2015b). Parents desired to involve their child in decisions (Erby et al., 2006, Nicholson, 2012, Wissow et al., 2001) and the child's feeling regarding the decision was frequently taken into consideration (Nicholson, 2012, Mitchell, 2014, Wissow et al., 2001). However, other parents were concerned about involving their child and protected them from this (Nicholson, 2012). Some young people were or wanted to be actively involved in decision making (Nicholson, 2012, Mitchell, 2012, Mitchell, 2014, Wissow et al., 2001), whereas others did not (Mitchell, 2012, Mitchell, 2014), highlighting the importance of establishing individuals' preferences for involvement. When children reached the legal age to make their own decisions, parents sometimes felt excluded from decision making by professionals (Nicholson, 2012), although young people identified that they valued and desired their parents' input (Mitchell, 2012, Mitchell, 2014). Parents valued the input of professionals in decision making for their child (Nicholson, 2012). Involvement in decision making also related to the presence, or absence, of perceived choice and existed in situations where the child's condition dictated a particular action to be taken, for example, an unsafe swallow leading to termination of oral feeding (Nicholson, 2012). Nicholson (2012) identified that even in situations

with little perceived choice, the process of agreeing to a treatment was important for families as it required them to understand the risks and benefits involved and come to their own conclusion that the treatment is necessary.

Communication and information exchange played an important role in decision making. Some families expressed problems in accessing information due to the rarity of their child's condition or lack of openness by professionals (Nicholson, 2012), whereas other families received open and honest information about the available options (Rapoport et al., 2013). Professionals expressed concern that often communication with families regarding the implications of intensive care treatment for their child is poor (Mitchell and Dale, 2015) and that written resources may help with such communication (Delany et al., 2017). Quality of communication between professionals and the child varied (Nicholson, 2012, Mitchell, 2014, Erby et al., 2006) and families appreciated when their knowledge of their child was welcomed and valued by professionals (Nicholson, 2012, Zaal-Schuller et al., 2016b), but this did not always occur (Popejoy, 2015b, Nicholson, 2012).

Trust within the family-professional relationship was important in facilitating candid conversations with the child and family (Erby et al., 2006, Delany et al., 2017, Zaal-Schuller et al., 2016b) and enabled professionals in certain circumstances to take the burden of decision making away from the parents (Popejoy, 2015b). Many instances were identified where a lack of trust was demonstrated and occurred when professionals did not listen to parental concerns or had previously made errors (Nicholson, 2012, Zaal-Schuller et al., 2016b). In the absence of trust, parents suspected a hidden agenda when treatment decisions were discussed (Nicholson, 2012), which may have impacted on their ability to make an informed decision. Agreement between various stakeholders in the decision also appeared to be important; both agreement within the family, between the family and professionals, and within the healthcare team (Rapoport et al., 2013, Nicholson, 2012, Mitchell and Dale, 2015). Professionals identified that it can be difficult to achieve consensus regarding the appropriate treatment for each child and the difficulty in ensuring that discussions have been had with families prior to an acute deterioration (Mitchell and Dale, 2015).

System Factors

System factors were identified as important and came through strongly in the studies by Mitchell and Dale (2015) and Nicholson (2012) and referred predominantly to health care systems, but also occasionally to social and educational systems. Some families felt that there were too many professionals involved in the care of their child, which made continuity and effective communication difficult (Nicholson, 2012). Rules and procedures often made decision making or accessing care difficult and prevented an individualised service (Nicholson, 2012). Additionally families encountered errors within the healthcare setting, such as medication errors, surgical complications or inability of professionals to notice the subtle signs of a deterioration in their child's condition; often families made the decision to stay with their child continually during a hospital admission, due to lack of confidence in staff (Nicholson, 2012). Additionally, professionals identified that likely deteriorations in the child's condition are often not anticipated or planned for appropriately and children are subsequently given inappropriate treatment, whereas better planning may have resulted in more appropriate outcomes (Mitchell and Dale, 2015).

Another issue related to the healthcare system was the difficulty in accessing advice or support. Families identified the difficulty in obtaining support and advice from their main doctor (Nicholson, 2012) which may have left them to make decisions alone. Other families expressed that their doctor made time for them when required, to discuss any issues (Nicholson, 2012). Families articulated that there is a difference between 'good' and 'bad' professionals who will either help them to navigate the system and overcome the rules, or who rigidly apply the rules (Nicholson, 2012). When families found a 'good' professional, they knew that they could rely on these people to help them make decisions and access the treatment their child needed (Nicholson, 2012). Professionals themselves acknowledged the problems of the system where care is often over medicalised and professionals become emotionally exhausted through providing care and dealing with death (Mitchell and Dale, 2015).

2.3.4. Thematic synthesis discussion

This thematic synthesis has identified numerous influential factors to decision making for children with life-limiting conditions. These were broadly

categorised as: decisional factors, family factors, relational factors and system factors. This review has highlighted that decision making for this population involves the complex interplay of many factors.

Within the wider chronic or life-threatening conditions literature, decisional factors have also been identified as important in decision making. Preference for the timing of the introduction of discussions appears to be individual for each family (Gibson, 2001, Jackson et al., 2008, Dellon et al., 2010, Edwards et al., 2012, Gotz et al., 1997, Noyes et al., 2013) and may impact on perceived choice in the decision (Sharman et al., 2005). Prognostic uncertainty also influences decision making, making it more difficult (De Graves and Aranda, 2005, Kirschen and Walter, 2015).

Family factors were noted to influence decision making and these included experience and awareness of the disease trajectory. Although not specifically identified within the reviewed papers, it is possible that the multiple phases of the child's illness and previous experience of the child's ill health link into these and impact on decision making. Changes in the child's disease trajectory are viewed as significant by families (Wood et al., 2010) and influences the way they view the disease (Bluebond-Langner et al., 2016). It follows that this may factor into the decisions that families make, but this requires further investigation.

Other family factors have also been noted also in the wider literature, with family characteristics, such as hope and the child's Quality of Life (QoL), influencing decision making (Maurer et al., 2010, Higgins and Kayser-Jones, 1996, Tomlinson et al., 2011, Hinds et al., 2000, Lan et al., 2007, Tomlinson et al., 2006). The role of these characteristics however, appear to be different between the thematic synthesis and wider literature. For children with cancer, for example, families pursue cure-directed therapy even when the chance of cure is minimal, as they try to extend the child's time and not give up hope (Bluebond-Langner et al., 2007). Although hope was identified in the thematic synthesis, it did not appear to impact on decision making, rather assisted families to cope with life and the child's illness on a day to day basis. Anticipation of the child's

future in light of a life-limiting diagnosis appeared to be different than for those with a potentially curable illness.

The wider body of literature, including children with life-threatening and chronic conditions has identified the need for families to seek information on their own in order to make informed decisions (Watson et al., 2002, Bluebond-Langner et al., 2007) and exert pressure on health professionals to ensure their preferred course of action is taken (Lindblad et al., 2005, Russell, 2003). This seems to reflect the descriptive theme within the synthesis of 'Educating and Fighting' and may reflect the need for those in regular contact with the health care system to have their individual needs recognised.

Additionally, relationships have been identified in the wider literature as important in decision making. Primarily families feel that decision making is their responsibility (Carroll et al., 2012, Sullivan et al., 2014), but despite this, families value the input and guidance provided by professionals (Rodriguez and King, 2014). The thematic synthesis suggested that families want to be involved in decision making, but did not identify who they considered held the ultimate responsibility for this. The inclusion of children in decision making has also been discussed, although the majority of this research has been conducted with children and young people who are healthy, or have HIV or cancer (Berger, 2013). From the limited research in this area, it appears that there is a consensus that young people have the right to be involved in decision making and future planning discussions (Pousset et al., 2009, Talati et al., 2010). The literature suggests that such discussions are safe to have and increase decisional congruence between young people and their families (Lyon et al., 2010a, Lyon et al., 2010b, Lyon et al., 2009a, Lyon et al., 2009b, Lyon et al., 2013). These findings may not be directly transferrable to children with life-limiting conditions, as many have some degree of cognitive impairment and may need additional support to participate in such discussions. Further research is needed to identify what role children and young people with life-limiting conditions have in making decisions about their care and treatment and the impact of their age and prognosis on their involvement.

The lack of acknowledgement by professionals of families' specific expertise regarding their child has been reported in relation to paediatric chronic illness (Lindblad et al., 2005, Watson et al., 2002, Graungaard and Skov, 2007). This may impact on other relational issues such as trust, which has also been highlighted as problematic in the family-professional relationship for children with chronic illnesses (Graungaard and Skov, 2007, Lindblad et al., 2005, Russell, 2003). However, as would be expected there is variability in the quality of professional relationships and communication, with caring and trustworthy professionals also being acknowledged (McMaster et al., 2004, Nuutila and Salanterä, 2006, Moore and Kordick, 2006, Konrad, 2008). Trust and acknowledgement of the family's expertise was identified within this synthesis as a descriptive theme relating to difficulties in decision making. The specific impact, in relation to the process of decision making, of trust and acknowledgement of parental expertise needs further investigation.

System factors have been highlighted within the chronic or life-threatening conditions literature in relation to incidents of medical errors (Sacchetti et al., 2000, Srivastava et al., 2005, National Reporting and Learning Service, 2009, Chua et al., 2010), rather than the impact that these factors have on decision making. This synthesis has identified that errors in hospital and problems accessing support and advice, impact on relationships with professionals and decision making. Further research is needed to identify how system factors are considered within decision making.

As can be seen from this discussion, the analytical themes identified within this synthesis have some commonalities with the wider literature on chronic and life-threatening conditions, however the manifestation of these themes appear to be different for those with chronic, life-limiting or life-threatening conditions. This synthesis has revealed the individuality of family preferences for engaging in decision making and future planning, but the included studies provide no indication of how professionals can best discuss and identify these or ensure that these are accommodated. Further research investigating how professionals might identify and meet family preferences for involvement is required. Additionally this synthesis has shown that future planning has not been a central focus within

the literature so far. Included studies focussed only on ACP, suggesting that the concept of future planning, more broadly, requires further investigation.

The themes resulting from this thematic synthesis are similar to those identified by Nicholson (2012), which is unsurprising given its weighting in terms of data for consideration in the synthesis. However, although Nicholson (2012) recognises the individual role of professionals and families, this is presented from the family's perspective only and there is little discussion of the interaction between them and its impact on decision making. The model of decision making proposed by Nicholson (2012) suggests that process and decisional factors are separate systems which rarely interact. This synthesis, however seems to suggest that professionals may act as a bridge between these two systems.

Limitations

As with any literature synthesis, the results are limited by the data reported in the included studies, which are necessarily a 'snapshot' of the data collected. However, the inclusion of individual study authors' findings in the synthesis (Thomas and Harden, 2008) aimed to mitigate this limitation, as the authors will have had an overview of the whole data set before analysing it into themes.

This synthesis aimed to provide insight into the process of decision making for children with life-limiting conditions. Papers investigating decision making for children with life-threatening conditions were excluded, although as highlighted in the discussion there may be some similarities and differences between these groups. As the wider literature enabling this comparison was not identified through a systematic search, it may be that there are more similarities between the groups and that excluding the studies investigating children with life-threatening conditions has prevented deeper insights into the process of decision making from being revealed.

Finally, the lack of multiple perspective research in this area and the dominance of the family or parent view in the included studies mean that little is known about the relational aspects of decision making. Additionally, the included studies were predominantly small and cross-sectional in nature and although two of the papers presented data from one longitudinal study, they only presented data from one time point, meaning that the dynamic process of decision making

and future planning could not be analysed. The evidence base in this area is limited and further research is needed.

2.4. Gaps in the literature

Despite the growing recognition that decision making is relational and temporal most of the research to date has been cross-sectional in nature and predominantly elicited a single perspective. The lack of research considering the multiple perspectives involved has resulted in a limited understanding of how the process of decision making occurs in context, how the various factors influencing individuals' perspectives are considered in relation to each other, and therefore the processes involved in reaching a decision. An investigation of decision making from multiple perspectives is required in order to identify how different opinions are shared and combined, and decisions negotiated.

The predominantly cross-sectional nature of the current literature has resulted in a perception that decision making occurs as a one off event. Some literature has alluded to the temporal nature of decision making and the evolving understandings which are present in the process of decision making. Lipstein and Britto (2015) conducted longitudinal research using interviews and observations for children with chronic illnesses without participant attrition, demonstrating the acceptability of this approach for further research. Prospective research investigating the process of decision making within the relational context and over time is required in order to fully appreciate the complexity of the process of decision making, specifically for children with life-limiting conditions. Such investigation should include observations of decision making interactions in order to capture the complexity of the process (Beecham et al., 2016).

Although a consideration of the wider chronic and life-threatening conditions literature has been useful in contextualising decision making within paediatrics, some of this may not be directly applicable to children with life-limiting conditions. Much of the literature has been conducted on the PICU with families of children who were otherwise well, presenting in an acute life-threatening episode, and therefore different challenges for decision making emerge. A number of authors have also suggested that differences in family experiences and decision making may exist between children with life-limiting and life-

threatening conditions (Berger, 2013, Fraser et al., 2014, Bužgová and Páleníková, 2015, Popejoy, 2015a, Inglin et al., 2011), further drawing doubt on any direct transferability of the wider literature to the life-limited population. Additionally, Price et al. (2012) have drawn attention to the lack of research within the non-malignant life-limited population, despite this being the largest group accessing children's palliative care and they therefore highlight the need to conduct such research. Therefore there is a clear rationale for additional research to better understand the complexity of decision making for this population.

Despite a growing recognition that decision making in paediatrics is a complex, relational endeavour, the methods which have been used to investigate decision making to date, mean that this complexity has not been fully illustrated. Longitudinal and multi-perspectival research is warranted in order to identify the extent to which each individual is involved in the process of decision making, how their preferences are combined, how the process evolves over time and how a decision reached. It should be conducted within the UK context in order to further illuminate the specifics of decision making in this cultural, legal and professional context.

Chapter 3: Methodology and Methods

3.1. Introduction

This chapter will provide an overview of the study aims and objectives, followed by the philosophical assumptions and methodological approaches underpinning it. It will then detail the study design, setting and methods used to carry out the investigation, followed by a discussion of the study procedures. The ethical issues encountered during the conduct of this study will then be considered, followed by an explanation of the data analysis and the chapter will conclude with a discussion of the procedures employed to ensure research credibility.

3.2. Study aims and objectives

The study aim was to explore decision making and future planning for children with life-limiting conditions, from a multi-stakeholder perspective. The specific objectives were to explore:

- The experiences and preferences of families, professionals and 'significant others' for engaging in decision making and future planning for children with life-limiting conditions.
- Communication between families, professionals and 'significant others' about decision making and future planning.
- The networks of care that exist for children with life-limiting conditions and how decisions are negotiated and distributed through these networks.

3.3. Methodology: Understanding the approach

3.3.1. Qualitative investigation

The processes of decision making and future planning have been identified to occur in a relational context and research investigating these phenomena therefore necessitates the use of an approach which can sufficiently capture this complexity. Qualitative research provides flexibility in relation to the available methods, which allow the complexity of the phenomena to be investigated within a small number of cases in great depth, rather than achieving a large sample size at a more superficial level of understanding (Miles and Huberman, 1994). The qualitative approach is more focussed on understanding phenomena, rather than explaining them with reference to cause and effect (Stake, 1995). Stake (1995)

asserts that for *"the qualitative scholar, the understanding of human experience is a matter of chronologies more than of causes and effects"* (p.39). Stake (2006) also identifies that in order to fully understand the context and setting that a phenomenon occurs within, the qualitative approach is required. This approach is also favoured in situations where little empirical evidence exists regarding the topic and as such an exploratory investigation is required (Smith et al., 2011). As has been demonstrated in the previous chapter, little is known about decision making for children with life-limiting conditions and therefore a qualitative exploratory investigation was an appropriate choice in order to enable a better understanding of the topic generally (Hesse-Biber and Leavy, 2011). Additionally, a generic qualitative approach facilitates a detailed focus on the research question, rather than moulding the research question to fit within a particular philosophical stance (Smith et al., 2011). The importance of this research question, as outlined in the literature review chapter and through my own clinical practice, suggested the need to retain the question as the driving force behind the choice of methodology, and not alter it to fit within a specific approach.

Hesse-Biber and Leavy (2011) discuss that qualitative research needs to be holistic and flexible, where methodological and methods decisions are made as a fluid process as opposed to a set of decision events in sequence. The flexibility of the approach for individual participants was also required, in order to make the research appropriate for each participant and reduce the burden of participation. Qualitative research is emergent in its design (Sandelowski, 2010) and allows the methodology to be revised, if required, as a result of unforeseen circumstances during the study (Hesse-Biber and Leavy, 2011). Design decisions therefore need to be flexible and guided by the nature of the research question (Morse, 2010, Thorne, 2011). Within this research, such flexibility was required in relation to accessing additional sites in order to follow up with participants and adding other interview participants within cases, as they presented themselves. Flexibility regarding the types of decisions focussed on was also demonstrated. Initially, the focus was to be on medical decisions, but it quickly became apparent that other social and educational decisions pose

significant issues for the recruited families and may shed additional light on the process of decision making; consequently the focus of the topic was broadened.

Qualitative research aims to obtain rich contextualised data, whereas closed-ended data collection instruments often prevent detailed data from being recorded (Miles and Huberman, 1994). In keeping with Miles and Huberman's (1994) assertion that structured instruments are unnecessary at the start of a study, this research began with broad questions, which were continually focussed throughout the study as a result of preliminary data analysis (Stake, 1995, Murphy et al., 1999) and resulted in the development of additional questions and foci for observations.

Qualitative approaches, specifically those utilising open ended interviews, have also been demonstrated to be acceptable and to some extent useful for families of children with life-threatening conditions. McGrath (2003) concluded from her research with families of children with cancer, that the *"process of qualitative research in longitudinal studies is actually a beneficial process that can make an important contribution to the lives of individuals going through a challenging health care experience"* (p.77). This provides further justification for use of the qualitative approach.

3.3.2. Philosophical orientation: interpretive constructionism

There are a number of paradigms within which research can be conducted, with three of the major paradigms being positivism, interpretivism and critical realism. Positivism is underpinned by realism and holds that knowledge can be gained about reality through systematic and careful experimentation. Such research is valuable in ascertaining the experiences of a larger number of individuals, once exploratory research has been conducted, for testing hypotheses and can be more readily generalizable. Interpretivism, in contrast, is underpinned by relativism, where the reality experienced by each individual is constructed based on their culture and experiences, and social constructionism is situated within the interpretive paradigm. The relativist epistemology denies that objective 'truth' or 'knowledge' can ever be gained, as no single shared reality exists. Critical realism partially closes the divide between positivism and interpretivism and argues that a universal reality does exist but that gaining

knowledge of it is difficult and subjective (Moses and Knutsen, 2012). Each paradigm provides valuable research insights and the choice of paradigm should be driven by the research question.

Given the lack of previous research about decision making for this population, a qualitative exploratory investigation was appropriate. The literature presented in the previous chapter has characterised the process of decision making as relative to culture and previous experiences and therefore a qualitative approach which acknowledges a relativist epistemology is required. Although critical realism could provide a paradigm for this investigation, the relative nature of decision making requires an investigation which is dedicated to understanding the phenomenon in the interactions between individuals at the micro-level (Cruickshank, 2012), which social constructionism is geared towards.

Constructionism is oriented towards the understanding of how life is interpreted and made meaningful by the people who live it (Berger and Luckmann, 1966) and challenges the traditional view that our observations of the world represent a single reality (Burr, 2015). Knowledge and understanding are relative, due to their situation within historical and cultural contexts and therefore any one way of understanding an event cannot be described as truth (Burr, 2015). Rubin and Rubin (2005) argue that the constructionist approach expects people to come to different conclusions about their experiences and acknowledges that *"multiple and even conflicting versions of the same event or object can be true at the same time"* (p.27). Social constructionism makes sense within a background of nursing, where we are taught that patients experience their symptoms differently and require individualised care, commensurate with their values and beliefs. Constructionism is therefore the lens through which I view the world and make sense of my own experiences, such that I can better come to understand the experiences of others.

Through the consideration and study of multiple perspectives regarding reality and knowledge, understandings of these interpretations become clearer (Berger and Luckmann, 1966). Constructionism also posits that realities are temporally and spatially situated and the events occurring within our area of influence shape our biographies. As socially situated and influenced beings, our experiences and

interactions continually shape us and our identities are therefore continually constructed (Goffman, 1990). Within interpretive constructionist theory, researchers may try to identify the experiences of different individuals and analyse these different accounts in order to achieve a better understanding (Rubin and Rubin, 2005). Social constructionists recognise that the positivist approach to data collection only permits simple responses, despite the complex answers that the question requires (Rubin and Rubin, 2005) and therefore favour a qualitative approach which enables the context (including history and culture), experience and accounts of people to be preserved as much as possible (Burr, 2015).

Decisions for children with life-limiting conditions are made in a relational context, taking account of a variety of perspectives. Conducting multi-perspectival research recognises this and is in keeping with the interpretive constructionist underpinning. The collection of data from a variety of participants, such as families, their significant others and professionals, will allow a richer understanding of the factors influencing decision making and future planning. The constructionist underpinning acknowledges that no one perspective is 'true' and that decision making is the outcome of negotiated understandings within networks of social relationships and action.

3.4. Study Design: Case Study

A case study design was chosen as the approach to conduct this research in order to gather data from multiple sources and analyse it within the complex contexts within which it was obtained. Stake (2005), argues that case study is not a methodology, but rather defines the phenomenon to be studied. The case study approach enables the investigation of a phenomenon in the complexity of its natural setting, from a variety of perspectives, thus providing a detailed understanding of the phenomenon (Thomas, 2011). Two types of case study exist and may be termed 'intrinsic' if the focus of interest is gaining a better understanding of the specific case or 'instrumental' if the interest is in gaining a better understanding of a phenomenon through understanding the case (Stake, 2005). The aim of this study is to explore the process of decision making for children with life-limiting conditions (the phenomenon) and therefore this research adopted the 'instrumental' approach. This research aimed to understand

the phenomenon across multiple cases and therefore the collective case study design (Stake, 1995) was utilised. The purpose of the multi case study is to understand the phenomenon, through reference to a number of cases, thereby detailing how the phenomenon operates in different contexts and settings (Stake, 2006). Within each case it will be important to uncover a variety of perspectives, not with the aim of identifying the 'true' perspective (such an aim does not exist within constructionism), but rather to identify what we can learn about the phenomena as a result of these divergent perspectives (Stake, 2006). The study aim of exploring decision making from a multi-stakeholder perspective necessitates the use of the case study approach, in which the multiple perspectives can be combined to provide a detailed understanding of the specific contexts in which decision making takes place (Swanborn, 2010).

In an instrumental case study, despite the interest in what the case can tell us about a phenomenon, Stake (2005) reminds us that there must be a detailed focus on the particulars of the case. It is only through gaining a detailed understanding of the case in its own right, that understanding of the phenomenon of interest is facilitated. He argues that too great a focus on cross-case generalisations early on in the process of analysis may result in overlooking particular and essential details of the individual cases, making the subsequent understandings generated, incomplete.

Case study research enables phenomena to be studied in their natural context, thus illuminating the social processes which normally occur within the case, to be observed and not disrupted. This also allows for the comparison of different contexts and their effects on the phenomenon (Swanborn, 2010). Studying the case naturally presents a *"unique opportunity to focus on social interactions and the developing meanings that participants in the system attach to each other, and how they interpret each other's acts. Another object of our attention is the existence of multiple realities: the different, and sometimes contrasting, views participants in a system have, and their diverging interpretations of events and conditions"* (Swanborn, 2010, p.16). This assertion further demonstrates the suitability of the case study approach to constructionist research and also its suitability for research within the complex context of children's palliative care.

3.4.1. Longitudinal design

The processes of decision making and future planning occur over time and are changeable. It was decided that in order to capture the evolution in these processes and any changes that occurred, a longitudinal design was required. Participants were followed for up to 12 months and numerous data collection activities occurred within this time. The longitudinal nature of this research enabled the chronology of the child's engagement with health and social care organisations to be observed and thus a more detailed understanding of the processes of decision making and future planning obtained. The longitudinal design also facilitated the development of a good research relationship and rapport with participants, which has been suggested to aid the discussion of sensitive topics over time (Murray et al., 2009, Carduff et al., 2015), something which occurred in this research. In second and third interviews, families appeared to feel more able to disclose aspects of their decision making and future planning that they perhaps did not feel comfortable to discuss earlier in the study period.

3.4.2. Multiple perspectives

As has been demonstrated within the previous chapter, research to date has investigated decision making from a single perspective, or included a variety of perspectives, but not within a case format. Such studies have neglected to illuminate the complex contexts of individual cases and therefore discussed decision making as a concept more broadly. It was identified that decision making takes place within a relational context and therefore obtaining multiple perspectives would assist in providing a better understanding of the phenomenon, something which Stake (2006) acknowledges case study research is appropriate for. Uncovering multiple perspectives is therefore commensurate with the study design, the underpinning philosophy and is relevant to the topic, which necessarily requires multiple stakeholders to make and implement decisions. Additionally, Kendall et al. (2009) argue that obtaining multiple perspectives in health research is beneficial in understanding the relationships and dynamics involved, exploring similarities and differences in perspectives and understanding the needs of each individual. Within this study multiple perspectives were accessed wherever possible.

3.5. Study Setting

The research was carried out in four large NHS Acute Care Trusts within England. Two sites were initially chosen based on their location and my professional relationships with the clinical teams. Subsequent to this, a third site was added to increase recruitment and was chosen based on the size of the hospital and breadth of specialities available, and within an acceptable distance to travel. Subsequent to adding the third site and a further drive for recruitment at one of the initial sites, the recruitment of additional cases with sufficient complexity enabled recruitment to cease. A fourth site was added as a follow-up site, rather than a patient identification site, in order to observe episodes of care for one of the participants at which important decisions would be made.

Data collection took place in various locations within the study sites, such as wards and out-patient departments, professional offices, family homes and in a number of schools[6].

3.6. Data collection

3.6.1. Observation

The study of decision making requires attention to the context in which it occurs and therefore necessitates the use of self-report practices and observation (Carnevale, 1997). Observation was therefore employed within this study, which enabled immersion into the setting, facilitating an understanding of the *"complexities and shared cultural nuances"* (Jones and Smith, 2017, p.98) of the context and therefore a detailed consideration of the process of decision making. Observation enables an understanding of the shared meanings of those involved and the environmental and relational context within which decision making occurs. However, it is important to recognise that what is observed and recorded will be determined by the culture, background and previous experiences of the observer. The product of the observation is therefore co-produced between the participants being observed and the researcher observing them (Carnevale et al., 2008, Mulhall, 2003).

[6] Observations took place in schools when Team Around the Child or therapy meetings occurred there, as these involved medical and social care professionals making decisions regarding the child's care. Medical reviews conducted within schools were also attended.

Observations of clinic appointments, ward rounds, Team Around the Child meetings and a Clinical Ethics Committee meeting were undertaken. Unstructured observation was utilised as it is in keeping with the constructionist underpinning of the study, whereby context is seen as important and understanding is co-constructed within social interactions (McKechnie, 2008). Unstructured observation does not imply that the activity was haphazard or lacked rigour, rather that there was no checklist used to search for predetermined notions of what may occur in the observational setting (Mulhall, 2003). Specific aspects which were looked for were: the individuals present and their influence on others, apparent feelings and emotions of individuals, activities carried out, information seeking and provision, and the impact of time. Pertinent information, including the content of discussions and non-verbal communication, were abbreviated and written in a notebook, and any relevant quotations were written down verbatim. Immediately following the observation, field notes were typed up and expanded. Although I tried to be open to what was being observed, I was continually seeking any evidence to suggest that decision making was occurring, however hidden or unspoken it may have been.

3.6.2. Serial interviews

The interviews were guided by the Responsive Interviewing approach as outlined by Rubin and Rubin (2005). They argue that the purpose of this type of interviewing is to obtain a deep understanding of the topic and it was therefore considered an appropriate choice of method for the research question. They define five characteristics of Responsive Interviewing:

1. The purpose is to gather participants' interpretations and understandings of their experiences.
2. Interviews are conducted within conversational partnerships and therefore the personality and beliefs of the researcher are important and should be at the forefront of their mind.
3. Responsive Interviewers have significant ethical obligations to their participants.
4. Researchers should ask broad questions to avoid limiting participants' responses and imposing their own views on the interview.
5. The approach requires flexibility and adaptation in order to follow up on

new insights provided within the interview (Rubin and Rubin, 2005).

These characteristics assisted with guiding the research design and were employed within interviews. Rubin and Rubin (2005) suggest that for serial interviews, it may be appropriate to only ask non-threatening questions in the first encounter. Therefore a topic guide (Appendix 4) was used in initial interviews as a prompt for discussion and although the interviews were largely unstructured to begin with, there were a number of main questions, which provided the *"scaffolding of the interview"* (Rubin and Rubin, 2005, p.134). The broad opening questions enabled participants to discuss issues which they felt comfortable sharing and which were pertinent to their experiences. This provided the participant with more control to discuss issues related to the topic that they felt were pertinent (Corbin and Morse, 2003). Follow-up questions were then asked in order to explore further the answers participants gave to the main questions.

Prior to follow-up interviews, initial transcripts were revised and follow-up questions to participants' responses in the initial interviews were identified as questions to be asked. Therefore subsequent interviews were able to build on topics of discussion from previous interviews, in a process of ongoing conversation (Davies et al., 2009, McGrath, 2003, Campbell and Lassiter, 2014) and progressive focussing (Stake, 1995, Murphy et al., 1999). The interview questions were also guided by the ongoing observations and steered towards discussion of the topics of interest (Roulston, 2010): decision making and future planning. Serial interviews allowed for the establishment of a relationship and rapport between myself and the participants, such that more sensitive disclosures were made once the participants felt they were able to trust me (Hunter Revell, 2013). The final interview also provided an opportunity for participants to provide feedback about their participation in the research and helped to mark the end of data collection, as part of the exit strategy.

The study aimed to achieve approximately three interviews with each participant during the 12 month study period. The number and frequency of interviews was flexible and negotiated with the participants on an individual basis, providing the option to discuss events within an interview setting at an appropriate time.

More or less frequent contact with each participant occurred as appropriate and in keeping with their desired level of engagement. Arranging interviews with the significant others, particularly for the professional participants took some time. This was due to the difficulties in making contact initially and also due their clinical commitments and a struggle to find the time. Given the difficulty in arranging initial interviews with the professional participants, it was decided that these participants would only be interviewed twice, once at the beginning and once at the end of the study.

Interviews were primarily conducted in family homes for the family participants and in professional offices for the professional participants. There were a few exceptions to this: three family interviews and two professional interviews were conducted over the telephone and one professional interview was conducted in their home. This was important to ensure flexibility to meet the needs and schedule of the participant.

Family participants were given the option (if more than one family member was participating) of having interviews together or alone. Predominantly interviews were conducted on an individual basis. It is important to note that the presence of additional people in the interview may alter the content of the discussion as a result of the social interaction between the participants (Morgan et al., 2016, Eisikovits and Koren, 2010). However, given the constructionist underpinning of this study, the difference in data gained from such interviews is not considered better or worse, rather recognised as being constructed differently given the social interaction between the participants (Morris, 2001, Polak and Green, 2016). Only one family chose to have joint interviews, yet two other families had joint interviews which resulted from additional family members entering the room and joining in the discussion. In such instances the participants were consulted to ensure that they were happy to proceed with a joint interview.

3.6.3. Notes review

Review and analysis of the patients' medical notes also formed part of the data collection. The use of documents in constructionist research is not aimed at uncovering the facts of a situation, rather it provides additional data regarding individuals' interpretations (Rubin and Rubin, 2011). Utilising documents in

addition to interviews and observations enables comparison between the different sources in order to highlight where the accounts are consistent or where they contradict each other, and a consideration of how such inconsistencies can be explained (Rubin and Rubin, 2011). It is important when utilising documents within constructionist research to acknowledge who produced them, under what constraints and for what purpose. This acknowledgement helps to overcome some of the methodological challenges, such as any biases which influenced the document's production and therefore determined what was included, omitted or erroneously recorded (Holstein and Gubrium, 2008). Within this study, medical records were recognised as being produced by professionals, with the primary intended audience being professionals, and often written in a time constrained environment with specific requirements regarding what needed documenting.

The patient's medical records were accessed (with permission) after all other data collection was completed within the case. This enabled the identification of any additional periods of discussion or decision making and provided some account of interactions which occurred prior to the study or that I was unable to attend during the study period, as well as providing another perspective on any interactions at which I was present. All records were accessed in either paper or electronic form on NHS Property. Medical notes were reviewed for content relating to decisions that had been made. Records extracted included nursing notes, medical notes, clinic letters and care plans. Data were fully anonymised before extraction and transfer out of the organisation. Reviewing medical notes helped to illuminate the extent to which professionals documented their discussions and their perceptions about the level of family engagement.

3.6.4. Case Structure

Each case was centred around one child and family. Families were invited to nominate 'significant others' who they considered important in their decision making to also participate in the study (Figure 5**Error! Reference source not found.**). It was hoped that 'significant others' would include at least one health professional, in order to gain an understanding of the process of decision making from a professional perspective, but this was not achieved in all cases. The boundary of each case, in relation to the interview component, was determined

by the child and family based on who they nominated as their 'significant others', within the limits of 5 participants per case.

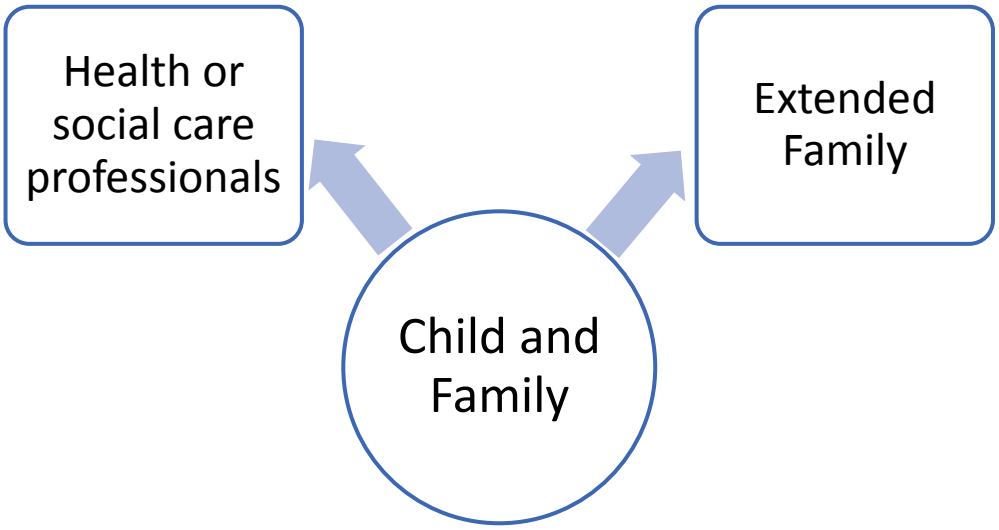


Figure 5: Case Structure

The actual boundary of each case was much wider and almost impossible to define, given the vast number of professionals and significant others present at observations and who often influenced the decision making from afar. The minimum requirements for a case to be included initially was that there was one participant and at least two data sources, for example, serial interviews or more than one method of data collection. The minimal requirements for a case reflected the individual nature of family contexts and the ability to accommodate family preferences for participation.

The iterative and evolving nature of qualitative research prompted a reconsideration of the minimal requirements for a case to be recruited. Following invitation to take part in the study, one family contacted me declining to participate in the full study, but offering to take part in a single interview, which they highlighted *"could help provide powerful context to your study"*. Stake (2005) highlights that the opportunity to learn is often the most important aspect in sampling decisions and as a result, the decision was made to revise the requirements of a case and include this case in the study.

3.7. Study procedures

3.7.1. Ethics and governance approvals

Governance approvals were required in order to conduct this research. In accordance with the HRA guidelines for research conducted within the NHS, an application to an NHS Research Ethics Committee, via the Integrated Research Application System was made. The research was granted Favourable Opinion by the Nottingham 1 Research Ethics Committee in September 2015 (Ref: 15/EM/0377, see Appendix 2), in addition, applications to the Research and Development departments of two NHS organisations were made and both approvals were granted in October 2015. Agreement from the relevant Heads of Service at both Trusts was also obtained prior to commencing the research.

The addition of a third site was necessary due to recruitment difficulties and governance approvals for this site were granted in January 2016. One further site was added as a follow-up site. This required registration with the HRA for the study to come in line with the new research processes. HRA approval was granted and governance approval for the fourth site was granted in July 2016 (Appendix 2).

3.7.2. Sampling

A maximum of 15 cases was considered appropriate to meet the study aims and is in keeping with case study literature (Stake, 2006). Stake (2006) argues that more than this may yield too much data for the researcher to comprehend. Additionally, Morse (2000) argues that the larger amount of useable data collected from each participant, the fewer the number of participants required. As this research aimed to collect significant amounts of data from each case, it followed that the number of cases would be small.

Purposive sampling was utilised for this research, whereby strategic choices were made about what or who to study in relation to the study objectives (Palys, 2008). Purposive sampling is typical for qualitative research where the aim is to achieve in depth understanding about a topic within a small number of cases, rather than gain statistical significance through studying large numbers of cases outside of their context (Miles and Huberman, 1994). Within multiple case study

research, purposive sampling is also related to choosing the case which offers the best opportunity to learn, such as the most accessible case (Stake, 2005).

The case study design employed within this research required within case sampling. This involved a 'snowball sampling' approach, whereby informants within the case recommend others who may be appropriate to study. Again the primary concern with this type of sampling is opportunity to learn (Stake, 2005) and the 'snowball' approach enabled the identification of individuals who would present the best opportunity to learn, albeit from the perspective of the child and family. Additionally within-case sampling is theoretically driven by the research question, not a concern for representativeness (Miles and Huberman, 1994), therefore making the 'snowball sampling' approach appropriate. Within the current study the opportunity to learn about decision making was determined by the parent and child. Their nomination of significant others (or not) provided information about their view on decision making and the extent to which they believed they were assisted in making decisions. They knew who they involved in their decision making and which individuals were consulted about decisions; these individuals presented the best opportunity to learn and were therefore included in the case.

Eligibility Criteria

The eligibility criteria for each type of participant are shown in Figure 6.

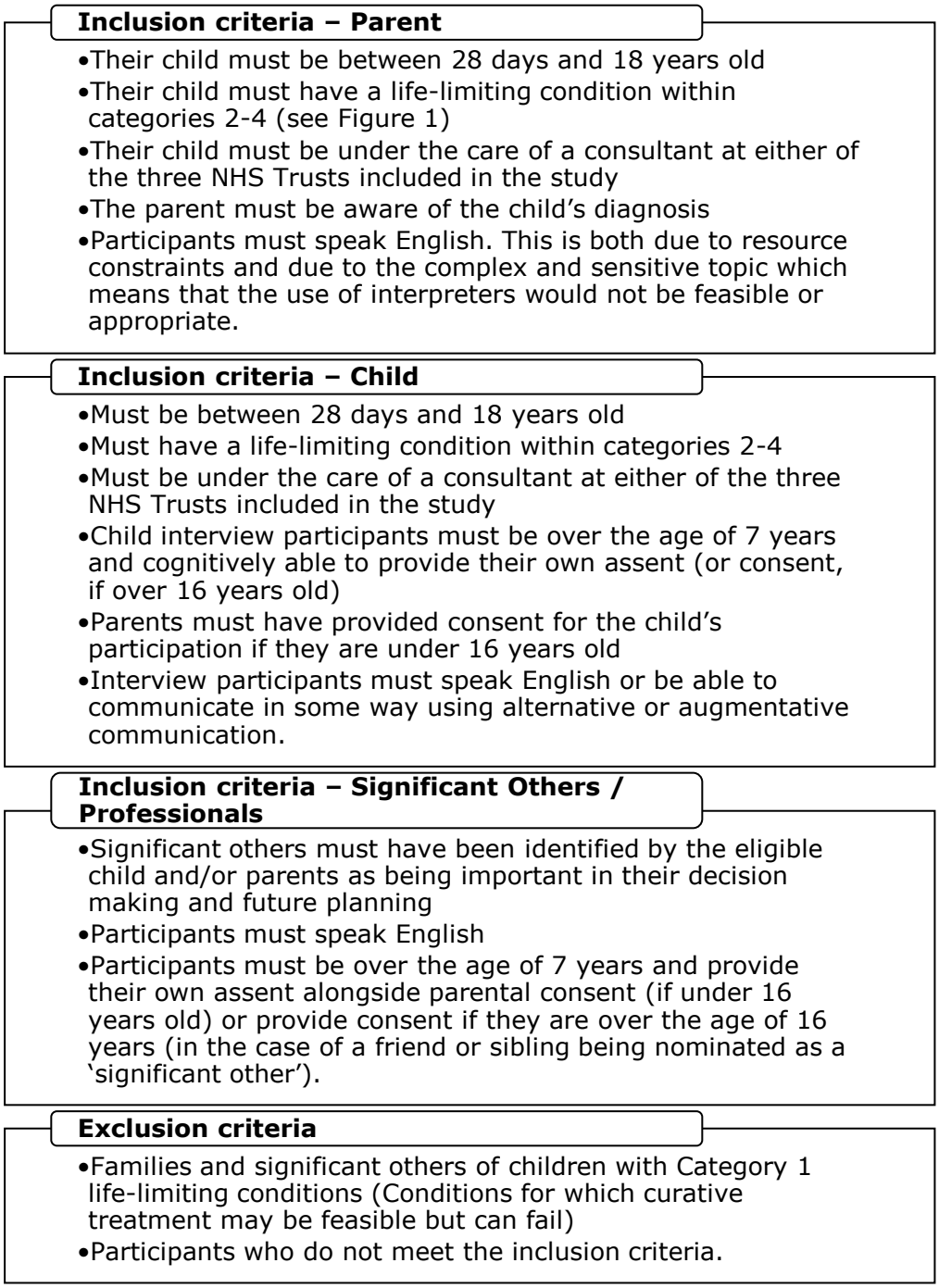


Figure 6: Inclusion and exclusion criteria

Complexity of the child as a sampling decision

The primary interest of the study was regarding decision making and future planning and it was presumed that recruiting families of children with

particularly complex care needs would enable a more in depth understanding of decision making, as more decisions would be likely to be needed to be made within the study period. It became clear that important decisions for the family were not confined to the realms of medical decision making and that other decisions surrounding housing, education and the wider family may be incredibly significant for the family. Predominantly families of children with more complex care needs were recruited, but enabling the inclusion of children at different stages of their illness facilitated an overview of the different levels of complexity that exist in decision making for families over the course of children's lives and their evolving disease trajectory.

3.7.3. Screening and recruitment

Consultants covering a range of specialities in the relevant fields at each of the three initial participating NHS Trusts were approached and the research presented to them. They were given information about the study, including a copy of the eligibility criteria and were then asked to invite eligible families to participate in the study. The invitation to participate in the study was made by the child's main consultant or a member of their clinical care team, who provided them with an information pack[7]. In sensitive health research, it is recognised that recruitment is often dependent on health professionals who introduce a degree of gatekeeping (Crocker et al., 2015). It is not possible to identify what other criteria they applied, additional to the eligibility criteria, to decide who would be (and who would not be) appropriate to invite to participate.

Most consultants screened and invited families without my assistance, but one consultant wanted to help in capturing a diversity of cases and invited me to assist her in deciding which families to invite to participate. In all cases, including the consultant who screened potential participants with my help, it was not possible to identify whether there were any particular additional criteria that

[7] In total 116 information packs were distributed to consultants and their clinical teams. A minimum of 32 information packs were sent to families and this number is known as some professionals informed me of the families they had invited. Other professionals did not provide such feedback and it is therefore not possible to know how many of the other 84 information packs were sent out to families.

were applied to potential participants. However, it appeared that for the consultant who I assisted with screening, general reference to the family's current emotional state or coping was made when she decided not to invite a family.

An information pack which included a cover letter from the child's main consultant was sent to the family. If the family wished to participate, they returned the reply slip to me in the stamp addressed envelope (included in the information pack), consenting for me to contact them. I contacted the family member and further explained the research to them. All families that were able to be contacted following their return of the reply slip, agreed to meeting me for an interview and written consent was gained at this encounter, in advance of the interview. Two families who returned a reply slip, were uncontactable despite a number of attempts.

Significant others were nominated at the first interview with the family and were contacted directly and invited to participate. Written consent for these participants was obtained in advance of their first interview.

Data were collected from 11 cases, within which 24 individual participants were interviewed up to three times and many more were observed. Serial interviews and/or multiple observations were achieved in 10 of the cases and a single interview obtained in the 11th case.

3.7.4. Practicalities of participant engagement

At the initial family interview, participants were asked for details of their upcoming appointments and provided their consent for me to access their child's medical records in order to identify any other scheduled appointments. The electronic appointment's system for the three sites that were not my employing organisation were not able to be accessed, so appointments occurring at these sites were only able to be identified through contact with the family or the professionals caring for them. The longitudinal nature of this study and the changeability of the children's conditions meant that it was incredibly difficult, to keep up to date with and attend all medical interactions. Wherever possible, appointments, hospital admissions and other scheduled meetings, were attended. Some emergency appointments or admissions that participants forgot to inform

me of, were not attended. Additionally appointments which occurred at other sites where governance approvals had not been gained were not attended.

Identifying the appropriate level of contact for each family throughout the study was required. Some families had fewer appointments and so it felt appropriate to contact them less frequently, as there would be less to discuss between contacts and less risk of missing out on scheduled appointments. Conversely, other families had such a high level of medical intervention and almost daily appointments with health or social care professionals, that for these families, it felt appropriate to minimise the contact and further intrusion into their family life. Most families were contacted via telephone, email or text on a monthly or six weekly basis, but sometimes more regularly, depending on the timing of appointments. It seemed appropriate to telephone them between appointments, as a way of maintaining contact with the families and ensuring that the research relationship was sustained. It demonstrated my interest in their family and the non-medical aspects of their lives, such as how the child was getting on in their transition to secondary school. The method of contact was largely determined by the family and this started out with telephone calls, but it was identified that some families preferred text contact, so this was adopted in such cases. Prior to each appointment, contact was made with the family to check that they were still happy for me to attend and also, whenever possible, contact was made with the professional who the appointment was with, to ensure that they were also happy for me to be present and observe the consultation.

3.8. Ethical issues and data collection challenges

3.8.1. Consent and assent

Prior to obtaining informed consent, I explained the details of the study and provided a Participant Information Sheet (Appendix 6), ensuring that participants had sufficient time to consider participating. Children's Participant Information Sheets were designed to be engaging and ensure that the necessary information was communicated in an age appropriate manner (Twycross and Smith, 2017). Possible participants were then provided with the opportunity to ask any questions concerning study participation. Adult participants provided written informed consent at the start of the study and subsequent process consent was then employed. Within qualitative research, it has been acknowledged that

one single event of obtaining consent is not sufficient, but rather an ongoing process of *"consensual decision-making"* (Ramos, 1989, cited in Usher and Arthur, 1998, p.695) and continual awareness of the participant's motivation and willingness to continue to participate in the study is required.

Modi et al. (2014) highlight the need for process consent, especially in longitudinal children's research, due to the child's evolving legal status and capacity to make decisions. They also stress that written consent should be documented prior to enrolling in the study and this should be confirmed with the participant prior to subsequent data collection activities, although the confirmation may be verbal. Explicit consent to ongoing participation was periodically confirmed throughout the course of normal interaction at each stage of data collection. I was particularly sensitive to behavioural cues from children which indicated that they wished to disengage from data collection activities, something which Coad et al. (2015) highlight as essential for research with children. One family withdrew from the study after three observations, prior to arranging an interview, highlighting that the process of consensual decision making was sufficient that participants felt able to withdraw. No information was given or collected regarding the reason for this case withdrawing.

Children who possessed sufficient cognitive understanding were required to provide their consent or assent to participate, and their parents' written agreement was also required (for those under 16 years old). These children were also asked to provide their consent or assent to participate in interviews. Most children are able to provide their assent from the age of 7 years (Modi et al., 2014) and therefore children under this age were not invited to take part in interviews. For those children who were able to understand and provide consent or assent, written or verbal confirmation was obtained, as deemed most appropriate. Ongoing assessment of the child's understanding of their participation, as outlined by Twycross and Smith (2017), was undertaken to ensure that their consent or assent remained valid. There were no cases where conflict occurred between the child and parents regarding study participation.

In cases when it was known which professionals would be present at observations, they were contacted in advance to request permission for me to be

present and take field notes about the interaction. However, it was not always possible to anticipate who would be present. In cases where it was not possible to gain consent in advance, participants were briefly informed about the purpose of the research and gave their verbal consent. Such an approach has been highlighted by Carnevale et al. (2008) who state that when written consent is not possible to obtain, *"the level of consent requirement should be proportional to the level of personal disclosure and potential obtrusion anticipated for the study"* (p.27). The verbal consent obtained during such interactions was deemed to be proportional.

3.8.2. Confidentiality and anonymity

Confidentiality was another issue encountered within this study, especially as families nominated significant others to participate in the research. It was important for me and the participants to be aware that even though the issues discussed in one interview are relevant to the case as a whole, the information collected within that interview would be kept confidential and therefore not shared with the other members of the case. As the study included a small number of cases, it was difficult to fully anonymise and decontextualize the results, but where appropriate, attempts to do this have been made.

Another issue regarding confidentiality was the inclusion of children as participants. Children were able to request to have interviews separately from their parents and may wish to know that what they have said will not be fed back to their parents (Huang et al., 2014, Duncan et al., 2009). However, children were made aware that I am bound by a duty of care and would be required to share information if I felt that they were at risk of harm. Separate child interviews occurred only for Michael's case and was facilitated by his mother, Kate, as she felt that a non-threatening interaction between Michael and I, whilst playing video games, would elicit more insight into his thoughts and feelings. The door to the room where Michael and I were playing games remained open, whilst Kate did housework and occasionally brought us some refreshments. Michael appeared unconcerned about his mother's 'interruptions' and, as we had previously had discussions about his thoughts together as a group, seemed to be happy for his mother to know what he was sharing with me. Nonetheless, I felt conflicted when Kate asked what we had discussed, as I was conscious about the

need to maintain Michael's confidentiality, but also trust within the relationship with Kate. I therefore discussed with her the broad topics that we had touched on, which I knew that she had heard when bringing us refreshments. This highlighted the difficult nature of confidentiality in research involving children and the need for a delicate balance between confidentiality and maintaining trusting relationships.

3.8.3. The researcher-participant relationship

During the initial stages of the research, a rapport was built with the participants through telephone calls, interviews and contact at observations. This was an essential part of the research process and assisted in gaining access to each individuals' story (Dickson-Swift et al., 2007, Huang et al., 2014). These relationships were subsequently built on over the study duration. The longitudinal nature of the study, requiring my journeying alongside the families whilst they made difficult decisions, therefore posed challenges regarding relationship boundaries and termination. Over time, the participants and I developed a bond, which was felt more strongly in some cases than others, as a result of the sensitive stories they shared and my intensive involvement in their lives, something which has been acknowledged by Sterling and Peterson (2005). It was important from the outset of the research to be clear about the timescale of contact that I would have with participants and I reminded them of this as the study came to an end (van Wijk, 2014). However, this was something that I only truly appreciated once these relationships had begun to develop and I realised my own emotional ties to the families. This realisation highlighted the need to begin discussions regarding termination of relationships and my exit strategy. Towards the end of the study, the reminders of the number of contact points prior to the end of the study served as much as a reminder for me to prepare myself for the end of data collection and termination of the relationships, as it did for the participants. Given the intensity of the contact in some cases, I anticipated that leaving the field would result in a feeling that I had abandoned the families. During the final interview as part of the exit strategy, there was the opportunity for participants to provide feedback about their experience of participating in the research and the offer of providing the participants feedback, once the data had been analysed and findings written. These strategies served as a way to mark the

end of the data collection and confirm the termination of the research relationship.

3.8.4. Role boundaries

The information sheets for this study detailed my status as a paediatric nurse, an amendment that the Research Ethics Committee had requested during the ethical approvals process. I was open and honest with participants about my background as a paediatric nurse and my continued clinical contract. The reasons for this openness were twofold: firstly, there existed a real possibility that some of the children would be admitted to my ward during the course of the study which, had I kept my role from them at the outset could have caused trust issues, if they found out during an admission. Secondly, being a nurse is a core aspect of my identity and is the reason for my interest in this topic area. Therefore if I did not reveal my true identity I would struggle to communicate with families about who I am and why I embarked on this research, and would feel that I were deceiving them. As Rubin and Rubin (2005) argue, researchers have ethical responsibilities to those they interview and that *"respecting interviewees normally means not deceiving them, not pretending to be someone you are not"* (p.97).

Given my clinical role as a paediatric nurse, it was a possibility that families would ask for my clinical advice throughout the study. At the initial interview, and indeed at all interviews, I was clear to discuss with the participant that I was present as a researcher and would not give any advice or opinion about the course of action that they were deciding upon, explaining that doing so may influence their interaction with me and the study findings. Only two incidents occurred throughout the study period where I felt true conflict between my role as nurse and researcher. The first incident occurred when a mother telephoned me and asked about obtaining feed supplies for her child. She seemed to realise that I wasn't the correct person to ask, but that I may be able to help, given my nursing role. I found the telephone number for the appropriate department and advised the mother to contact them for further assistance. The second incident involved a mother asking for my opinion about the decision that she should make. This highlighted my role as nurse and researcher, and further illuminated that despite an attempt to be an observer rather than participant, my presence in the

participants' lives meant that this separation was impossible. Further discussion of these role boundary issues is provided in section 8.9.

An additional issue that arose in relation to my role was when one child was admitted to my ward during the study period. I had seen the family a few days before and explained that when I was going to be on shift, I would not be looking after the child, because I felt that it may cause further role confusion (for both myself and the family) and represent a conflict of interest. In this case the family understood and agreed that this was appropriate.

Role boundary issues also arose in relation to professionals. Prior to observations I contacted the professional who was due to see the child and obtained consent to conduct the observation. This contact did not discuss my role as a nurse, rather focussed on the study purpose and methods. The reason for this was because I did not wish to draw attention to myself as a nurse in appointments, something that at times did happen when in appointments with professionals who knew me professionally. Additionally, I did not wish to influence the way in which professionals conducted their appointments, which may have occurred if they knew that a nurse was present. During some observations with professionals who did not know me well professionally, but perhaps recognised me, it was clear that they were slightly confused about who I was. This highlighted the conflict between my desire to remain as invisible as possible within observations and the need to provide all relevant information to participants.

3.8.5. Reflexivity

In recent years there has been a growing interest in reflexivity in qualitative research and significant academic outputs regarding the topic. Etherington (2007) describes reflexivity as:

> *"A tool whereby we can include our "selves" at any stage, making transparent the values and beliefs we hold that almost certainly influence the research process and its outcomes. Reflexive research encourages us to display in our writing/conversations the interactions between our selves and our participants from our first point of contact until we end those relationships, so that our work*

> *can be understood, not only in terms of what we have discovered, but how we have discovered it"* (p.601).

This explanation of reflexivity demonstrates the need to consider the influence of the researcher on the whole research process, something that was facilitated through the use of reflective diary entries following all data collection episodes. Rubin and Rubin (2005) highlight the importance of addressing any biases or preconceptions which may influence the research process and take steps necessary to reduce, or explicitly address, these wherever possible. Practicing this self-awareness and reflexivity will result in more transparent research. Situations such as those discussed in relation to role boundaries also are in need of reflexive contemplation in order to identify my potential impact on the data, in these specific situations.

The positionality of the researcher in relation to their role as an 'insider' or 'outsider' has been extensively discussed in the literature. Insider research implies that the researcher belongs to the group that they are investigating and outsider research implies that the researcher is not a member of the group they are researching (Dwyer and Buckle, 2009). Insider research has been argued to provide benefits in relation to trust and acceptance of the researcher, whereas outsiders may have 'fresh eyes' for looking at the phenomenon (Bonner and Tohurst, 2002). Dwyer and Buckle (2009) argue that the traditional conceptualisation of positionality as being dichotomous is flawed and that in reality, the researcher's role will be on a continuum from insider to outsider. Similarly Davies (2008) acknowledges that all researchers are to some degree connected to the research topic. Within this research, I occupied different spaces on the insider-outsider continuum, depending on the data collection activity and participants involved. As I was to some degree connected to the topic at all times, reflexivity was of vital importance in order to identify the impact of my role on the data and its interpretation (Davies, 2008, Bonner and Tohurst, 2002, Pellatt, 2003).

3.9. Data Analysis

3.9.1. Data Management

The Data Protection Act (1998) was adhered to when managing study data. Interviews were audio recorded with the participants' permission. These data files were transferred to secure servers via University of Nottingham computers and deleted from the recording device as soon as possible after data collection. I transcribed all interview recordings verbatim and subsequently checked for accuracy by listening to the audio file and reading the transcription. All identifiable information was removed from transcripts. As identified by Ochs (1979), transcription is necessarily selective and certain aspects of the interaction are filtered out, resulting in a more useful, albeit partial transcript. Transcription focussed on the language used and any non-verbal communication which appeared relevant to what was being said. Verbal fillers were omitted in order to make the transcript more readable. Field notes were taken during the observations and detailed notes made immediately after observations. These were stored in locked cupboards in locked rooms on University premises. As with the interview transcripts, it is important to note the partiality of field notes and as suggested by Clandinin and Connelly (2000), a reflexive journal was kept in order to identify my position in relation to the data recorded. The participant's medical records were accessed after their final period of data collection and relevant sections of the notes were copied, anonymised and stored in an encrypted space on a University of Nottingham computer. Analysis of anonymised data was conducted by hand and all printed study data were kept securely in a locked cabinet in a locked University office.

3.9.2. Narrative Analysis

Data were analysed using Narrative analysis. Narrative analysis has been recognised as a way of coming to understand others' experiences, through collaboration between researcher and participants and through *"living and telling, reliving and retelling, the stories of the experiences that made up people's lives, both individual and social"* (Clandinin and Connelly, 2000, p.20). Narrative inquiry aims not only to identify individuals' experiences, but to understand these within the wider social and cultural narratives within which they are formed, through observation, journeying alongside and speaking with

individuals and through writing and interpretation (Clandinin and Rosiek, 2007). This research aims to explore decision making from a multi-stakeholder perspective and therefore narrative analysis is an appropriate choice, given its focus on individuals' experiences in relation to their wider social and cultural context and attention to re-storying over time. Narrative analysis also recognises that the story is told and re-told through the research process and that the subsequent analysis produced has been co-constructed in a process of collaboration with researcher and participants (Holloway and Freshwater, 2007). Narrative analysis is commensurate with the constructionist underpinning of the study and as such, does not seek to identify a single 'truth' or the historical facts involved in the experiences of participants, rather it seeks to identify how they construct meaning from their experiences through telling stories, striving for a coherence theory of truth, rather than for a correspondence theory of truth (Sandelowski, 1991). As Riessman (2005) puts it, *"narratives do not mirror, they refract the past"* (p.6). Holloway and Freshwater (2007) outline that multiple and conflicting narratives can be brought together in order to present a meta-analysis, rather than focussing on a single perspective, such as intended within this study. Additionally narrative inquiry recognises the temporal and changeable nature of narrative truth (Sandelowski, 1991) and is therefore highly appropriate for application within a longitudinal investigation such as this.

Although other types of analysis could have been used on this data, the specific aim of the study, to understand the process of decision making, required attention to the temporal and relational context within which this occurred. The narrative approach enabled the context of accounts to be preserved and specific themes analysed within this context, rather than breaking the accounts up into specific codes as would have been the case with Framework Analysis, where data are extracted from their initial context (Richie and Spencer, 1994) or Grounded Theory, where data are fractured and become abstracted from their context (Holton, 2007). As with all methods, pitfalls also exist within narrative analysis and the possibility of privileging certain voices (Gottlieb and Lasser, 2001) exists perhaps more so than in other methods of analysis. Attention to this possible pitfall was paid throughout the process of analysis and the strategies

that were used to reduce this risk are discussed in relation to the research credibility (page 102).

Within the field of narrative analysis, there are a variety of approaches available to the researcher. The form of narrative analysis that was employed within this study was the interactional, or three-dimensional space, approach (Riessman, 2005, Clandinin and Connelly, 2000, Ollerenshaw and Creswell, 2002). This specific approach acknowledges the personal and social aspects of experience and the contexts within which individuals' experiences occur, focussing on the interactions, temporality and situation or environment (Ollerenshaw and Creswell, 2002). It is highlighted that these aspects are considered through the personal practical and professional knowledge of those involved in the narrative (Ollerenshaw and Creswell, 2002) and the value of the researcher disclosing aspects of their own stories to contextualise their analytical decisions (Clandinin and Connelly, 2000), which has become known as reflexivity.

Data analysis initially occurred within case; all of the data for each case was compiled and analysed in accordance with the three-dimensional space approach outlined by Clandinin and Connelly (2000). Each case was treated as a separate study during this initial phase of analysis, where correspondence and patterns were sought (Stake, 2005), using triangulation of data sources to contextualise each case. Subsequent cross-case analysis was undertaken, with the aim *"to ensure that we have the picture as clear and suitably meaningful as we can get it"* (Stake, 2006, p.77).

Process of analysis

As outlined by Clandinin and Connelly (2000), the process of analysis began with continual familiarisation and immersion within the data and an understanding of how different data sources fit together. A summarised narrative of each case was then compiled from the case specific data, through attention to the types of decisions made, the strategies used and any pertinent contextual details, in a process of restorying. The resultant case narratives were then imported into NVivo 11 for cross case analysis and underwent a process of coding, where key concepts were coded. These concepts, once identified were interrogated and either merged or expanded upon, depending upon their content,

thus refining the hierarchy of codes and themes. Once all case analyses were coded within NVivo, a cross-case analysis was facilitated. Figure 7 depicts the process of data analysis.

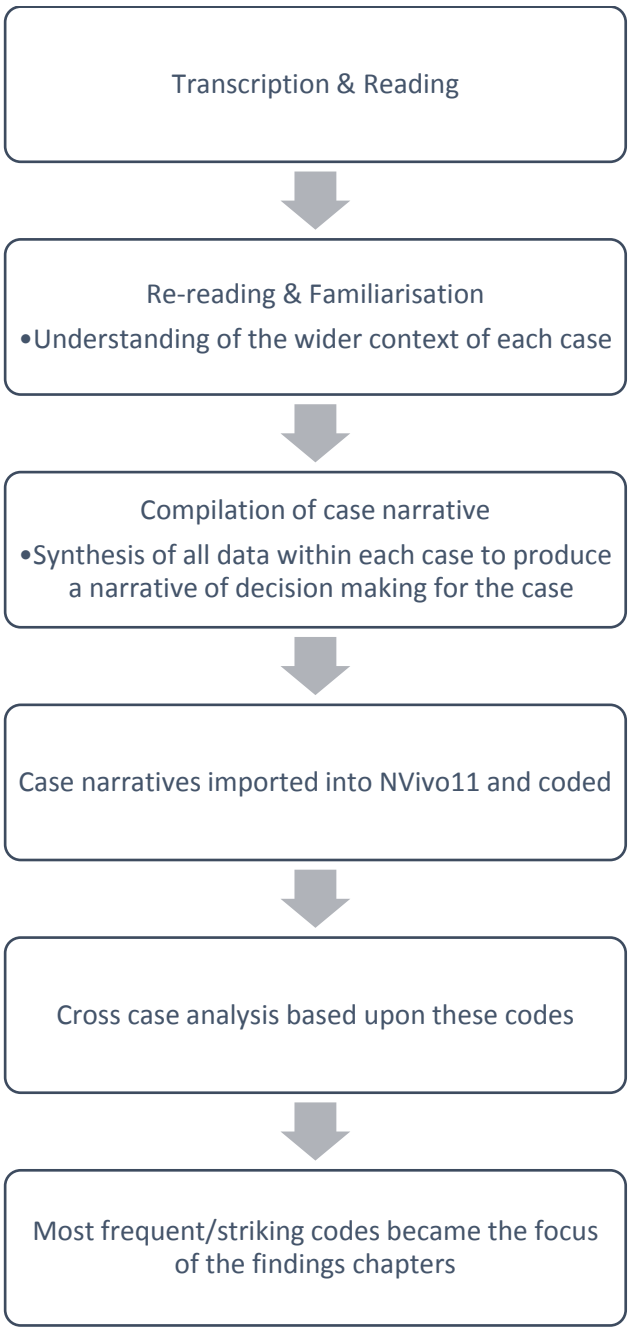


Figure 7: Process of data analysis

Stake (2006) argues that an understanding of the phenomena of interest is facilitated through cross-case analysis. The most pervasive themes identified as significant in the process of decision making and future planning were identified

through reference to the NVivo dataset and coding hierarchy, and the cross-case analysis was thereby facilitated.

A coding hierarchy chart was exported from NVivo 11 following the cross-case analysis and the prominence of themes identified within the data resulted in the findings chapters presented in this thesis. This hierarchy chart and an explanation are provided in Appendix 7 and demonstrate how the findings chapters have evolved from the data.

3.10. Research credibility

Credibility in qualitative research is concerned with the trustworthiness of the findings and whether they make sense (Miles and Huberman, 1994). Through carefully checking our results and considering our own presumptions, the research is made credible by minimising the impact of these presumptions on the resultant data (Kvale, 1994). Miles and Huberman (1994) suggest that thick description, triangulation with complimentary methods, and the presentation of confirmatory and contradictory cases all add to the credibility of qualitative research. In addition they ask whether the account is comprehensive and presented within the temporal context within which it occurred. This research has attended to these concerns through ongoing reflexivity, detailed attention to the specific contexts and triangulation of data collection methods. Attention to my own presumptions and searching for rival explanations has been facilitated through detailed discussion with supervisors and critical friends. The resultant findings are also plausible (Miles and Huberman, 1994) and therefore in conjunction with the strategies as described above, can be considered credible.

3.11. Conclusion

This chapter has detailed the study aim and objectives and outlined the philosophical assumptions underpinning this research. It has clarified and justified the methodology and methods which were used and outlined the governance procedures followed and sampling decisions made. It then referred to the specific ethical considerations which this research raised and concluded with an overview of the process of data analysis and strategies used to ensure research credibility.

Chapter 4. Introducing the cases

4.1. Introduction

This chapter provides a summary of the cases which made up this research. It aims to provide the reader with an understanding of the wider context of each case, the decisions required for each child and the factors which influenced their decision making. Within case study research, it is only through understanding the complexities and specifics of a case that a broader understanding of the phenomenon can be achieved (Stake, 2005). In order for the reader to develop an understanding of the specifics of the cases, an overview of each case will be presented. These will detail the broad context that the family operates within, the significant people involved in the case and the significant decisions observed and identified within each case.

Each case had different durations of participation and ranged from one single interview, to between 6 and 12 months. One case participated for less than two months and then withdrew from the study. The cases recruited, their composition and the data collection activities undertaken with each case are outlined in Table 4. In total, 46 interviews and 72 observations were conducted, and the medical notes of 9 children accessed and analysed.

Table 4: Participant characteristics and data collection activities

NB: All participants have been given pseudonyms in order to preserve their confidentiality and anonymity.

Case	Child's Name	Child's Characteristics	Interview participants (Role, number of interviews)	Observations	Notes Review
1	Daisy	2yr Female, Category 2	Catherine & Steve (Mum & Dad, 1) Catherine (Mum, 2)	5	✓
2	Maisie	1yr Female, Category 3	Fiona (Mum, 3) Dr Price (Paediatrician, 2) Dr Chase (Cardiologist, 2)	12	✓
3	Grace	10yr Female, Category 4	Kay (Mum, 3) Dr Price (Paediatrician, 2) Alice (School nurse, 2)	4	✓
4	Michael	10yr Male, Category 3	Kate (Mum, 3) Michael (3) Gail (Family Support Worker, 1)	5	✓
5	Elena	10yr Female, Category 3	Pauline (Mum, 3) Joanna (Physiotherapist, 1)	7	✓
6	Omar	4yr Male, Category 4	Nadia (Mum, 3) Hafisa (Omar's Cousin, 1)	9	✓
7	Hassan	1yr Male, Category 4	Salma (Mum, 2)	11	✓
8	Christopher	12yr Male, Category 4	Susan (Mum, 3) Dr Brett (Paediatrician, 2)	8	✓
9	Charlotte	14yr Female, Category 2	Charlotte and Rachel (Mum, 3) Jamie (Dad, 2) Jessica (Specialist Nurse, 1)	8	✓
10	Ella	13yr Female, Category 4	N/A*	3	N/A*
11	Evie	4yr Female, Category 4	Anthony & Ruth (Dad & Mum, 1)	N/A**	N/A**

*Ella's family withdrew after 3 observations, before an initial interview could be arranged and before the medical notes would have been accessed. **Evie's family felt that that they already had a myriad of professionals involved and did not wish to add another person into their life, and also expressed that they felt that it would not be useful for me to follow them for observations. Her parents did however consent to participating in one interview.

4.2. The Cases

The aim of this study was to recruit complex cases with multiple participants in each case and multiple sources of data. Not all cases wished to nominate significant others, but for all but one case, multiple sources of data were collected. In such cases, serial interviews and/or multiple observations and medical notes analysis enabled a detailed understanding of the case specific context and its individual complexity. One case consented to participate in one interview only. Despite the lesser volume of data gained from this case, it presented the opportunity to learn about the perspectives of a family who did not feel able to commit to participation in the full study.

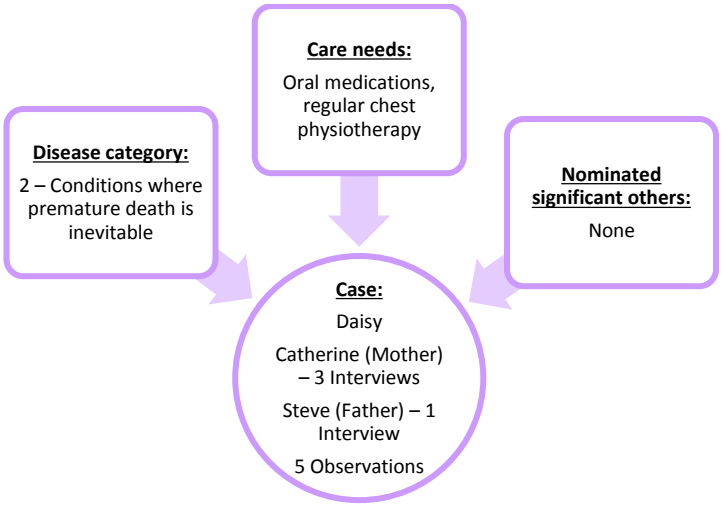
The decision has been made not to reveal some of the children's diagnoses (for those with very rare conditions) as, in conjunction with the description of the family context, doing so could result in a breach of anonymity. In such cases a broad description of the condition will be given. Each case will now be described following a standard format:

- Case composition and features - a discussion of the relevant people involved in the case, the child's disease category and their care needs.
- Case context – a description of the family structure, values, the child's health and wellbeing, and data collection activities.
- Important decisions – a discussion of the important decisions made by or being faced by the family.
- Case timeline – a graphic representation of the temporal location of important decisions, health care interactions and data collection activities.

4.2.1. Daisy's Case

Case composition and features

Name	**Relationship**
Catherine	Mother
Steve	Father
Dr Green	Consultant
Dr Dietrich	Consultant



Case context

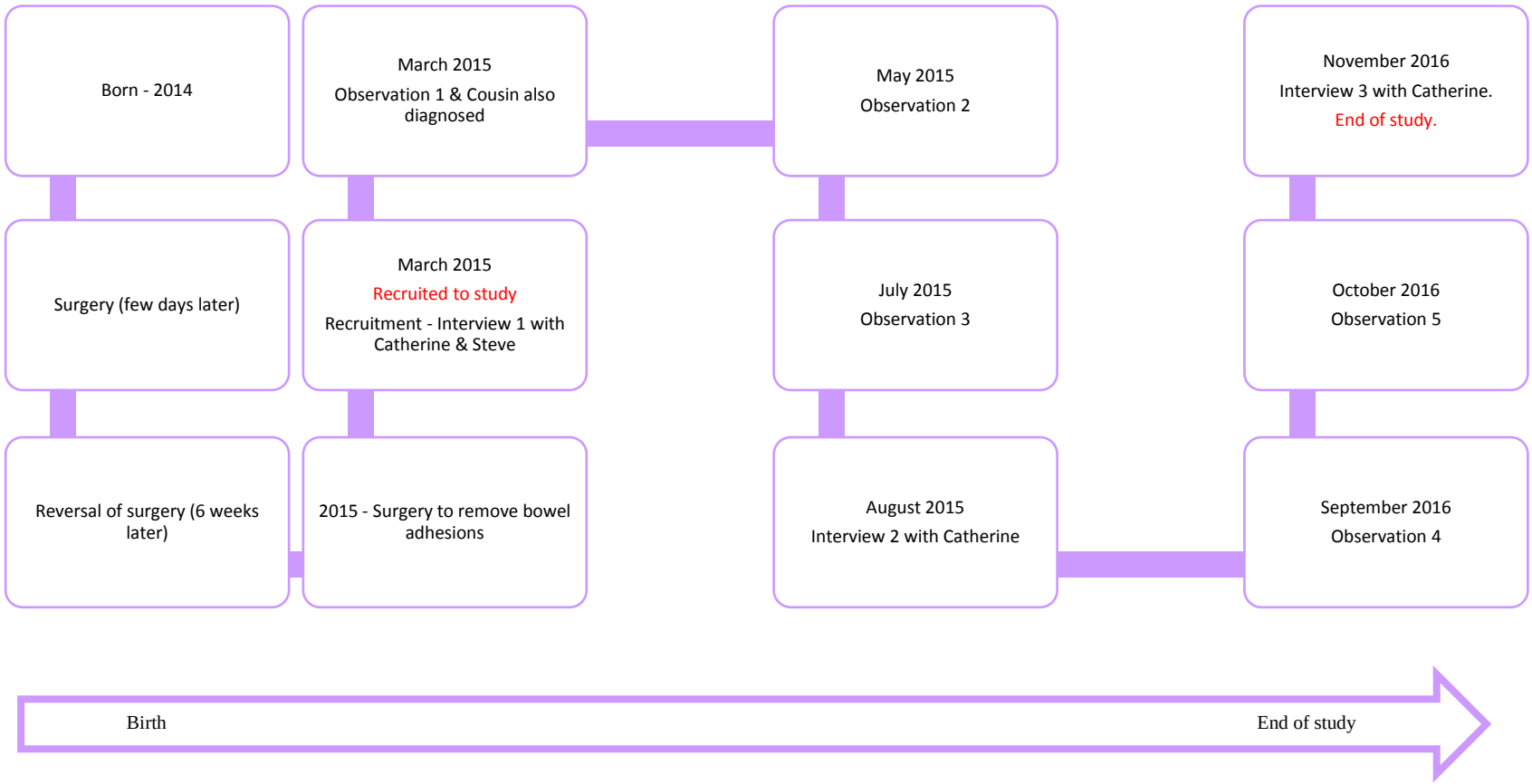
Daisy is a 2 year old girl with Cystic Fibrosis, who lives at home with her parents and 4 year old brother. Very little was articulated about Daisy's family's goals when making decisions for her, although it appeared that keeping her as well as possible, whilst enjoying life was what they aimed for. Catherine and Steve both acknowledged that Catherine was the main decision maker in the household regarding Daisy's care and treatment. Steve worked as a manager in the retail sector and Catherine did not work outside the home. The family were close, both geographically and in relation to contact, to Catherine's family and were well supported by them. The family expressed that they had less contact with Steve's family, which was partially due to them being more geographically distant, but Catherine also inferred that she had a better relationship with her side of the family. The family owned a car, although Catherine relied on Steve or public transport to take her places, as she was still in the process of learning to drive.

Data collection took place in two locations: interviews were conducted in the family home and observations all took place in the same clinic at the hospital at which Daisy was treated. The family stated that they did not have a religion.

During the final observation, Catherine's grandma (whom I had never met and who had never been mentioned before) was present. When Catherine took Daisy out of the room, I was informed by her grandma that Steve had left Catherine. This was not something that Catherine shared at the final interview and therefore the way in which decisions had been made since this change in family dynamic, or the perceived future negotiation and distribution of decision making within the family was not discussed.

Important decisions

The important decisions which the family had made prior to the study which were referenced in the interviews were regarding Daisy's gastrointestinal surgery, which she had first at a few days old, and then subsequently a few months later to reverse the procedure. Additionally she had another surgery at ten months old, due to a common complication following the initial surgeries. Ongoing everyday decisions the family faced were regarding her physiotherapy and the commencement of antibiotics when she developed a cough. The main decision facing the family during the study period was regarding Daisy's contact with her cousin, who had also been recently diagnosed with Cystic Fibrosis, as they had been made aware that there would be cross-infection considerations which could impact on the health of both children.

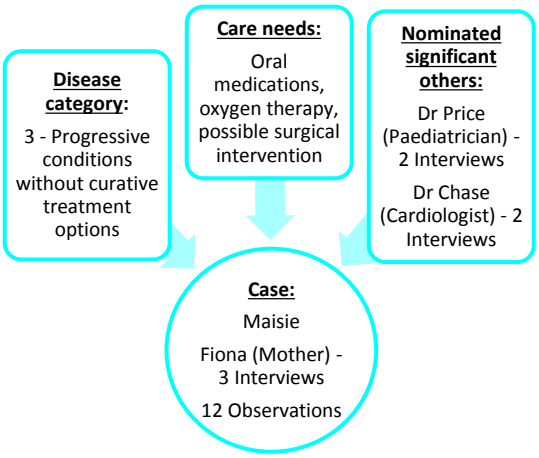


Timeline 1: Daisy's Case

4.2.2. Maisie's Case

Case composition and features

Name	Relationship	Name	Relationship
Fiona	Mother	***Dr Burton***	Paediatrician (with special interest in Cardiology)
Kieran	Father	***Dr Eltoft***	Cardiology Registrar
Luke	Brother	***Dr Alexis***	Initial Cardiac Interventionist
Tracey	Maternal Grandmother	***Dr Vijay***	Cardiac Interventionist
Dr Price	Paediatrician	***Dr Owens***	Paediatric Registrar (with special interest in Cardiology)
Dr Chase	Cardiologist	***Anna***	Family Support Worker



Case context

Maisie is a 22 month old girl born with congenital cardiac abnormalities. She has one 3 year old brother and lives with her parents, brother and her maternal Grandmother. The family own two cars and both parents are able to drive, something which Fiona valued in order to go on holiday and make memories as a family. The family had significant social support from their extensive network of family and friends and both Maisie and her brother would often go to stay at Fiona's friend's house at the weekend, to give Fiona and Kieran a break. Support

was available from Kieran's parents also, but Fiona was not always keen to accept this, as she felt that it was always on their terms. Fiona worked part time in the service industry and Kieran worked full time in logistics. Fiona placed an emphasis on working part time, explaining that this enabled the family to go on holidays often and provide her children with the opportunity to make memories. The family are aware that Maisie has a life-limiting condition and have undertaken planning for her end of life. Fiona also identified that her and her children were recently baptised as Roman Catholic.

Data collection took place in many locations. Interviews with Fiona took place in the family home and the extent of the social support was evident, as often friends would be present, or would enter the house during the interview, suggesting that the family operated an 'open door policy'. Interviews with Dr Price and Dr Chase took place in clinic rooms, or in offices, at various locations between the two hospitals at which Maisie was treated. Observations took place similarly in various clinic rooms between these organisations, and also on hospital wards when Maisie was admitted to hospital with an acute illness, or for investigations or surgery.

Maisie's diagnosis was discovered pre-natally on her 20 week scan, although the extent of her cardiac condition was unknown until birth. At birth, her diagnosis was considered palliative, and a palliative surgical procedure was provided in the first month of life. Maisie reached 22 months old by the end of the study and had met all of her developmental milestones, despite being told numerous times that this would be highly unlikely due to her condition. Fiona stated that she was the main decision maker for Maisie's care, and during appointments this was observed to be the case, as she was the family member present at all appointments, the one most engaged in asking questions and discussing the possible options, and the one responsible for signing any consent forms. However, Fiona stated that Kieran and Tracey also helped her to make decisions. Fiona expressed awareness that the decisions that she is offered are constrained within the boundaries of what professionals feel is appropriate.

Important decisions

The main decision to be made for Maisie during the course of the study was regarding further surgical intervention to her heart. The case involved a lack of consensus about the appropriate options to offer Maisie and her family and therefore the decision making process regarding further surgery was observed to be erratic. The oscillating nature of this decision is outlined in Figure 8, which describes the timeline of decision making for this specific decision. There was uncertainty regarding the outcome of any possible further surgery and therefore the decision was, after much discussion among the medical professionals, delegated to her family. Daily minor decisions for Maisie involved amount of oxygen delivered, commencement of oral antibiotics and admissions to hospital with normal childhood illnesses and injuries. In addition to this, planning for Maisie's deterioration and end of life were identified as important decisions that her family had made.

Timeline 2: Maisie's Case

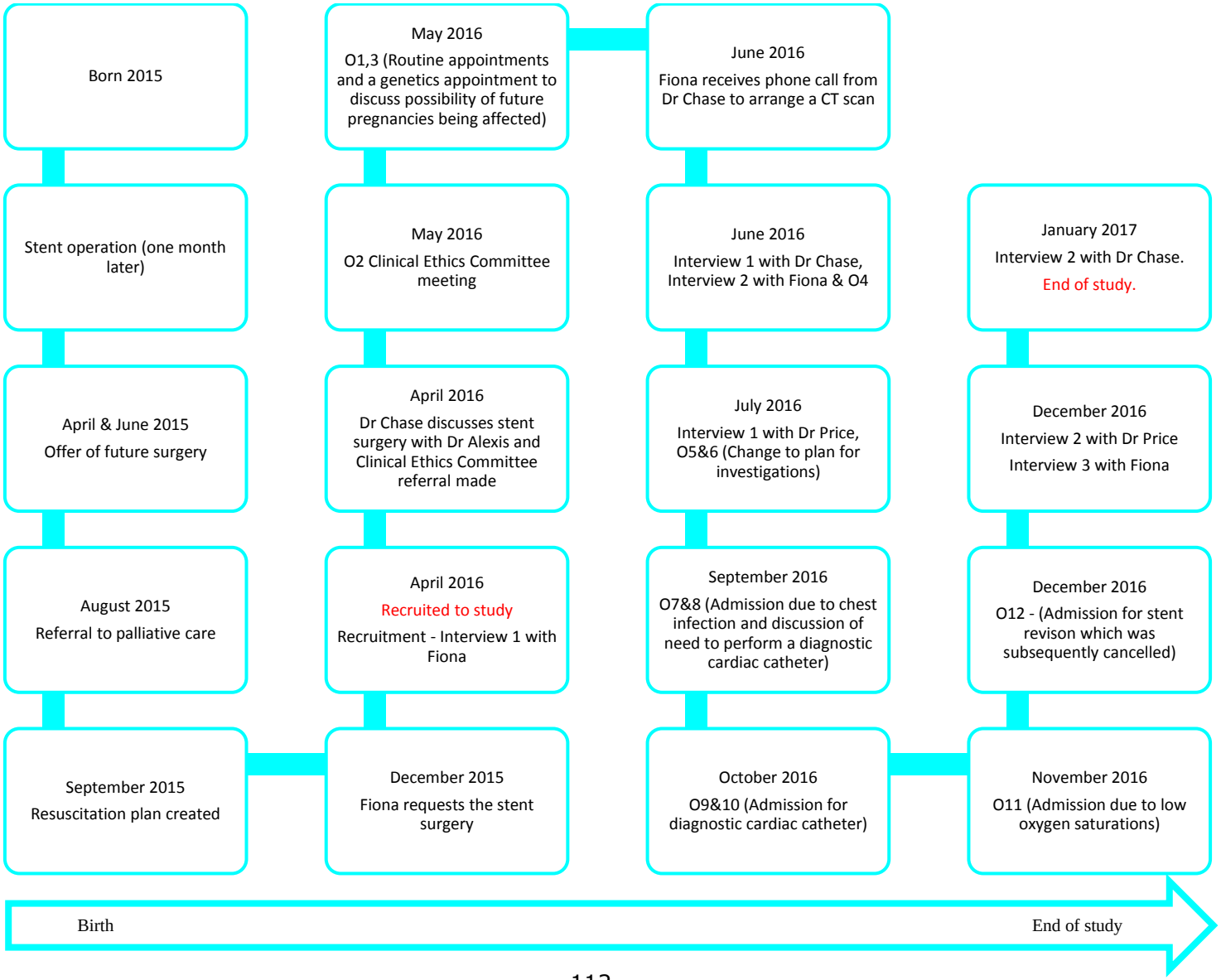
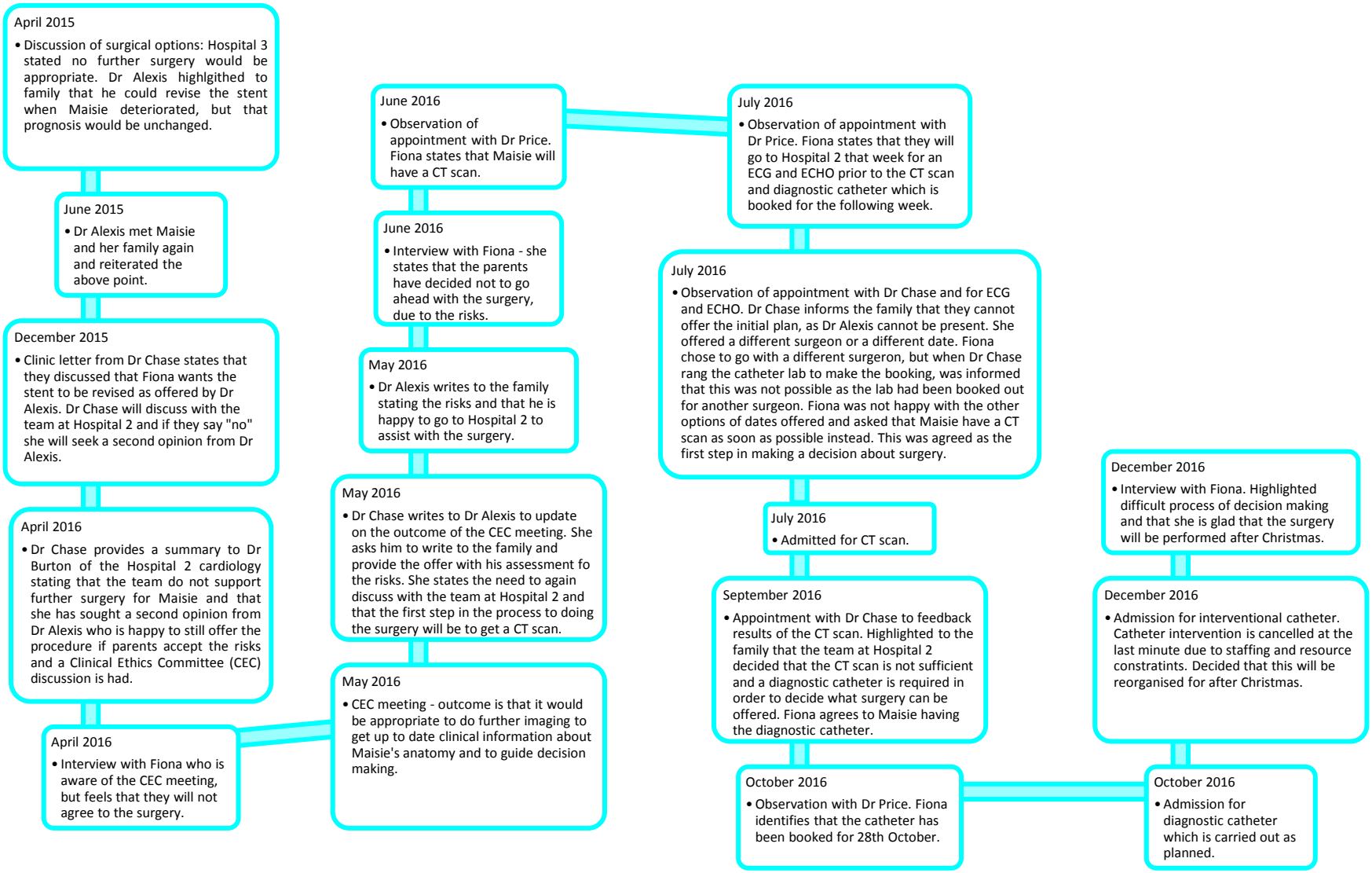


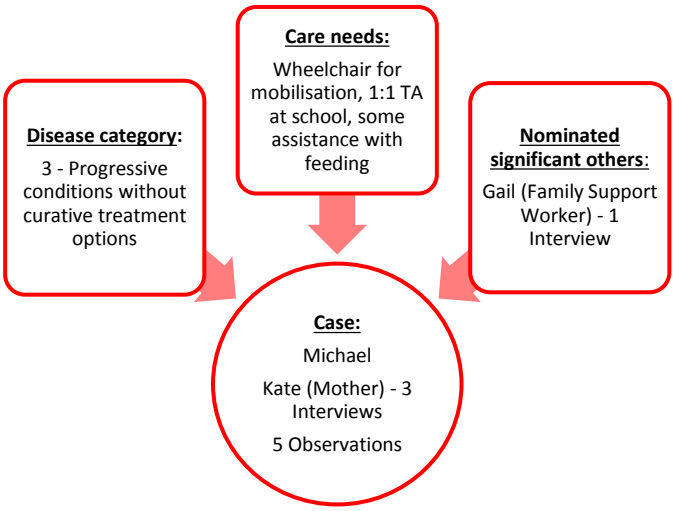
Figure 8: Maisie's cardiac surgery decision timeline



4.2.3. Michael's Case

Case composition and features

Name	Relationship	Name	Relationship
Kate	Mother	***Cilla***	School Teaching Assistant
Gail	Charity Family Support Worker	***Joanna***	Physiotherapist
Dr Price	Paediatrician	***Mr Smith***	Paediatric Surgeon
Dr Patel	Respiratory Consultant		



Case context

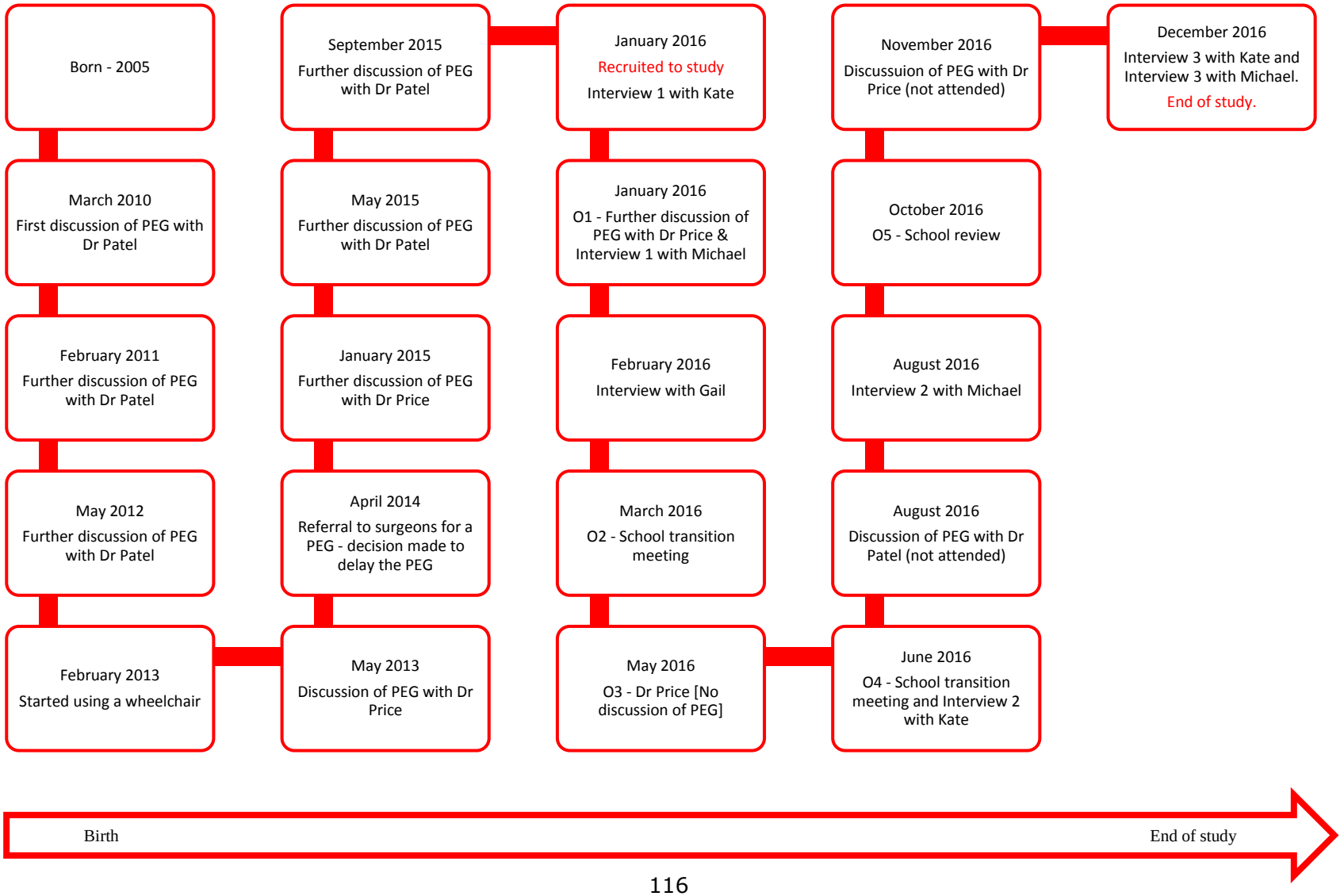
Michael is a 10 year old boy who has a condition under the broad category of 'systemic atrophies primarily affecting the central nervous system'. He lives at home with his Mum, Kate, and his 18 year old sister, but does not have any contact with his Dad. Kate drives and owns a van which has been modified to transport Michael in his wheelchair. The family has no contact with Michael's father or his side of the family, and although Kate has regular contact with her own family, the practical support that they provide to her in caring for Michael is limited. Kate is heavily involved in the disease community, receiving and offering support on social networks and it appeared that this was where she drew upon for the support that she needed. She also stated that she is often asked to

speak to groups of professionals about the condition and her experience of health care.

Michael was born following a normal pregnancy and uncomplicated birth, but became unwell with pneumonia during infancy and was admitted to hospital for treatment. When he was discharged he failed to meet some of his developmental milestones normally and after numerous investigations over the following years, received his life-limiting diagnosis at two years old. Michael has remained fairly well since this, suffering from only minor childhood illnesses. He is currently in a wheelchair and attends a mainstream secondary school. Maintaining normality for as long as possible and Michael's self-esteem appeared to be one of the most significant factors underpinning Kate's decision making for Michael. Kate identified that her family were not religious.

Interviews with Kate took place in the family home, or in a café at her request. Interviews with Michael took place at home or in the clinic waiting room. The interview with Gail was conducted over the phone. Observations of medical appointments took place in the same clinic room in the outpatient department of the hospital, and transition meetings took place in the staff room of Michael's primary school and the occupational therapy office of his secondary school.

Timeline 3: Michael's Case



Important decisions

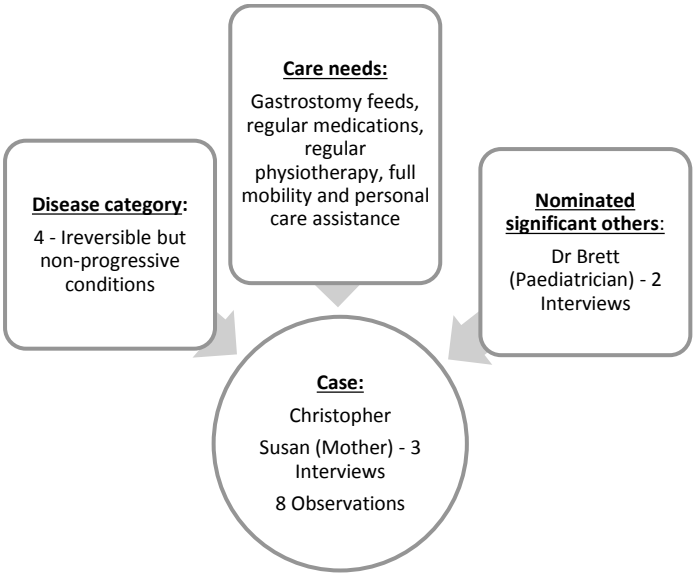
The main decisions for Michael during the study period were gastrostomy[8] placement and transition to secondary school. Michael's Respiratory Consultant, Dr Patel, introduced the idea of a gastrostomy when Michael was five years old and discussion of this had become a regular feature of his clinic appointments since; Michael and Kate are currently refusing this treatment. The other decision that Kate faced for Michael during the study period was regarding his transition to secondary school. The recommendation of Michael's educational professionals was that he start out in Nurture (supported learning) classes, but Kate felt that this would be detrimental to his self-esteem and his development, as it would not push him to achieve his potential. She therefore fought for Michael to start out in mainstream classes and during transition meetings, made her feelings known and argued her case vehemently. Other previous decisions that had been made throughout Michael's life had been regarding medications and the development of a resuscitation plan, although neither of these decisions appeared to hold much significance for Kate.

[8] Gastrostomy – also referred to as a PEG (Percutaneous Endoscopic Gastrostomy). This is a feeding tube which is passed directly through the wall of the abdomen and into the stomach. It is permanent in so far as it can remain in-situ forever, but can be removed (and the hole will close over) if it is no longer needed.

4.2.4. Christopher's Case

Significant people

Name	Relationship	Name	Relationship
Susan	Mother	***Dr Novick***	Consultant – Regular Botox injections
Jake	Father	***Mr Wallis***	Orthopaedic Surgeon
Dr Brett	Paediatrician	***Dr Chapman***	Paediatric Neurologist
Dr Martin	Respiratory Consultant		



Case context

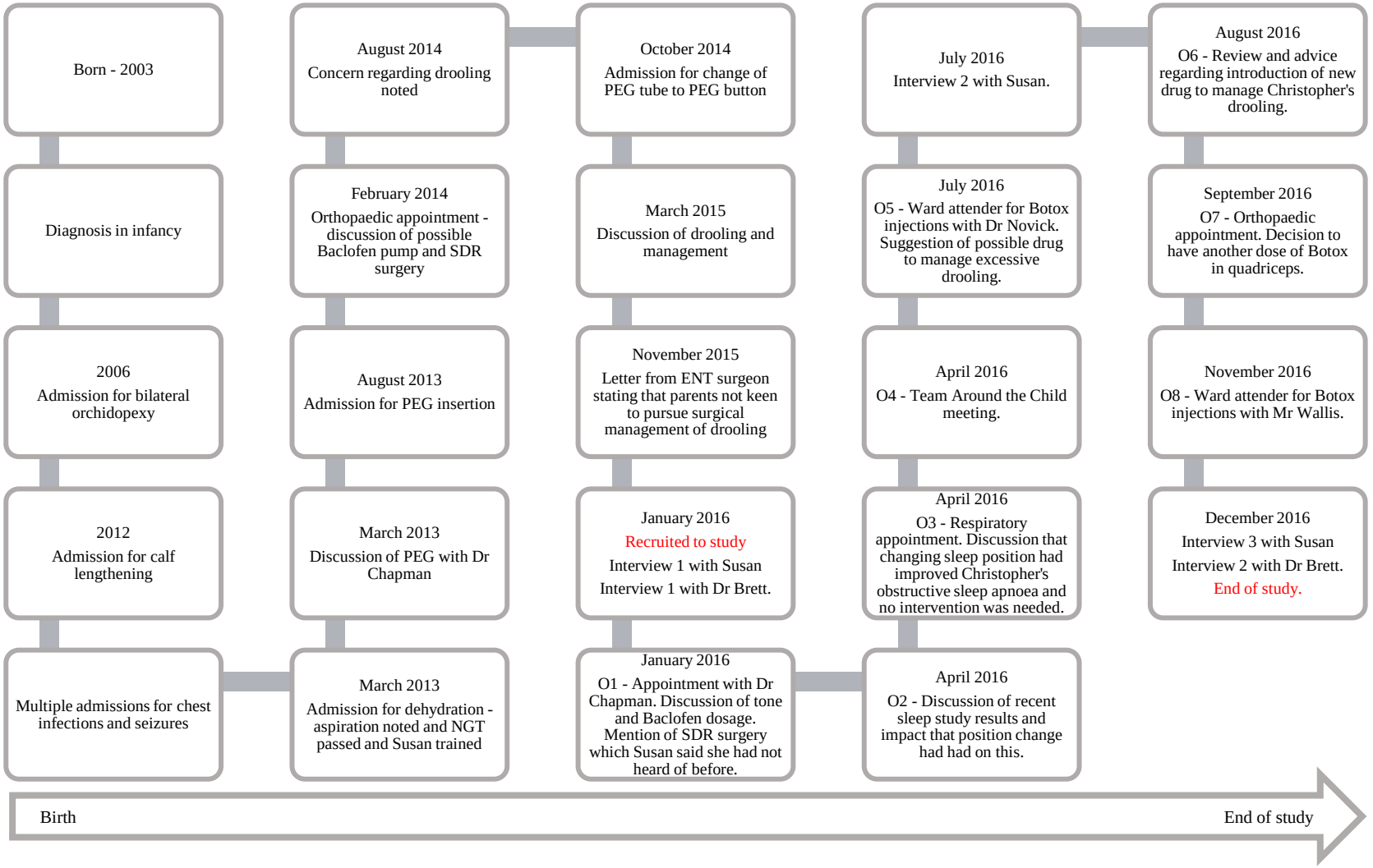
Christopher is a 12 year old boy who has severe Cerebral Palsy. Christopher is an only child and lives with his parents and his Grandfather, with whom he has a close bond. Susan co-owns a business and is able to work from home when Christopher is at school. Jake works in logistics and the family appear not to struggle with money. The family support that they receive is limited and additional support in terms of respite care has been resisted by Susan. She identified that having previously worked as a carer, she had seen poor practice and therefore would not feel confident leaving Christopher with anyone else.

The only support that she accepted was a few hours of home respite a week, which enabled her to go shopping and run any errands she needed.

Data collection took place in a variety of locations, with Susan's interviews being conducted in the family home and Dr Brett's interviews being conducted in her office, for the first interview, and via the telephone, for her second interview. Observations took place in a number of clinic rooms across the two hospitals at which Christopher received care and in the school staff room, for his Team Around the Child meeting.

Christopher has been on anti-convulsant medication all his life in order to maintain seizure control and when he was younger, he was frequently in hospital with chest infections. Since having a gastrostomy some years ago he has been relatively well. Christopher attends a local special needs school and Susan is heavily involved in the parent body. The family identify as being part of the Church of England.

Timeline 4: Christopher's Case



Important decisions

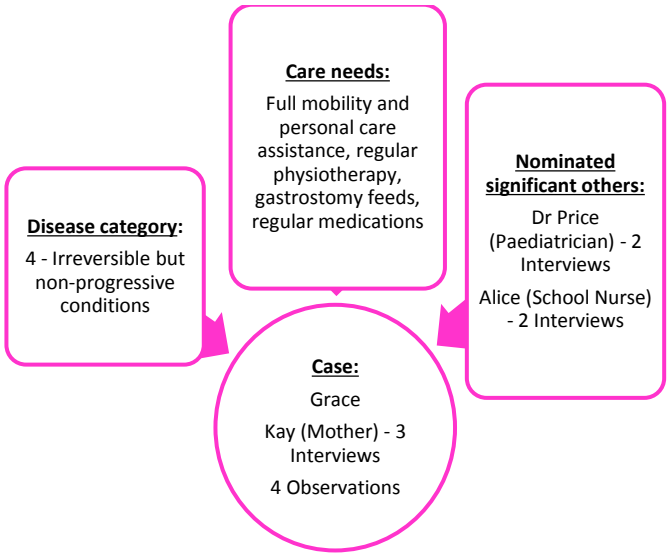
There were numerous decisions discussed during the study, some of which had previously been made, such as gastrostomy placement and Botox injections, whereas others were highlighted during the study and involved airway management and musculoskeletal management. Tube feeding had been commenced prior to the study during one hospital admission for a chest infection where it was noted by the professionals that Christopher was aspirating and they decided to terminate oral feeding and insert an NG tube[9]. After around 6 months of having the NG tube, Christopher had a gastrostomy and fundoplication. Since having terminated oral feeding three years ago he has had no further seizures and had not had any significant chest infections, until during the study. Christopher has regular Botox injections to relax his muscles, as they become very tight and uncomfortable. The decision to have Botox itself was not identified as being significant for the family, although the decisions regarding who they would allow to administer these injections were identified as holding greater significance for them and at times resulted in significant concern.

[9] NG tube (nasogastric tube). This is a tube which is passed through the nose, down the oesophagus and into the stomach. It is secured on the face and may be used during an acute illness whilst the child is temporarily unable to feed orally, or as a long term solution until a PEG can be inserted surgically.

4.2.5. Grace's Case

Case composition and features

Name	Relationship	Name	Relationship
Kay	Mother	***Glenn***	Social Worker
Alice	School Nurse	***Lucy***	Case Manager – Continuing Care
Dr Price	Paediatrician	***Mr Finlay***	Orthopaedic Surgeon



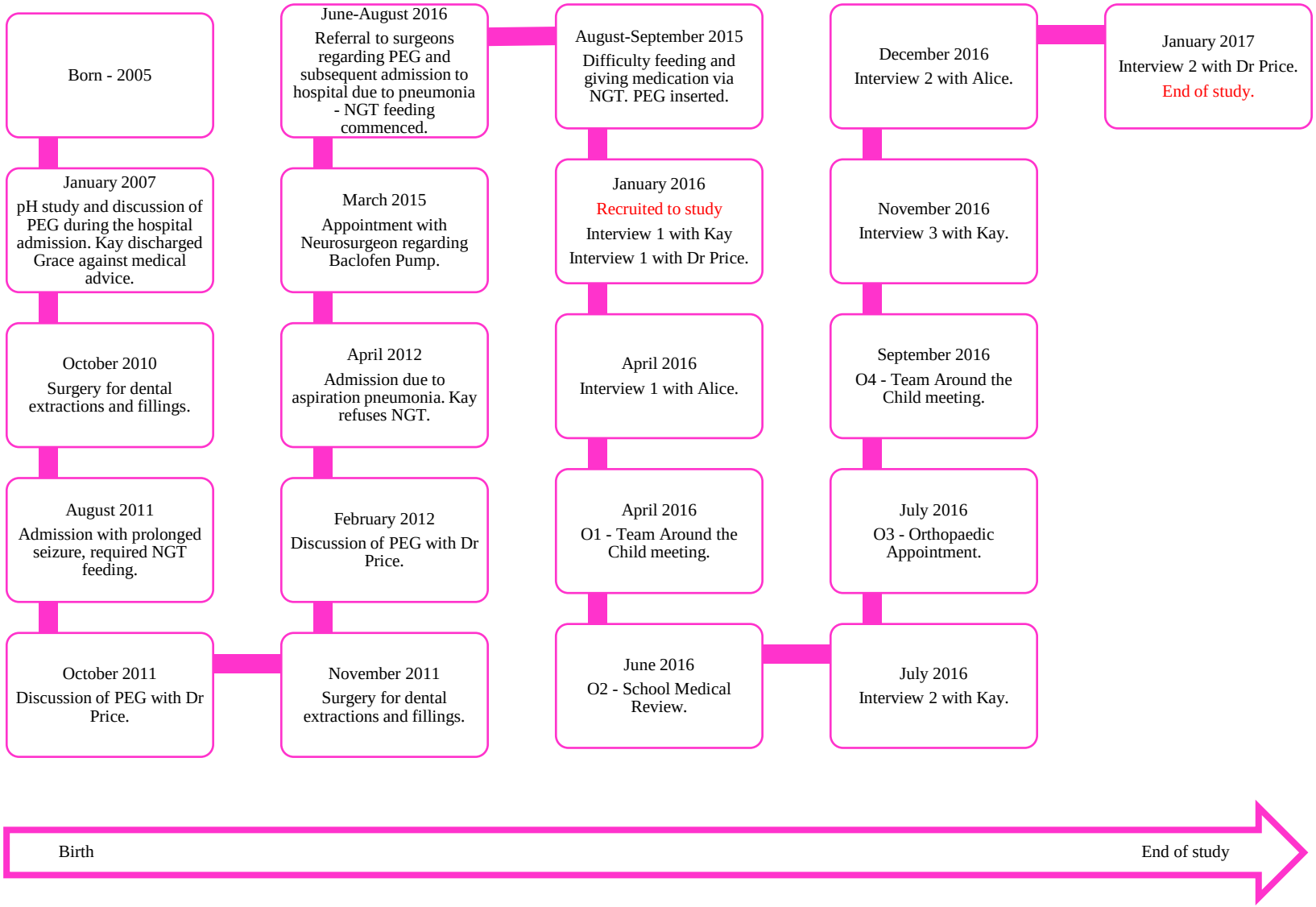
Case context

Grace is a 10 year old girl with severe Cerebral Palsy who lives with her Mum, Kay. Grace's father was denied contact with her from a young age because he did not believe in modern medicine and refused to allow Grace to have treatment. Grace's father has since died. She has half-sisters and nieces and nephews who she has never met, as they live abroad. Kay and Grace experience some degree of social isolation, as they were required to move away from their support network to a more suitable property on the other side of the city, to accommodate Grace as she grew. Kay and Grace's social support predominantly came from their church community, as Kay is a devout Christian. Since being rehoused, however, the family have been distanced from the support they received from the church. Kay herself has numerous medical conditions and struggles to

provide all Grace's care and therefore requires assistance for this. Her medical conditions and reliance on public transport mean that she is not always able to travel to visit her friends, therefore remains distanced from her social support.

Data collection took place in a number of locations. Interviews with Kay took place in the family home, interviews with Alice took place in the nurse's office at Grace's school and one interview with Dr Price took place in her clinic room and the other took place in her home. Observations of medical appointments took place in various clinic rooms at outpatient departments and the school medical room, and Team Around the Child meetings took place in the school 'Parent's Room'.

Timeline 5: Grace's Case



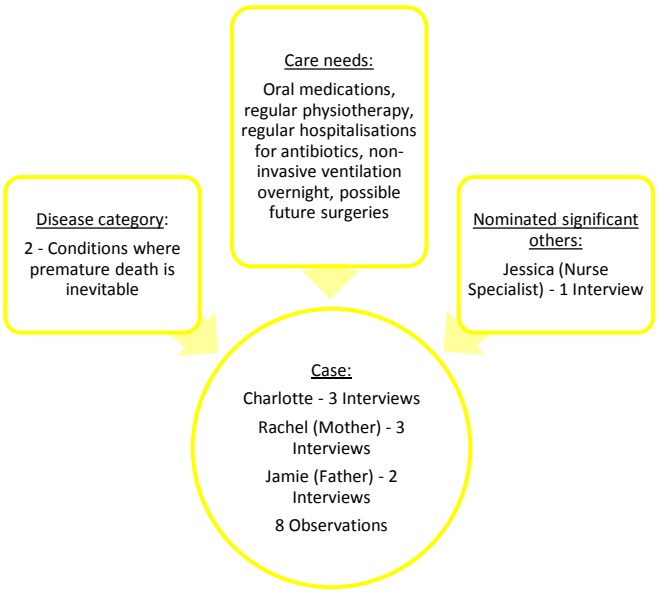
Important decisions

Grace has had numerous hospital admissions throughout her life and the year prior to the study experienced a life-threatening chest infection, requiring chest drains. During that admission Grace received a gastrostomy and has largely been well since then, avoiding any lengthy hospital admissions during the study period. The main decision that was discussed in Grace's case was her gastrostomy, which was identified as having been a protracted decision occurring over many years, with Kay's resistance to artificial feeding as the reason for the protracted nature of the decision. Dr Price identified that she had requested Alice to develop a good relationship with Kay in order to help persuade her that gastrostomy insertion was required. Termination of oral feeding was also identified as an important decision for Grace, as the professionals felt that she was unsafe to feed orally, something that Kay disagreed with, but was unable to resist this decision due to the intervention of Social Care. The other important decisions considered by Kay during the study period was regarding going on holiday with Grace and accessing respite care. Although not identified as important for Kay, Alice and Dr Price identified that an important ongoing decision for Grace was the development of an emergency health care plan.

4.2.6. Charlotte's Case

Case composition and features

Name	Relationship	Name	Relationship
Rachel	Mother	***Dr Randell***	Respiratory Consultant
Jamie	Father	***Jessica***	Specialist Nurse
Dr Green	Respiratory Consultant	***Dr Rutter***	Liver Consultant (Local hospital)
Dr Dietrich	Respiratory Consultant	***Dr Tate***	Liver Consultant (Regional hospital)
Dr Khan	Respiratory Consultant		



[10]

Case context

Charlotte is a 14 year old girl with Cystic Fibrosis, who lives with her Mum. Rachel has a car and drives and therefore does not need to rely on anyone else to assist with transport. Charlotte is in frequent telephone and text contact with her Dad, Jamie, although the amount she sees him is irregular because he is remarried with other children and often works abroad. Rachel does not work, as

[10] Jamie participated in two interviews, but arrived at Charlotte's house at the end of the third interview with Rachel and Charlotte and participated in a few minutes of that interview.

she cares for Charlotte full time. She expressed distress that she is no longer able to work and make her own money and that she is partially reliant on benefit, stating that this has a significant impact on her self-respect. Rachel receives some social support from Jamie's mother, who she has a good relationship with, but does not have such close or regular contact with her own family. Rachel has her own friendship group, but often expressed that she was unable to meet with them due to caring responsibilities for Charlotte. Charlotte herself appeared to be somewhat isolated from her peers, especially as she had stopped going to school and began home schooling. However, she expressed that she had a core group of friends that she was frequently in contact with over the internet and via text messaging. The family stated that they did not have a religion.

Cystic fibrosis is usually diagnosed in infancy, but Charlotte was undiagnosed until she was 2 years old, because she lived abroad, in a region where the condition is uncommon. Whilst living abroad, her treatment was arranged through their current medical team in England and Jamie was heavily involved in organising Charlotte's treatment during that time. She was reviewed by the team in England during regular holidays back to the UK and when she reached school age she moved back to the UK with her Mum and her parents separated. Charlotte is from a wealthy and well educated family and education is important to them. Despite her ill health, Charlotte expressed a desire to learn and achieve a good education so that she can go to University. Charlotte requires regular hospital admissions for antibiotics to maintain her health.

Interviews with Charlotte and Rachel took place in the family home or in Charlotte's room during one hospital admission. The interviews with Jamie took place over the phone and the interview with Jessica was conducted in a clinic room. Most observations were within her regular clinic department, but occasional observations took place in the general outpatient department or in her room on the ward, during admissions to hospital.

Important decisions

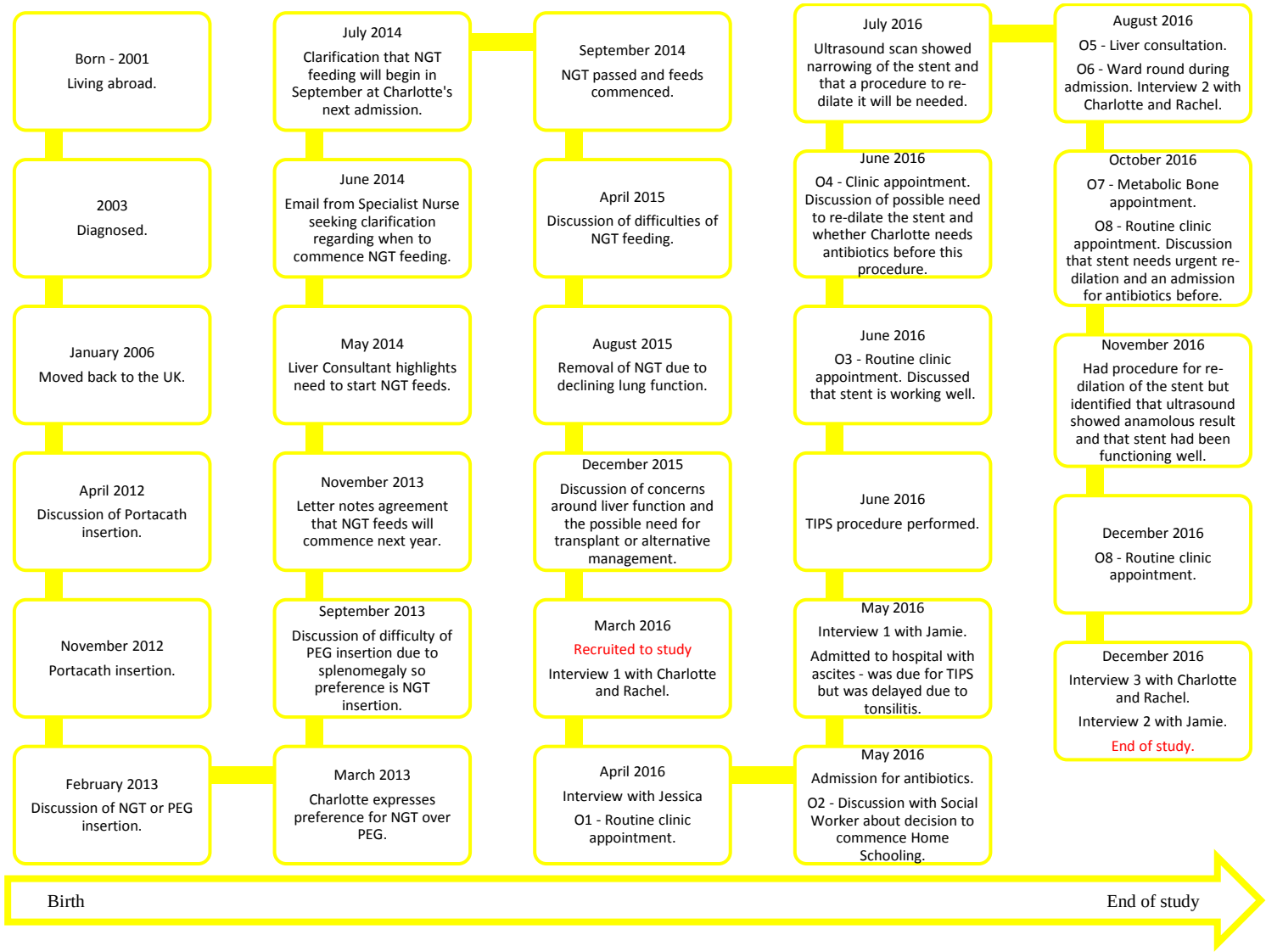
Charlotte has been faced with numerous decisions regarding surgery to site a Portacath[11], nutritional support, and schooling, and faced decisions around maintaining her liver function, through the TIPS procedure[12]. She may also face decisions regarding liver and /or lung transplant in the future, something that the family, and the professionals involved in her care discussed during the study. She has a complex treatment regime, requiring numerous medications, twice or three times daily physiotherapy and use of non-invasive ventilation[13] overnight and is required to make daily decisions regarding treatment management.

[11] This is a device which is inserted surgically and provides more reliable intravenous access. It reduces the need for cannulas (intravenous devices sited peripherally which are often unreliable and require re-siting more frequently) and needles to take blood samples.

[12] TIPS - Transjugular intrahepatic portosystemic shunt – a procedure that may be used to reduce portal hypertension and its complications, especially variceal bleeding.

[13] Non-invasive ventilation (NIV) – this is a device which provides breathing support and ensures that there is adequate ventilation.

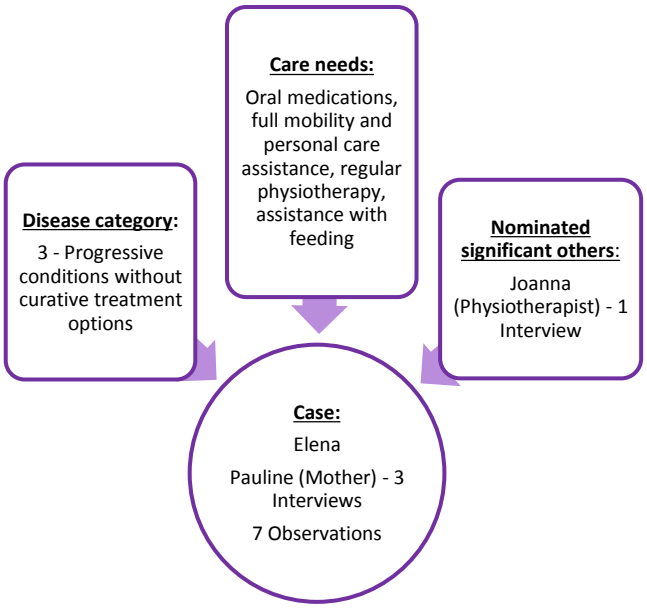
Timeline 6: Charlotte's Case



4.2.7. Elena's Case

Significant people

Name	Relationship	Name	Relationship
Pauline	Mother	***Joanna***	Physiotherapist
Richard	Father	***Polly***	New Physiotherapist (School Based)
Dr Price	Paediatrician	***Lucy***	Case Manager – Continuing Care
Dr Chapman	Paediatric Neurologist	***Doris***	Home carer
Dr Ludd	Specialist Consultant (Regional hospital)	***Mr Kelly***	Surgeon
Mr Machin	Spinal Surgeon		



Case context

Elena is a 10 year old girl with a very rare metabolic disorder. She lives with her Mum, Dad and older half-sister, who is now a paid part-time carer for Elena. They live in the family home and have a car which is modified to accommodate Elena in her wheelchair; both parents drive. She has two adult half-siblings who are from both her parents' previous relationships. Richard works full time in Management and Pauline cares for Elena full time, having given up her career in the service industry. The family support they receive is limited. Pauline's

mother and brother died a few years ago and they are geographically remote from her father. Pauline explained that she had a strained relationship with Richard's family and felt that they can be forceful in their opinions. Pauline articulated that they have an extremely supportive and close knit friendship group locally who provide support to the family. Pauline is involved in the global disease community through social networks and appeared to gain significant support from her participation in these. She has also gained awareness of the disease trajectory as a result of her involvement in the online community.

Elena was born normally and there were no complications during the pregnancy or early years. When Elena started school, she was identified as having some learning and co-ordination issues which were attributed to dyslexia and dyspraxia, but investigations conducted during an acute illness confirmed her diagnosis. Since diagnosis, Elena has lost the ability to walk and needs full assistance for mobilising and her personal care. Elena's parents are concerned about her having a general anaesthetic, because they have been informed that this speeds up degeneration for children with her specific condition and this has impacted on their decision making, as they are resistant to any surgery, for fear of Elena losing abilities sooner. Maintaining normality and Elena's abilities for as long as possible appeared key to the family's decision making. The family did not identify as religious.

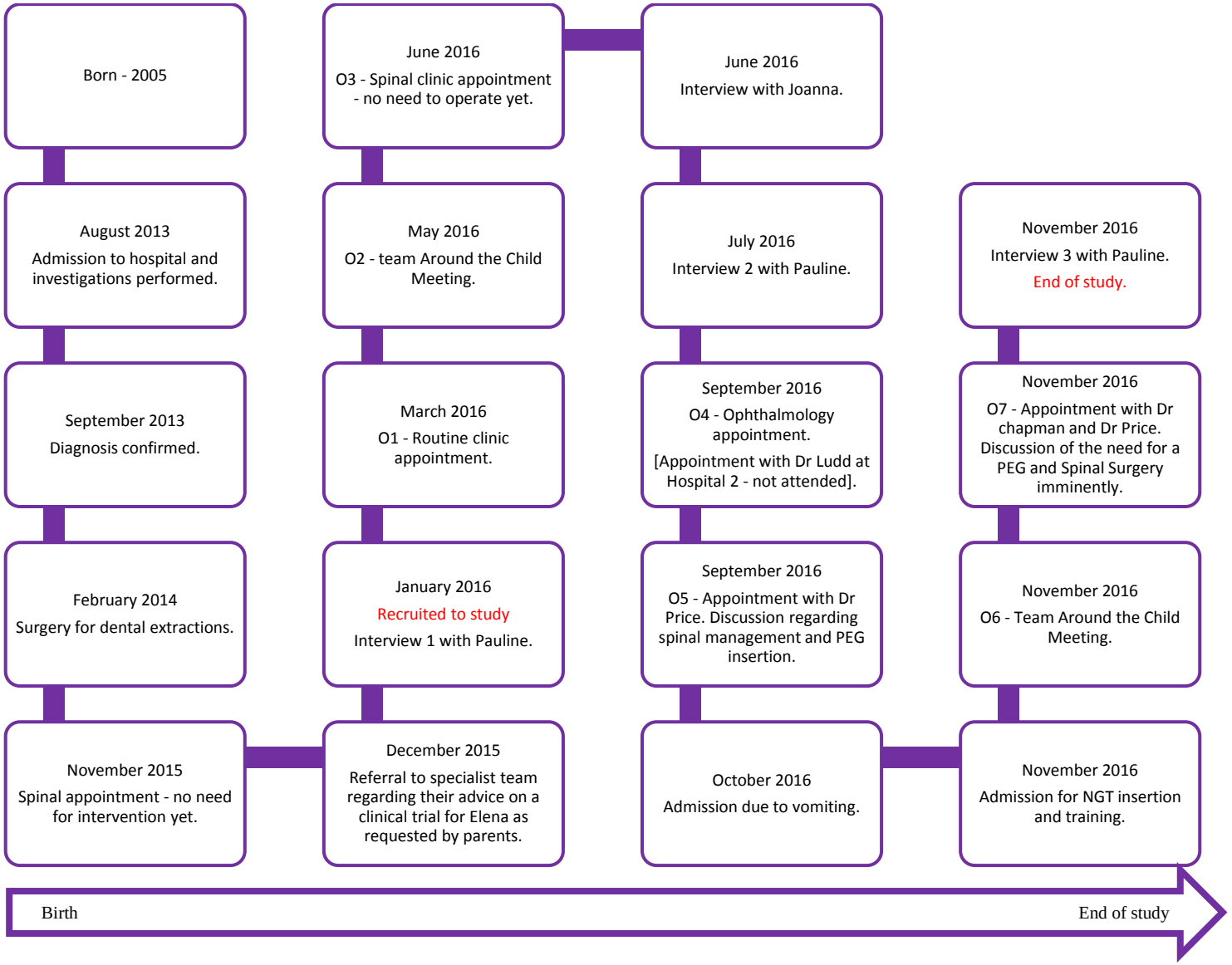
Data collection took place in a number of locations. Interviews with Pauline took place in the family home and the interview with Joanna was conducted in her office. Observations were in clinic rooms or in the school 'Parent's Room', for the Team Around the Child meetings.

Important decisions

The most significant decisions within this case were those regarding gastrostomy and scoliosis surgery, given the family's concerns about the impact of a general anaesthetic for Elena. Pauline spoke about housing and equipment decisions during the interviews, alongside decisions regarding a care package and development of a resuscitation plan, and despite being significant for her in relation to the frustration that these had caused, held less significance as they were not perceived to directly impact on Elena's condition. Other decisions such

as commencement of new medications and transition out of mainstream school were identified to be significant for Pauline.

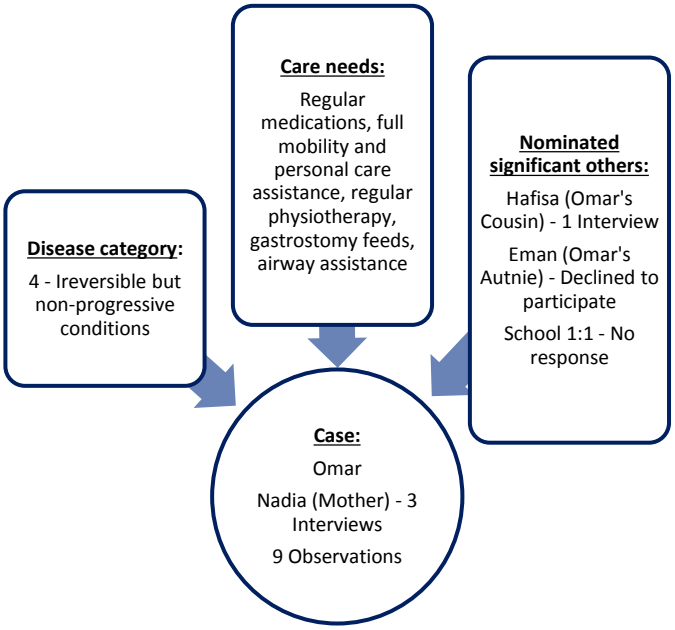
Timeline 7: Elena's Case



4.2.8. Omar's Case

Case composition and features

Name	Relationship	Name	Relationship
Nadia	Mother	***Dr Martin***	Respiratory Consultant
Jamal	Father	***Mr Finlay***	Orthopaedic Surgeon
Eman	Auntie	***Mr Field***	ENT Surgeon
Hafisa	Cousin	***Lucy***	Case Manager – Continuing Care
Dr Price	Paediatrician		



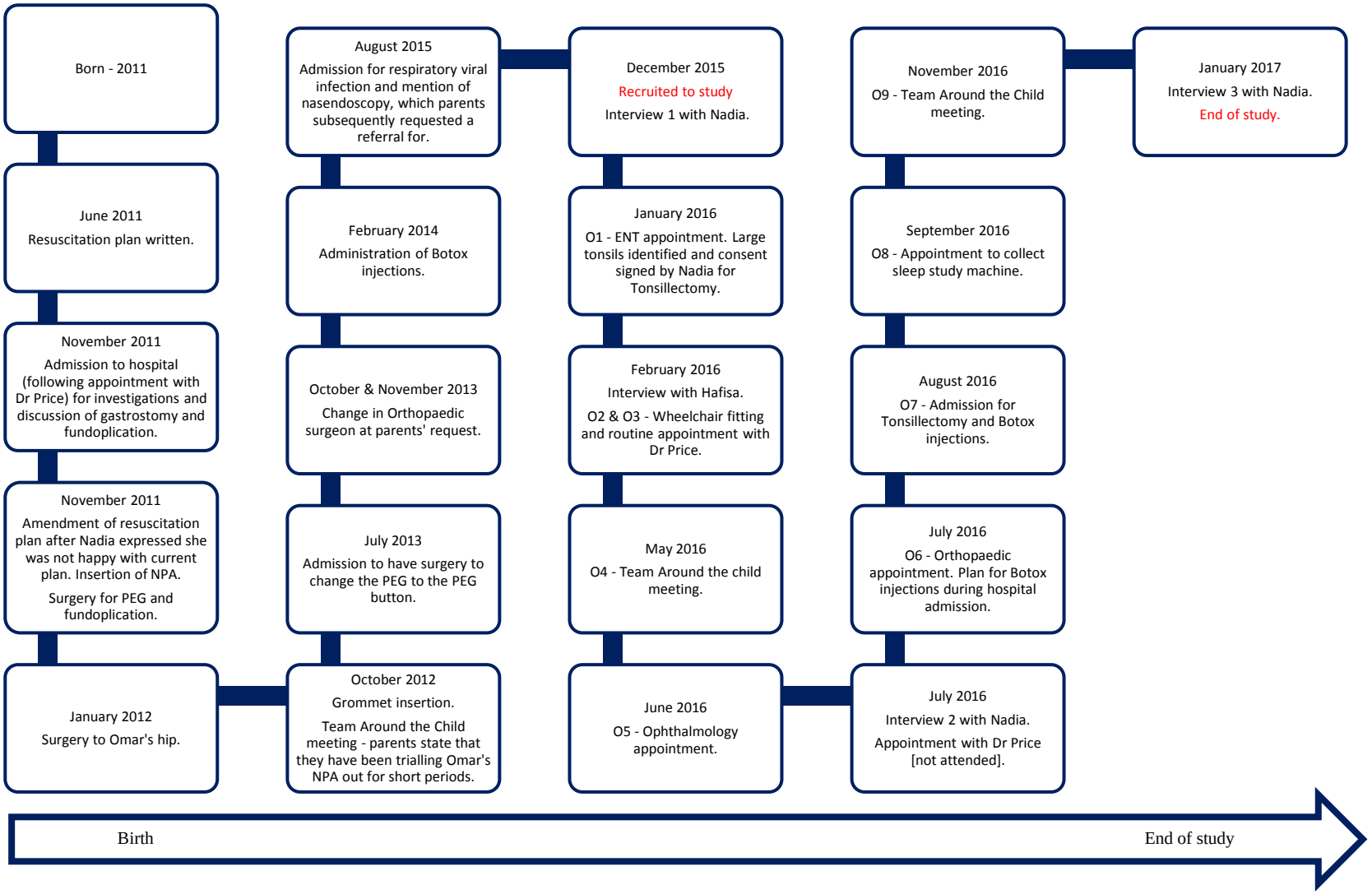
Case context

Omar is a 4 year old boy who has severe Cerebral Palsy as a result of complications at birth. Omar is from a devout Muslim family and lives at his auntie's house with Nadia, his mother. Omar's Dad, Jamal, lives in a different property and once they find a suitable property to move to, they will live as a family again. Jamal has his own car and drives, but Nadia is unable to drive and therefore relies on Jamal to take her to appointments, or is required to get a taxi. Jamal works full time in the service industry, Nadia does not work outside the home and Eman works part-time. Omar's extended family are very close and his cousin, Hafisa, was heavily involved with his care at birth and in infancy, as she was at that time studying to be a midwife. Omar had a very poor prognosis at

birth and was in hospital frequently in infancy. He has maintained good health in the last few years, avoiding any lengthy hospital admissions due to intercurrent illnesses, and is developing well. He attends a local school for children with special needs and his family and teachers expressed happiness at his progress developmentally.

Interviews all took place at the family home. Nadia nominated a number of people to participate in interviews, but not all of these participated. There was no response from Omar's 1:1 worker at school regarding participation and despite Eman initially agreeing to participate, eventually decided that talking about Omar would be too difficult emotionally for her, so did not participate. Jamal initially agreed to participate in an interview, however, following numerous attempts to arrange this, decided that it would not be possible to do so. Observations took place in a variety of locations: clinic rooms, ward areas and the 'Parent's Room' at the school for Omar's Team Around the Child meetings.

Timeline 8: Omar's Case



Important decisions

The important decisions for Omar's family in the past, which were discussed in interviews, were regarding previous surgeries (on his hips, for grommets[14] and gastrostomy and fundoplication[15]), airway management, which involved full time use of a nasopharyngeal airway[16], and development of a resuscitation plan. Throughout the study the major decisions ongoing were regarding long term airway management, musculoskeletal management, his care package and litigation against the hospital. The litigation referred to events surrounding Omar's birth, as it was identified that medical negligence was at least partially responsible for Omar's disability. By the end of the study the hospital had admitted two out of three aspects of their injury claim, but were refusing to accept causality. Nadia identified her perception that this was not good enough for the family and had been advised by the solicitor to pursue the full claim and not settle out of court, which is what they decided to do; the case is ongoing.

[14] Grommets – these are tiny tubes that are inserted into the child's eardrum when the child has a build-up of fluid in the middle ear (resulting in trouble hearing) and enables the drainage of this fluid away.

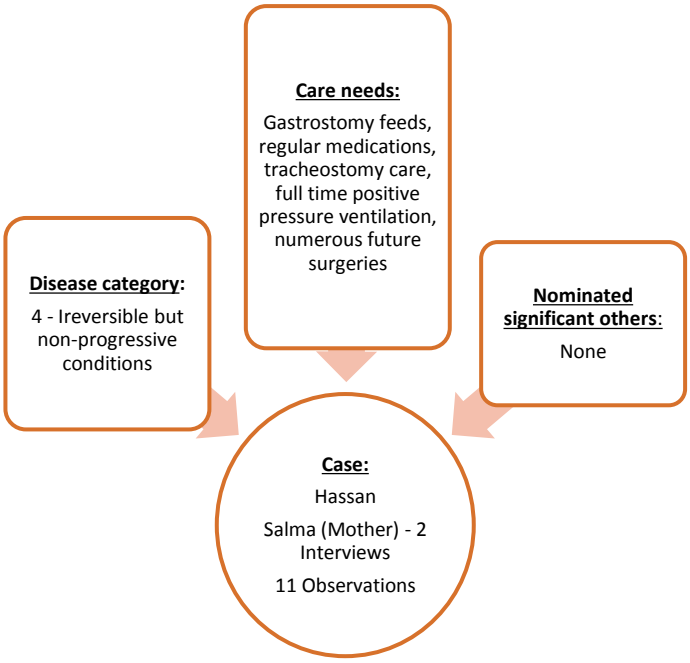
[15] Fundoplication (also referred to as a 'nissens' procedure) – this is a surgical procedure which makes the lower oesophageal sphincter (the opening between the oesophagus and stomach) tighter and helps to reduce or prevent gastro-oesophageal reflux and vomiting.

[16] Naso-pharyngeal airway (NPA) – this is a tube which is inserted into the nostril and reaches the pharyngeal space. It keeps the airway open and is often used for children who have partially obstructed airways, as a way of maintaining airway patency.

4.2.9. Hassan's Case

Case composition and features

Name	Relationship	Name	Relationship
Salma	Mother	***Caitlyn***	Long Term Ventilation Nurse Specialist
Ali	Father	***Mr Abraham***	Paediatric Surgeon
Fatima	Sister	***Lucy***	Case Manager – Continuing Care
Dr Martin	Respiratory Consultant	***Mrs Yannis***	Ophthalmology Surgeon
Mr Field	ENT Surgeon	***Anna***	Family Support Worker
Dr Tring	Paediatrician	***Jane***	Dietician
Sara	Long Term Ventilation Nurse Specialist		



Case context

Hassan is a 1 year old boy with conditions classified as 'congenital malformations, deformations and chromosomal abnormalities'. His syndromes mean that he has a variety of health complications and is at risk of life-threatening complications, and therefore classified as having a life-limiting

condition under category 4. Hassan was in hospital from birth until he was almost a year old, but he now lives at home, with his parents, Salma and Ali, and his older sister, Fatima, who also has severe behavioural issues. The family have two cars and both parents can drive. The family are Muslim and although Salma does not identify as very religious, she was observed to be praying for Hassan when he was distressed whilst having his bloods taken, suggesting that religion is important to her. Salma stated that religion is incredibly important to Ali and that he spends lots of time at the Mosque and at prayer, and had consulted religious scholars regarding the possible decision to terminate the pregnancy. Ali works full time in the retail sector and Salma is a full time carer for Hassan.

The family have some social support from Ali's family, who live locally, although the extent to which they are able to rely on the extended family for care is minimal, due to the extensive needs of both children. Salma identified that she is estranged from her own family due to their opinion that she should have had a termination and that her children brought shame on their family. Salma identified that she had severed most contact with them as she did not wish to have such negativity in her life or to hear such criticism of her children. It appears that outside of the paternal extended family and the religious community, the family receive little support from a social perspective. Due to Hassan's dependency, he has a care package which provides carers in the family home for most of the day and seven nights a week. Due to his sister's severe behavioural issues, the package aims to ensure that neither parent is ever alone with both of the children at the same time. Most of the support that Salma accessed in relation to care was from paid carers who staffed the care package and medical professionals involved in Hassan's care. Like many of the other parents of children with rare conditions in this study, Salma had educated herself about the condition, so that she was able to prepare herself for the future possible treatment options, although she did not identify that she was part of any social networks for support or information.

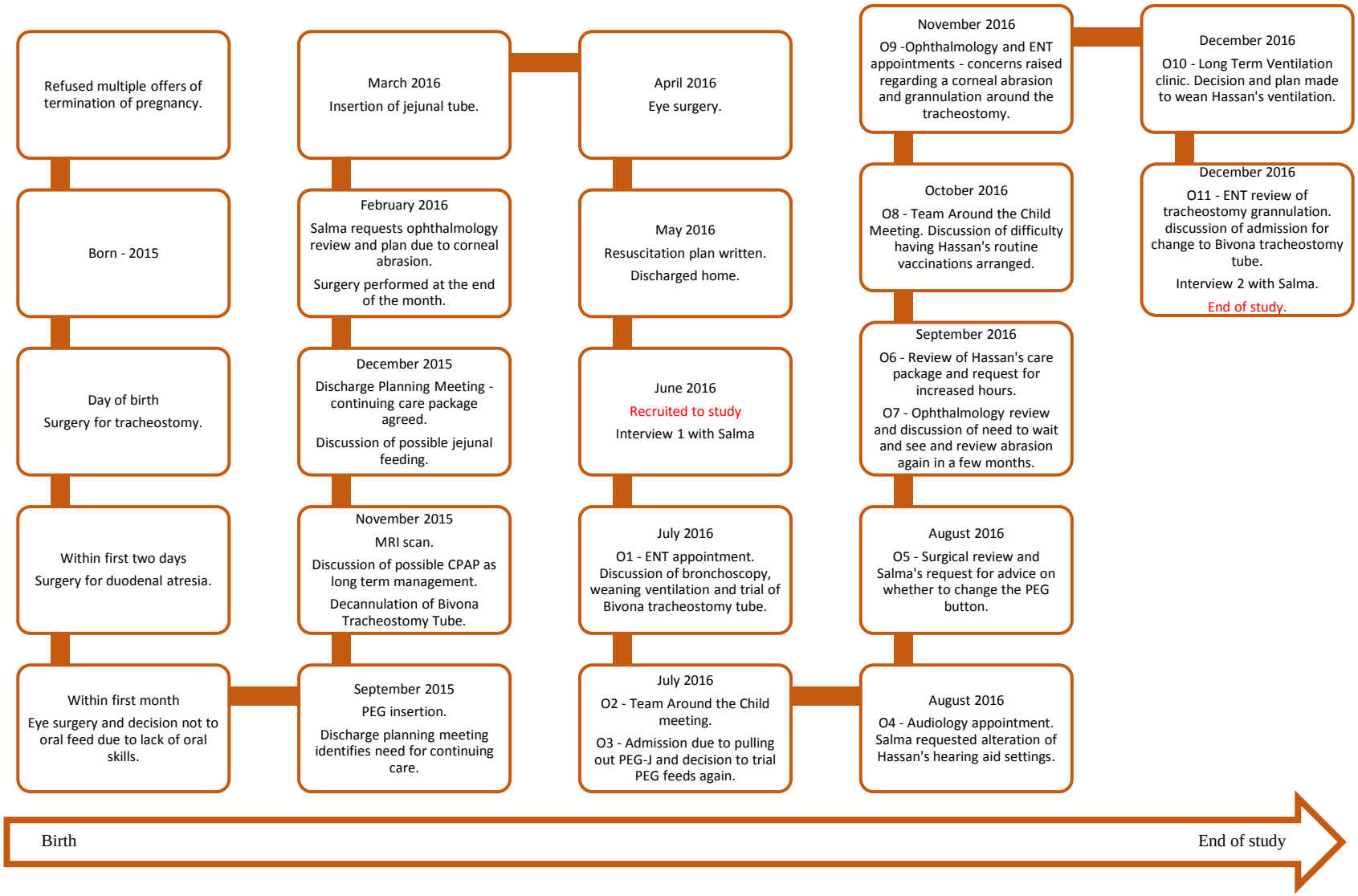
Salma did not identify any significant others to participate in interviews and both interviews with her were conducted in the family home. Observations were conducted in a variety of clinic rooms at the hospital, in outpatient departments, the high dependency unit and in the family home.

Important decisions

Salma identified that she was required to make important decisions prior to Hassan's birth, as she was repeatedly offered, and repeatedly turned down, termination of her pregnancy. Salma explained her philosophy that every living creature has the right to life and that she believed in her ability to be a mother to Hassan, despite his extensive needs. Decisions that were necessary shortly after birth, included the decision to agree to a tracheostomy[17] and gastrointestinal surgery. Since then decisions regarding Hassan's care have included the commencement of full time CPAP[18] and continuous jejunal[19] feeding, meaning that Hassan is highly dependent on medical intervention. Hassan's dependency meant that the family would need support to care for Hassan in their home and were subsequently presented with decisions surrounding his care package. These decisions were ongoing during the study period. Hassan's condition means that he has significant facial disfigurement, which required corrective surgery and will present the possibility for further surgery as he grows, both for functional and aesthetic purposes. Ongoing decisions throughout the study also included weaning of Hassan's ventilation and the possibility of changing his tracheostomy tube to a different type of tube.

[17] Tracheostomy – a hole created through the front of the neck into the trachea, through which a tube is inserted and secured, which assists with breathing in people with anatomical abnormalities or neurological issues meaning that they are unable to protect their own airway.
[18] Continuous Positive Airway Pressure – a method of ventilation in which positive air pressure is delivered. In contrast to Charlotte's ventilation (non-invasive ventilation), Hassan's ventilation was via a tracheostomy (a surgically created artificial airway) and is therefore considered as invasive ventilation.
[19] Jejunal feeding – this can be via a naso-jejunal tube (inserted through the nostril, into the stomach and through into the jejunum) or through the abdomen into the jejunum (often referred to as a PEG-J). It is used when the child cannot tolerate feeding into their stomach and experiences frequent vomiting or delayed gastric emptying (when their stomach does not pass the feed through into the intestines quick enough, meaning that the stomach gets too full).

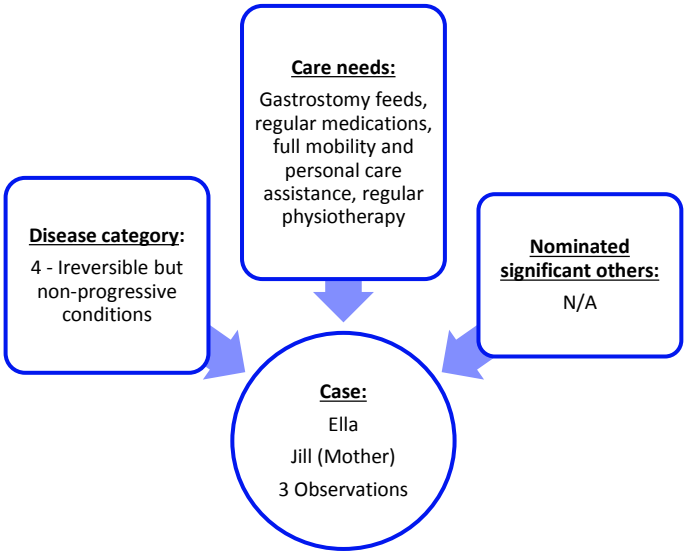
Timeline 9: Hassan's Case



4.2.10. Ella's Case

Case composition and features

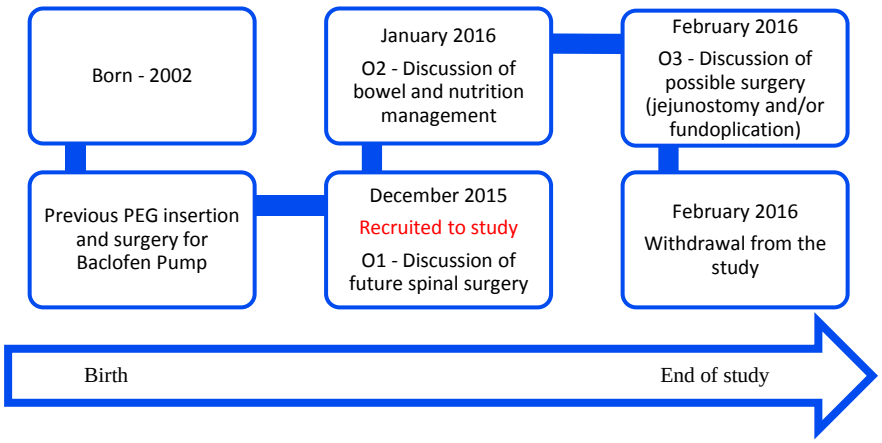
Name	Relationship
Jill	Mother
Dr Price	Paediatrician
Mr Machin	Spinal Surgeon
Mr Brown	Paediatric Surgeon



Case context

Ella is a 13 year old girl with severe Cerebral Palsy. She lives at home with her mother, Jill and her father. She is fully gastrostomy fed, although is allowed 'tastes' orally and requires assistance with all aspects of daily life. I attended three clinic appointments for Ella, but following the third observation prior to being able to arrange an interview with Jill, I received an email to say that the family wished to withdraw from the study. As Jill had not been interviewed prior to withdrawing from the study, it was therefore not possible to gain a detailed understanding about the family context, values or religion.

Timeline 10: Ella's Case



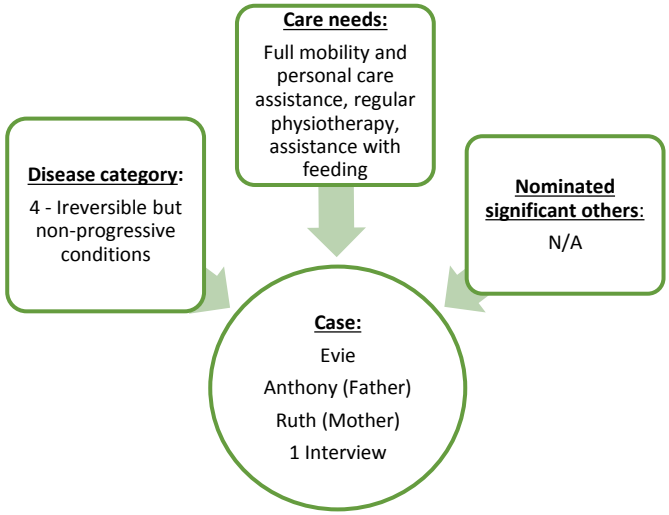
Important decisions

The three appointments observed signalled two major and challenging decisions for Ella: scoliosis surgery and gastrointestinal surgery. It appeared that these decisions would be ongoing and considered over the coming months and years. Another minor decision, albeit one which had a significant impact on her life was in relation to management of her chronic constipation.

4.2.12. Evie's Case

Significant people

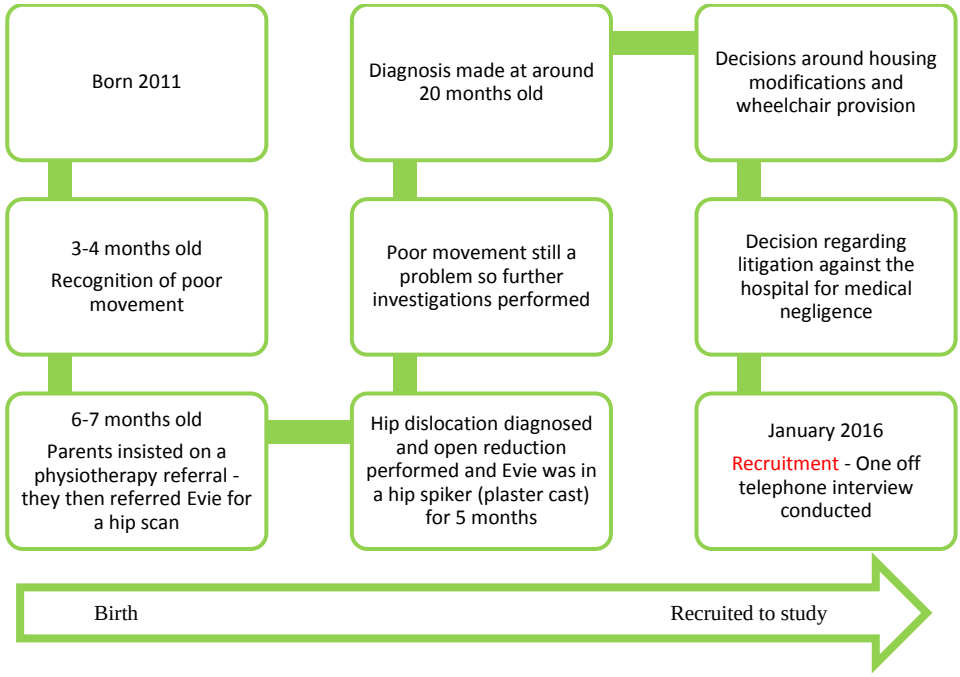
Name	Relationship
Anthony	Father
Ruth	Mother
Dr Brett	Paediatrician
Dr Wilkin	Paediatric Neurologist



Case context

Evie is a 4 year old girl with Muscular Dystrophy. She lives at home with her Mum, Dad and older brother. She attends the local primary school and is described by her Dad as funny, intelligent and a joy to be around. The family appear well educated and self-identify as such. Anthony works full time in the Technology sector. Ruth does not work outside the home, although Anthony identified that this is more than a full time role in relation to Evie. The family opted to take part in the study through a single telephone interview, stating that they did not feel that it would be appropriate to bring yet another person into Evie's life over an extended time period, as she already had contact with upwards of 25 professionals. It appeared that the family had an agenda for the interview and wished to provide some context and understanding about the difficulties that they have faced in caring for and making decisions about Evie. Religion was not discussed within the interview and therefore the family's religious beliefs are not known.

Timeline 11: Evie's Case



Important decisions

The main decisions which were discussed regarding Evie were those regarding housing and equipment. The family stated that the social decisions are often harder to make, as there are many more factors to take into consideration than for purely medical decisions. Evie required hip surgery, for a dislocation in infancy and her parents identified that they had taken the decision to pursue litigation against the hospital for failing to recognise and act on their concerns surrounding her general mobility around that time. Anthony stated that litigation was something that the family did not take lightly, but felt they needed to do as the hospital had not adequately taken responsibility or apologised for their mistake. They also discussed their awareness that in the future they may need to consider non-invasive ventilation and tube feeding, but did not discuss their opinions regarding decision making for these issues, as they were likely to only present themselves many years in the future.

4.3. Conclusion

This chapter has provided an overview of the eleven cases included in this study. It has demonstrated the complexity and variety of data collected within each case in order to demonstrate the unique contexts within which decision making operates. Although Evie's case was formed of less data, it still provided some powerful insights into the wider contexts of decision making and also illuminated how social decisions may be more problematic than medical ones, as they impact not only the ill child, but the siblings and wider family also. This chapter provides the context within which the data in the following chapters were collected and analysed.

Chapter 5. Findings Theme 1 – Communication and relationships

5.1. Preface to the findings chapters

The following three chapters will present the major findings from this study. The major themes have been identified through cross-case analysis (see Appendix 7) and represent the most significant aspects of decision making identified within the data. The importance of communication and relationships to the process of decision making was highlighted and is the focus of Chapter 5. Chapter 6 goes on to consider the process of decision making, with particular focus on decisional responsibility and the strategies used to reach agreement. Chapter 7 considers the issue of protracted disagreements and the role of best interests' discussions, with reference to possible court involvement. These chapters, although discussing separate and distinct themes, overlap significantly. For example, communication and relationships were identified as relevant to the process of decision making and the strategies involved in decision making required communication and took place within the family-professional relationship. Additionally the concept of best interests, although relevant to any decision making, has been presented in a separate chapter, as the terminology of 'best interests' was primarily used during protracted disagreements where there was concern about harm and when the possibility of court intervention was made. It was therefore deemed important for this theme to be presented in its own right, separate from the other data.

5.2. Introduction to communication and relationships

This chapter will explore the relational aspects of decision making, providing an understanding of the importance of relationships and communication in decision making. The relationships between stakeholders[20] were identified as being important in decision making, with good relationships facilitating decision making, and poor relationships acting as a barrier to the process. Communication was also essential in the process of decision making and was inextricably linked

[20] Throughout this thesis, the term 'stakeholder' refers to anyone involved in the decision making process within a case.

to relationships, with the quality of communication impacting on the quality of relationships and vice versa.

5.3. Communication

Communication was identified as being important in the development and maintenance of good relationships and conversely, poor communication resulted in damage to relationships. Communication played an essential role in decision making, as information was shared between stakeholders. Communication between professionals and families, and within the medical team, were identified as having the most significant impact on decision making.

5.3.1. Professional-family communication

Good communication

Good communication between families and professionals was identified to occur within every case. Predominantly families felt that they had good access to the professionals involved in their care and were able to contact them when necessary to obtain advice. Parents inferred that such availability was important and appeared to act as a source of support and reassurance for them, and professionals demonstrated their awareness that families needed this support. They were particularly aware of this when the child's health had recently deteriorated, or the family were having to deal with challenging and distressing symptoms, causing parental anxiety or making such anxiety likely. Dr Brett articulated that ensuring that she was available to provide support was often a challenge and required discussion out of hours, stating:

> *"It's quite hard work and there's quite a lot of negotiation and conversations going on so I often end up speaking to her late at night when I get round to it"* [C:Christopher, I2, Dr Brett, L:186[21]].

The availability of such channels of communication, particularly when it was perceived by the family to be above and beyond what would be expected of the professional, contributed to the development of trusting relationships. Families appeared grateful for and valued the attention to, and recognition of, their concerns from professionals, as alluded to by Rachel, who said:

[21]C = Case; I = Interview number; O = Observation number; L = Line in the transcript.

> *"So I've never been made to feel that I'm being an overcautious mother"* [C:Charlotte, I2, Rachel, L:483].

Access to the professionals for advice, including the availability of this 'out of hours', was a significant feature contributing to the perception of good communication. The provision of sufficient time to discuss the family's concerns and ensure that all questions were answered was another important factor influencing the quality of relationships. Waiting for appointments was a significant feature of the data collection period and observed to occur in all cases. Although there were occasional complaints about the frustration of waiting and the lack of communication regarding how long the wait would be, it was mostly forgiven. Parents felt that waiting meant that another family was in need of extra time and attention, a position that they could empathise with or had been in previously. Pauline identified:

> *"she listens…and I don't think she'd actually leave me without, if she needed to get her point across, I mean I've waited, I've had an appointment at 2 and not gone in until 5 o'clock with her, what I don't mind…cos I've spent 2 hours with her before. And I don't think she'd ever leave you going out upset or not understanding"* [C:Elena, I1, Pauline, L:392].

Waiting times were often over an hour long[22] but when it was an appointment with a trusted professional, the communication accessed was described as being worth the wait, suggesting the huge degree of value that parents placed on good communication and listening.

As well as the provision of sufficient time to discuss concerns, listening was a feature of relationships which was explicitly identified as valuable by Pauline, with reference to Joanna the physiotherapist:

> *"she'll just come up with what, any experience she's had or find out how I could go and find stuff so it's just, she just gives me all the options and stuff, or just lets me talk through it to be honest, I mean*

[22] For one particular paediatrician, it was unusual to be seen with less than an hour wait. The longest wait experienced during the study was in Hassan's case and was approximately 2 hours and 20 minutes long.

just hearing your own voice normally you come out with what you wanna do by the end of it" [C:Elena, I2, Pauline, L:289].

Pauline's explanation of the importance of providing time and listening was described to be partly in relation to discussing previous experience, but also about being provided with a safe place to talk through concerns and undertake the process of personal decision making.

Listening and being observant was also an important feature of high-quality appointments, which was both identified by the families during interviews and through my own observations. An example where this was explicitly demonstrated was in Michael's case. At the first observation, an appointment with Dr Price, it was highlighted that at the next appointment there would have to be a further discussion of the need for a gastrostomy. However, at the following appointment this discussion was not had. It appeared that the reason for this was that Michael had not been weighed or had his height measured, and Kate led the direction of the appointment, standing up and walking to the door to leave when she had addressed all of her concerns. Although Dr Price had not been nominated as a significant other in Michael's case and therefore her motivations for not discussing the gastrostomy at that time could not be verified, it appeared that she recognised the non-verbal cues which suggested that such discussion at that time may have been counterproductive.

This demonstrates the importance of detailed verbal communication in the process of decision making, but also the importance of detailed attention to non-verbal signs, which may indicate whether difficult discussions regarding decision making are likely to be productive.

Poor communication

Although effective communication was observed and/or reported to occur in most professional-parent relationships in all cases, poor communication in the professional-parent relationship was also identified in six of the cases. For Ella, the three observed appointments demonstrated the importance of listening and understanding non-verbal cues, and the inadequate communication which resulted if these were missing. A two way dialogue of information seeking and information provision, and some reference to the parental preference appeared

necessary for good communication, in addition to professional awareness of and response to any non-verbal signs of parental hesitation or concern. A good example in Ella's case was when Jill's responses to the surgeon demonstrated to him that she was unsure about the best option for Ella. He subsequently validated her apparent feeling that this decision would be a difficult one, suggesting that the family take some time to consider the options. In contrast, during Ella's spinal appointment, although stating a commitment to involving the family in decision making and enabling them to provide some information about Ella, it was observed that Mr Machin failed to recognise or respond to Jill's non-verbal indication that she required further information. At the end of the appointment Mr Machin narrated the clinic letter in front of Jill, providing important information, but failing to recognise Jill's expression which demonstrated concern about what was being said and her need for further explanation, as outlined in the field notes:

> *"During the narration on the Dictaphone, Mr Machin mentioned the baclofen pump and then realised that he had not adequately explained something to Jill. He paused the Dictaphone and explained, very briefly and not very clearly, that doing scoliosis surgery can cause a failure in the baclofen pump. He then continued narrating and finished dictating the letter…In the waiting room when putting on Ella's coat, it became clear that Jill did not know that the surgery could impact on the baclofen pump and that this knowledge would mean that she would resist the surgery, wherever possible, as the pump had been so beneficial"* [C:Ella, O1, L:67].

It appeared that the communication in this instance, particularly in relation to picking up on non-verbal cues, had not been as good as in the other appointments for Ella. As a result, despite a lack of information about the risks and benefits, it appeared that Jill had quickly arrived at a perception that the scoliosis surgery may have negative effects for Ella, and therefore struggle to make this decision in the future. This rushed communication and lack of recognition of Jill's concern, appeared likely to result in difficulties decision making regarding this surgery in the future.

Inattention to verbal and non-verbal cues were also observed to contribute to poor awareness of parental preference and poor communication in Maisie's case. Fiona felt that she did not have sufficient access to the cardiology team and that she was constantly having to chase them to see if any other intervention could be offered to Maisie. Despite this, Dr Chase appeared to be unaware that Fiona was keen for further investigation or intervention, as she said:

> *"I never got the feeling from her she was desperate for something to be done straight away and that she was very practical in terms of arranging procedures around sort of family occasions as well. So we gave them I thought, space to, space to enjoy Maisie's life as well"* [C:Maisie, I2, Dr Chase, L:86].

For Maisie, her poor prognosis at birth may have led Dr Chase to believe that her family's primary focus was on enjoying a good quality of life, rather than pursuing a potentially risky intervention. However, the poor communication appeared to maintain this perception, rather than enabling an awareness that Fiona viewed the communication between them as poor. Fiona said:

> *"I bet [Hospital 2] liaison team are sick of hearing my voice on that voicemail, I'm like "rar rar rar" and then they pass me on to Dr Chase's secretary, then the poor woman she gets it, but it is bad communication, when even if Dr Chase can't ring me, get somebody else to give you a call…I think it's poor communication, really poor because I'm forever chasing and then you're forever waiting for a phone call back…So everything's you wait wait wait, decision. They just spring things on you like you've got nothing else better to do"* [C:Maisie, I3, Fiona, L:323].

Poor communication in the family-professional relationship was also observed to occur in the opposite direction in Grace's case, with professionals perceiving it to be difficult to communicate with Kay. This was a unique case, as although some observational data suggested that professionals at times struggled to effectively communicate with families in the other cases, owing to emotional factors and limits to their technical understanding, on the whole there was little mention of the difficulties they had in communicating with families more

generally. In Grace's case, communicating with Kay however, was observed and explicitly identified as being challenging. Kay had an extremely poor relationship with Grace's social worker, Glenn, which resulted from events at the beginning of their professional relationship in which he was involved in forcing Kay to comply with the medical plan that she could no longer feed Grace orally. This relationship was the only one in which communication was directly observed to be particularly problematic, however reference to Grace's medical notes provided evidence of other professionals struggling to communicate effectively with Kay. The below extract from a clinic letter demonstrates the difficulty that one of the neurosurgeons had in communicating and making decisions with Kay about the possible use of Intrathecal Baclofen for Grace. The manner in which the letter was written suggests significant frustration and the feeling that the procedure would not be appropriate for Grace as a result of Kay's behaviour, as outlined by the following extract:

Thank you for referring Grace to my Assessment Clinic. I saw Grace with her mother, who is a person who is difficult to engage and for some reason had been told she was coming in to stay a few days. She then largely complained about how much she had packed, rather than focusing on Grace Grace is a severely disabled child with significant contractures at 10 years old. Grace's mother's main concern is her left hip, which is said to be out of joint. Grace does not have a gastrostomy, but is under weight and very fragile. The mother was suspecting that she actually had a chest infection on the day.

Grace's mother had reviewed the DVD and immediately told me she didn't want anything to happen to her Princess. I explained the Baclofen pump procedure again and mentioned the complications and the overheads of refill and, as mentioned previously, Grace's mother was difficult to really engage as she was too upset. Ultimately, we agreed to do a test procedure, which I will book in the near future, and we can discuss matters again.

My personal feeling is that although Grace is a fair candidate for intrathecal Baclofen, she is very frail and likely to be prone to complications and her mother is not the ideal parent to be burdened with intrathecal Baclofen. She is very worried and was already asking about the emergency numbers she could contact in case of mishap. We will come to a final decision after the test procedure.

[C:Grace, Clinic Letter from the Neurosurgeon to Dr Price, 25/03/2015].

This difficulty in communicating with Kay was identified by Dr Price, who demonstrated an understanding of why other professionals may struggle with making decisions with her, saying:

> *"it is so helpful to have a Key Worker or communication facilitator or somebody who's able to sit and get to know them, a mummy like this very well, because communication is quite difficult, with lots of my patients, but I think you could very easily as a doctor not get very far with Kay because you are often the first, the stone wall, you feel*

> *like you're not getting anywhere and I think a lot of people might just shrug and walk off and think "well you can't actually get, make any decisions together with this mummy because she's not listening, she's not understanding, she's not willing to see anybody else's view or listen to the medical issues""* [C:Grace, I2, Dr Price, L:191].

Dr Price's quotation seems to outline the reasons for the difficulty that the neurosurgeon had in his communication with Kay. It highlighted that this signals to the importance of having a key worker, or someone to have a good relationship with her to assist the process of communication and decision making. Dr Price's quote highlights how some professionals may "walk off" and not engage Kay in the process of decision making for a procedure which may be in Grace's best interests, due to the challenges that doing so may pose. This appears to have been the case regarding the intrathecal Baclofen pump, where Grace's health and best interests may not have been optimised, due to the challenges encountered by the surgeon in communicating and making decisions with Kay.

5.3.2. Inter-professional communication

Poor inter-professional communication was identified in five of the eleven cases studied. It was highlighted as a significant concern both for the child's care and for decision making regarding the most appropriate treatment. Ten of the 11 cases were cared for under multiple speciality teams, as can be seen from the case composition tables in Chapter 4. Specialist teams were often based at different organisations than the child's primary hospital and this posed challenges to information sharing between these teams, particularly because effective communication systems between organisations did not exist. Both families and professionals expressed concerns regarding the implications of lack of communication between teams, and these concerns ranged from not being able to provide a full account of the options available, to concerns for the child's health and safety if information was miscommunicated.

This lack of information sharing and communication between professionals often resulted in parents being forced to take on the role of communicator, something which they felt was inappropriate and posed risks to their child's care. For Pauline, even within one organisation, she felt that the professionals were

not communicating together, rather they were communicating through the family about the plan for Elena's upcoming surgeries. She highlighted:

> *"we've got Mr Kelly pencilled in to do her stomach PEG, so we've got two surgeries coming up so they don't know which one is coming first and we don't know how Elena is going to tolerate and I just think, I think all three should be talking. Cos the only person, cos I'm going have to go in there and explain everything to both surgeons and I'd like to know which one should come first, rather than which one comes first"* [C:Elena, I3, Pauline, L:31].

Pauline had demonstrated concern during a clinic appointment that she wanted to know which surgery, the scoliosis surgery or the gastrostomy insertion, would be better for Elena to have first. However, due to the lack of communication between the teams, Pauline felt that she was lacking information about which surgery would be beneficial to come first and that the situation would result in Elena being booked for the first available surgery, rather than the one which would have the better outcome for her. This case exemplified how decision making became more troublesome when professionals did not communicate with each other and therefore failed to provide advice to the family. Pauline's assertion also demonstrated the additional burden on the family to communicate between teams regarding the plan of care which resulted from poor inter-professional communication. Frustration was also expressed about the problems with electronic systems for information sharing within individual organisations and that these were perceived to be insufficient to ensure effective communication. However, inter-professional communication between different organisations was often the primary concern. All five of the cases in which poor inter-professional communication was identified had received care from two or more NHS Trusts, as well as other community based or third sector organisations.

The primary mechanism of inter-professional communication appeared to be the clinic letter, which was used to share information from appointments and investigations with other professionals involved in the child's care. This was observed to be a sub-optimal mechanism of effectively communicating

discussions, given that a brief A4 letter would infrequently be able to capture the detail of discussions that professionals conducted with the child and family during extended consultations. This was illustrated in Elena's case where during a clinic appointment with Dr Price, reference to discussions at a previous appointment with Dr Ludd, was made. The following extract from my field notes signalled that the clinic letter sent to Dr Price by Dr Ludd was not sufficient to clarify to Dr Price the content of their discussion. Pauline and Richard provided an account of what had been discussed, but given the lack of detail in the clinic letter it was not clear to Dr Price whether they had misunderstood anything or forgotten important information. Dr Price identified that many of the things that Pauline said were discussed, had either not been documented in the clinic letter, or were articulated differently than Pauline suggested:

> *"Dr Price asked about the spinal brace, saying that Dr Ludd hadn't written about this in the letter…Dr Price asked if Dr Ludd had mentioned the issue with the General Anaesthetic (GA), which Pauline said he did and Richard told Pauline to get out the fact sheet that they had about this…Dr Price then said that she doesn't know where the concern about the GA has come from and that she has not seen what evidence is behind that, which she identified concerns her slightly. She continued to say that Dr Ludd in the letter hasn't said that a GA is a problem, rather that the parents are concerned about a GA"* [C:Elena, O5, L:253].

This extract demonstrates that the issues which Pauline and Richard felt that they had been advised about by Dr Ludd, had not been sufficiently communicated in the clinic letter, to enable adequate shared understanding. This resulted in difficulty with the further provision of advice, as Dr Price was unable to identify whether Elena's parents had misunderstood the information from Dr Ludd, or whether the clinic letter just failed to document their discussions adequately. In order to provide information to the family to enable them to make informed decisions regarding spinal bracing and any possible surgery, clarification regarding the specialist opinion on the matter would be useful, but would be likely to fall to Dr Price to seek out. An appointment two

months later seemed to demonstrate that such clarification had not been obtained, providing further testament to the lack of communication between professionals at different organisations.

For Charlotte, the issue of inter-professional communication between separate organisations had become a significant concern. The family expressed concern that they perceived a fundamental disconnection between the different teams and that instead of them viewing themselves as Charlotte's team, they saw themselves as distinct teams, working at different organisations and therefore treating each other as such. The consequence of this was that communication between them was poor and the family were tasked to act as the bridge in communication, as explained by Jamie:

> *"We've seen at times where they've almost asked us to take that role proactively right and said "well we expect you to be doing this" sort of thing and then I've been given tasks to go and facilitate communication, that's not OK. I mean acting as a back stop is clearly you've got a role to play and acting as a back stop is OK, but this sort of proactive "well you guys kinda create a bridge" is not, that's just not OK"* [C:Charlotte, I3, Charlotte, Jamie and Rachel, L:719].

Being asked to create a bridge in communication was something which professionals seemed to expect in a number of cases, as a way of saving time and gaining quicker access to the information by asking parents, despite the possible implications relating to the accurateness of the information provided. Similar reference to parents as a bridge in communication was identified in Evie's case at her diagnosis. When the Neurologist met the family for the first time and disclosed Evie's life-limiting diagnosis, she did not have all the information relating to Evie and asked her parents to contact the other hospital in order to obtain it:

> *"Anthony: She'd clearly not read the notes or read Evie's red book because she ended up phoning up [Hospital 2] to try and get information*
>
> *Ruth: She got me on the phone to them*

Anthony: Yeah she got Ruth on the phone to [Hospital 2] to try and get information" [C:Evie, I1, Anthony and Ruth, L:253].

Ruth and Anthony explained this encounter in a way which suggested that over two years later, they were still shocked about how the communication had been handled. Ruth's interjection that the Neurologist asked her to contact Hospital 2, rather than doing it herself, suggests that this was something which caused her concern, perceiving that she had been placed as the lynchpin in communication between the two hospitals. It appeared that this experience was instrumental in damaging the relationship between the family and the Neurologist.

The expectation that families acted as facilitators of communication between different teams or organisations was identified in five cases, however it was only identified as a significant concern in four of them. However, in Christopher's case, it was not something that the family highlighted concern about or felt was significant. An example of this expectation occurred during an appointment with Dr Brett where she suggested that Susan could help relay information from Hospital 1 to the specialist nurse when she was in contact with her next:

"Susan asked whether Dr Brett would find out what was said at the appointment with Dr Martin tomorrow and Dr Brett said that usually the communication with Dr Martin is not great and that the system at Hospital 1 is quite slow. The Specialist Nurse told Susan that she would need to speak to her next week anyway about when Christopher's PEG button needs changing so Susan can update her then" [C:Christopher, O2, L:260].

Susan accepted this request without challenge and without any signs that she did not feel that this was her role. Susan's acceptance of this role may be related to Dr Brett's complete honesty with her about the poor systems of communication, the availability of her advice out of hours and her perception that acting as the communicator was in Christopher's best interests. Given their good relationship, it may also be that Susan perceived the request from Dr Brett

as arising from a true concern for Christopher's health, rather than a lack of effort.

Dr Brett's acknowledgement regarding the poor systems of communication was also echoed by Dr Price who found the poor communication between teams frustrating. At times she appeared to suggest that the onus for communication was on her, as the lead professional, but when other professionals failed to include her in correspondence, her frustration and offence at this appeared to translate into further breakdown in channels of communication. Service constraints in terms of time were identified as being significant barriers to communication between teams, meaning that writing emails was problematic, let alone trying to organise multi-professional meetings. However, this lack of communication resulted in issues around the provision of choice to families, as Dr Price identified:

> *"You can't on your own share the choices, because they can't say what can be done in the community, they don't know. They can't do a, I can't do, none of us individually can show the full menu to the family. They've got half the menu and I've got half the menu. And maybe Mum wants a bit of this and a bit of that and a bit of the other. So we're showing the family the options and the choices separately and that isn't the full a la carte. So we have to be able to speak to the family together"* [C:Maisie, I2, Dr Price, L:126].

Additionally and perhaps the most concerning consequence of the lack of communication for families was their perception that this presented a potential risk to their child's care. This was something that Rachel was particularly concerned about in relation to Charlotte's care, saying:

> *"And I just dread something very important being missed or miscommunicated at some point, we're OK now, but you never know"* [C:Charlotte, I2, Charlotte and Rachel, L:457]

The emotional stress that this placed on the family to communicate effectively and accurately in order to make decisions, represented a significant burden which was observed for the cases in which poor inter-professional communication was identified.

5.4. Relationships

5.4.1. Trust

Parental trust in professionals

The importance of trust was explicitly identified in nine of the cases and was inferred during interviews or observed during appointments in the other two cases. Predominantly trust in professionals was something that developed over time and was based on previous experience of working with them. It appeared to be something that was built upon the foundations of good communication and professional knowledge as articulated by Kate:

> *"we've got Dr Price which is amazing cos I think the first time we went in to see her she was just chatting away to me and then she spoke about Michael to me and I thought "how do you know all this?" and it's cos she literally observed him in her room and I was like, she's brilliant. She was fantastic, I felt really comfortable straight away because I just thought she knows her stuff, she'd only watched him for 5 minutes and she told me stuff I didn't know and he was my child, so I was happy there"* [C:Michael, I1, Kate, L:164].

Although the first encounter appeared to be instrumental in whether trust in the professional would be developed, it was not a foregone conclusion. Rather Catherine described it in a way which suggested that it had been earned:

> *"Mainly it's just something that's been built up. We've never thought they've made a wrong decision, we've never thought that were a waste of time, that didn't work or that made her ill or, it's always I suppose, I suppose trust is something we've built, it builds up and I think every time it's worked out…it's just something that's just built up and now we're just, rely on them I suppose to give us the right decision"* [C:Daisy, Catherine, I2, L:266].

The data demonstrated that ongoing relationships allowed for the development of trust which assisted in decision making, but the observation of the initial formation of such relationships was not facilitated within this study, as most of the cases involved pre-established relationships. The one example where the development of a new relationship was observed was in Elena's case, where a

new physiotherapist was introduced, due to Elena moving school. It was evident that trust had not yet been established within this relationship and appeared that its development may be challenging due to a number of difficult discussions that were had when she was first introduced, and the close relationship that Pauline had with Joanna, Elena's previous physiotherapist. Pauline said:

> *"I did come home that night and I was a bit upset with Richard that she'd [Polly] actually talked to Dr Price about my daughter without discussing it with me, but I don't know whether that was the dynamics I had with Joanna and I didn't know who this. I mean Polly I think is lovely, but I was just a bit, that I got told that Dr Price has OK'd it and. I'm sure a lot of that goes behind my back and sometimes you feel that they're all making decisions behind your back without being involved. And then on Thursday I saw her and she said "are you alright for the appointment tomorrow, are you feeling alright about it?" and I went "yeah" and she went "alright then, am I still alright coming?" and I went "yeah" and that's all I knew, so what she said, and I couldn't hear her because she spoke so quietly, it's all new to us"* [C:Elena, I3, Pauline, L:176].

Pauline alluded to the difficulty of establishing a trusting relationship with a new professional in cases where they are replacing a professional with whom they had had a close relationship and within which there had been different dynamics. The way that Pauline described this situation also highlighted the importance of straightforward communication and ensuring understanding in relation to developing trust. Pauline did not request clarification about what had been said by Polly at the appointment described, despite not having heard what she said and this appeared to further contribute to her feeling that things had been discussed without her knowledge and may result in the delayed formation of a trusting relationship.

Although Elena's case was the only one in which the formation of a new professional relationship was observed, interviews provided some understanding of the factors which had facilitated the development of trust in the other cases.

For example, Fiona developed trust in Dr Alexis through experience, observations of other patients' outcomes and reference to the internet. She said:

> "*I felt that I trusted it and I could only watch what was happening in my time on [Ward] at [Hospital 2]. So based on that, of me watching and him as a surgeon and googling him, I felt I trusted him and he made my decision*" [C:Maisie, I1, Fiona, L:147].

Fiona's trust in Dr Alexis was something that appeared to grow and develop throughout the account, with increasingly more emphasis placed on the trust that she had in him. Given that Maisie had vastly exceeded her prognosis, this may have promoted the further development of the trust that Fiona had in Dr Alexis, as he had performed the initial surgery, when other cardiologists had said that nothing should be done. His offer to perform a second surgery, again despite the consensus previously having been that nothing more should be done, appeared to have appealed to Fiona's need to maintain hope and therefore strengthened the trust that she had in him. This demonstrates the importance of trust when making uncertain and difficult decisions; it appeared to be an essential component of relationships and highly influential on decision making.

Despite the majority of families identifying trust as a feature of their relationships which had to be earned, in Omar's case, it appeared that having trust in professionals was the family's predisposition, something which Nadia articulated during an observation:

> *"Nadia said that she always agrees with what the doctors recommend, as they know what will be best"* [C:Omar, O7, L:469].

For Nadia, rather than building trust through experience, trust pre-existed the relationship and experience served to either solidify or degrade this trust.

Trust within the family

Similarly within families, trust in relation to decision making was either built or damaged over time. Mothers frequently made decisions without reference to the child's father when the decision was one which they perceived was not particularly significant, or had become part of their new normal and integrated into their responsibility as the mother of a child with complex needs.

Mothers were the principal the decision makers within the family regarding the child's care. In all but one case, fathers either explicitly identified that they were happy with the mother taking the primary decision making role, or were observed to voluntarily defer to the mother's decision, indicating a trust in their decisions. Steve said:

> *"Obviously her Mum…she know what she's doing. I kind of leave it to her in the end, I can never win an argument so"* [C:Daisy, I1, Catherine and Steve, L:344].

Mothers also highlighted that the fathers had developed trust in them through their expertise and detailed knowledge of the child. Nadia stated that although sometimes Jamal disagreed with her decisions, he deferred to them, through reference to her greater expertise in Omar's care and therefore trust in what she thought would be best. Nadia explained:

> *"before we used to clash a lot in the beginning, but as times has gone on and Dad has realised that he's not here 24/7 with Omar, so I do know Omar a bit more than he does, he will say "whatever you think is best""* [C:Omar, I3, Nadia, L:230].

For big decisions, however, mothers stated that they would try to involve the father, but often signified that their own opinion may hold more weight due to a greater trust in their own opinion than the father's opinion. For Hassan, Salma appeared to feel that Ali's decision making in relation to weaning his ventilation would directly put Hassan's health at risk and therefore was prepared to openly overrule him. She explained:

> "*Dad was like "give him 20 minutes no one will know" and I was like "we can't do that because that's dangerous for his health", I would not do something that would you know, and he was like "he'll be fine" and I said "no he won't" and that's why I'm in charge of this weaning programme, not the carers, not Dad, but me, because I, call it my maternal instinct, but I know when it's going to be pushing him too much"* [C:Hassan, I2, Salma, L:288].

This decision regarding weaning Hassan's ventilation seemed to demonstrate some degree of damage to the trust that Salma had in Ali's decision making capacity. Following this episode, Salma described herself as the main decision maker, rather than as she had done before, referring to both her and Ali as the decision makers for Hassan's care.

The deviant case in relation to parental decision making role was Charlotte's case. Hers was the only case in which the parents had separated, but the child still had contact with the father. In her case, it appeared that Jamie's opinion held more weight than Rachel's in relation to decision making and although the family described that they generally agreed on Charlotte's treatment and care, they agreed that Jamie's forceful personality meant that his opinions were often the ones that were trusted and implemented. Rachel articulated:

> *"Well often Charlotte's Dad is the strongest voice on, he's the one who questions and doesn't um just go with the flow…We tend to agree with him don't we?"* [C:Charlotte, I2, Charlotte and Rachel, L:53].

It appeared that for many decisions, Rachel trusted Jamie's opinion, because of his technical understanding of some of the concepts involved, but at other times, it appeared that she did not trust his decision, but was unable to overrule him, or was required to refer to the professionals to back up her opinion.

Inter-professional trust

The final component of trust, was in relation to professional trust of their colleagues. This was something which was implicit when they referred a child to a specialist and subsequently trusted the specialist's recommendation. This trust of professional colleagues was made more explicit in Christopher's case, where Dr Novick suggested a new medicine to manage his drooling and Dr Brett demonstrated her trust in Dr Novick by saying:

> *"if Dr Novick thinks it's a reasonable thing, I'm happy to do it"* [C:Christopher, I2, Dr Brett, L:147].

However, in Maisie's case, trust in the specialist was not so forthcoming. This appeared to be related to fundamental differences in opinion regarding the role

of the specialists in her care. Dr Price did not trust the cardiologists' initial assessment of the most appropriate plan of care for Maisie, which was that she should receive solely palliative care and no further surgical treatment should be performed, regardless of the impact on her quality of life. In contrast Dr Price's view was that palliative care involved parallel planning, where curative treatment and symptom management should be delivered alongside each other. This opinion appeared to have led Dr Price to distrust the medical opinion of the cardiologists and subsequently to try to encourage Fiona to fight for Maisie to have further surgical treatment.

This section on trust has served to demonstrate the importance of experience in relation to developing and maintaining trust in relationships. Trust has been demonstrated as being essential to decision making and is often delegated within the parental dyad, implicitly or explicitly to the mother, although this was not always the case.

5.4.2. Poor relationships

Poor relationships were identified in six of the cases. When communication during the initial interaction between the professionals and families was perceived by the family as being substandard, poor relationships resulted. Poor relationships precipitated by poor communication in the initial interaction were reported in Evie, Maisie and Grace's cases. For Evie, during the appointment where her diagnosis was communicated, as outlined in section 5.3.2, her parents were required to contact the other hospital to obtain information to provide to the Neurologist. They perceived that this was inappropriate and inferred that it signalled a lack of preparation prior to Evie's appointment and a lack of compassion, when they were about to receive a devastating diagnosis. Despite identifying that they now have a *"reasonable working relationship"* [I1, Anthony, L:247] with the Neurologist, Anthony was keen to provide the details of the initial encounter around the diagnosis, demonstrating their residual shock about how it had been handled, suggesting that they have maintained the reasonable working relationship out of necessity, rather than desire.

For Fiona, when she met Dr Chase prior to Maisie's birth, she identified that it was Dr Chase's quietness and apparent uncertainty which made Fiona take an

instant dislike to her. Uncertainty was something which Dr Price also identified as contributing to the poor relationship that Fiona had with the cardiology team, stating that this, combined with a lack of medical consensus can lead to poor relationships and distrust. She said:

> *"I think all of these uncertainties and lack of singing with a common voice has led parents to distrust the team"* [C:Maisie, I2, Dr Price, L:34].

Although uncertainty appeared to be a key feature of many of the cases, the explicit demonstration of a high degree of uncertainty from professionals appeared to cause concern for some families and a breakdown in family-professional relationships. Both of the above examples of poor relationships resulted from the way that communication was handled at the initial interaction. However, for Grace, the poor relationship that Kay developed with Glenn, the social worker, was related to the content of the initial discussion, as well as the way in which the communication occurred. In her case, Glenn was required to have a difficult conversation with Kay at the very start of their relationship which resulted in a very poor ongoing relationship. Alice described that:

> *"When Glenn started there was a lot of issues. He didn't have that previous relationship that I had with Mum, so when we had to go out and pretty much get Mum to sign a contract to say that she wouldn't oral feed, it was a very very difficult visit. And I think that got them off on the wrong foot. I think that was one of the first experiences"* [C:Grace, I2, Alice, L:335].

Grace's School Nurse, Alice, highlighted that as one of the first encounters between Kay and Glenn, this tension had implications had for their ongoing relationship. This encounter saw Glenn challenge Kay's role as a good mother, by forcing her to comply with professional recommendations and thereby implicitly accusing her of acting in such a way that she was causing her child harm. Despite Alice's presence at this difficult visit, she had managed to maintain a good working relationship with Kay. This encounter occurred before the study had commenced and therefore it is not possible to identify how the good relationship between Kay and Alice had been maintained, but it is possible that

in this encounter Alice behaved as an advocate for Kay and Grace. Alternatively, Kay may have recognised the need to maintain good working relationships with the medical professionals and therefore either consciously or subconsciously sacrificed the relationship with Glenn, by attributing the blame onto him and relieving Alice and Dr Price of any association with this blame. The poor relationship between Kay and Glenn required ongoing mediation and advocacy during meetings, predominantly provided by Alice, which appeared to further cement the good relationship that they shared.

The other three cases within which poor relationships were identified, appeared to be caused primarily by previous negative experiences. In Michael's case, the poor relationship was between Kate and the Special Educational Needs Co-ordinator (SENCO) at his school. This relationship resulted from a perceived lack of listening or understanding from the SENCO and a perception that she was not enabling Michael to perform to his ability. Kate said:

> *"I mean did you notice a couple of times as well she said "you have met them before Kate", like I'm an idiot, that does me … the last report I read on him from her I actually did turn round and say "do you even like him?"… I find them a bit argumentative and it's like to be in a meeting and someone to give a suggestion about your child that you don't agree with and then be "no actually he really does", I'm like "OK" do you know what I mean it's a bit sharp…and a bit, "well I'm telling you that it isn't"* [C:Michael, I2, Kate, L:274].

Kate described how she felt that the SENCO would tell her something about Michael and would still push her opinion, even if Kate disagreed. This poor relationship led to difficulties in decision making, particularly in relation to Michael's transition to secondary school. The transition meetings at times felt tense and the poor relationship between Kate and the SENCO was palpable, despite many other people being present and expressing their views. This relationship, however, again may have been a sacrificial one, as it came to an end when Michael transitioned to secondary school and a new SENCO took over his care, and may have been aimed at trying to get what the school deemed was in

Michael's best interests in a situation where Kate disagreed with their assessment.

The poor relationships in both Christopher and Omar's cases existed between their parents and Mr Wallis the Orthopaedic surgeon and given that these poor relationships both existed with the same doctor, it may signal to a personality issue. Omar's family's experience was that Mr Wallis was not very personable and they felt that he did not care for Omar as a person, as Nadia explained:

> *"he was one of those doctors that when we went to see him he was always rush rush and he always wouldn't explain things to us properly and was like he really talked really quickly and we always felt that to us Omar was just another number to him. So we weren't happy with him, because we didn't think he wasn't actually telling us what you know really explaining things"* [C:Omar, I2, Nadia, L:402].

Nadia's quote above demonstrates her dissatisfaction at the communication with Mr Wallis and went on to state that that was why they requested a second opinion. In Christopher's case, the relationship between the family and Mr Wallis was not poor per-se, rather their confidence in his ability to perform Botox injections appropriately for Christopher was the single aspect of the relationship which had been damaged as a result of previous experience. Susan identified that she had no problems with other aspects of Mr Wallis' care, but that it was their experience of Christopher having had Botox injections from him which resulted in a reduction in their confidence in his abilities. She said:

> *"[Mr Wallis] was quite good with everything else. It was just the way he did that it was just so old school, it just, Christopher's face, I've never seen him cry so much and it was, he was pinned on the bed"* [C:Christopher, I2, Susan, L:466].

As Susan stated in the above extract, she felt that Mr Wallis was quite good with everything else and Christopher therefore continued seeing him for consultations, but Susan ensured that a different Consultant performed Christopher's ongoing Botox injections. Later on in the study, however, Christopher ended up having Botox from Mr Wallis and Susan was happy with the outcome, suggesting that although experience can precipitate damage to relationships, similarly good

experiences with the same professional can ensure that any previous damage is somewhat repaired.

5.4.3. Second opinions

Second opinions were requested in four of the cases. A second opinion was requested in Omar's case as a result of the poor relationship with Mr Wallis, as described above, and the family's desire to have a professional involved that they felt explained things in a way they understood, and with compassion. Evie and Elena's families also requested second opinions, but not as a result of poor relationships, rather out of a desire to obtain a specialist opinion. Both children had extremely rare conditions and their families were aware that the local professionals lacked expert knowledge about these conditions. Finally in Maisie's case, a second opinion was sought as a result of previous mixed communication and the desire to obtain an offer of surgery which may prolong Maisie's life.

Although some requests for second opinions were not viewed as problematic, in Elena's case, Pauline identified that she was concerned about stepping on Dr Price's toes, someone who she valued and had a good relationship with. In contrast, Evie's parents did not have this same concern, perhaps as a result of the poor relationship that they had with the Neurologist and a referral to Great Ormond Street Hospital was requested as they felt that they were not being given the full picture about her disease. Anthony, explained that they felt that the local Neurology team did not have enough experience to provide them with sufficient information. Anthony explained that in their case, the referral had had a positive impact on their decision making, as it provided them with an experienced professional who could provide in depth information about the condition and a more balanced picture:

> *"what we got from Great Ormond Street was somebody with 30 years of experience who knew exactly the condition had seen lots and lots of kids with the same condition and basically was able to have a qualified judgement on what the what the severity of her condition was and how it might progress and you know and all those sort of aspects. It had gone from a lifespan of somewhere in the region of*

just up to 10 years, to living into young adulthood, but with the potential because of the advances in medical technology for want of a better word…So you know it was a completely different way of looking at it and a completely different sort of outlook for us as a family" [C:Evie, I1, Anthony, L:308].

Anthony explained that his desire to know more detailed information about Evie's condition, and therefore requesting a second opinion, was partially about anticipating and preparing for decisions which they were likely to face in the future.

For Maisie the second opinion had a direct impact on decision making and was requested by way of obtaining options for her further treatment. There had previously been an offer of additional surgery, by Dr Alexis, but when he moved away, the local team felt that his offer would not be appropriate and was withdrawn. In order to reinstate the initial offer however, a second opinion was required and Fiona stated that obtaining this had been a fight:

"Fiona: I feel like it's been a fighting process to get her to where it's been, because I'm still fighting over a year on to get to where I want to be.

Emma: Which is the surgery?

Fiona: The extending of the stent. That's what I originally went for, so yeah I'm still fighting and it's took me a long time to get where I am" [C:Maisie, I3, Fiona, L:503].

Despite Fiona's feeling that it had been a fight to get the team to listen and to seek a second opinion, Dr Chase identified that she felt that obtaining a second opinion would be appropriate, as it was her feeling that Maisie should have further investigation with the view to possibly offering surgery. The poor communication described in section 5.3.1 resulted in Fiona feeling that she was fighting against Dr Chase, when it appeared that they actually had the same goal. In Maisie's case, obtaining a second opinion in the form of speaking with Dr Alexis and discussing with the clinical ethics committee, resulted in the offer of

further investigations and subsequently the offer and acceptance of the stent revision, as Fiona had initially wanted.

Despite the difficulties, both personal and structural, in requesting and obtaining second opinions, it appeared that they could be immensely useful in the provision of expertise, compatible personalities and the offer of further treatment options. However, at times they could pose problems, particularly in relation to the issues surrounding inter-professional communication (Section 5.3.2), when the expert advice that was given had not been adequately communicated to the other relevant professionals. In such cases it made it difficult for a consensus to be achieved and for other professionals in the child's care to provide advice regarding decision making.

5.5. Conclusion

This chapter has demonstrated the importance of communication and relationships in decision making for children with life-limiting conditions. Communication between the professionals and families was the basis on which relationships were developed and was evaluated predominantly as being good. Communication between professionals and organisations was identified as essential in the process of decision making, but was observed to be suboptimal in five cases and which resulted in significant challenges when making decisions. Although lack of communication between professionals may not have caused concern for all families, it may have the potential to have a significant impact on their ability to make decisions, as the full range of choices may not have been shared with them or they may have received contradictory information and advice. Good quality relationships were demonstrated to develop over time and were primarily based on positive previous experiences. Trust was an integral part of good relationships and was impacted on by communication, in relation to availability for advice provision and previous experience. Poor relationships often developed from poor initial interactions and poor communication. This chapter has highlighted the importance of communication in the establishment and maintenance of good relationships, which are essential for decision making.

Chapter 6. Findings Theme 2 - The process of decision making

6.1. Introduction

This chapter will provide a discussion of the characteristics of the decisions observed in this study and typify these. It will discuss the individuals' perceived moral accountability for decision making, primarily focussing on how parents perceive this as a core component of their parental responsibility. It will provide an understanding of how choice and options are communicated and the moral work involved in both the presentation of this information from a professional perspective, and the acceptance or rejection of options, from a family perspective. Finally the chapter will discuss the strategies used in the quest for agreement and the role of parental justification of decisions, with reference to the moral work being undertaken through these strategies.

6.2. Identified decisional types

The data collected from this study has illuminated the concept of decision making and illustrated that it is a dynamic process which often occurs over time, as outlined in the case timelines in Chapter 4. However, in addition to its dynamic and temporal nature, the concept was shown to take different forms, depending on the specific circumstances of the choice under consideration.

6.2.1. Immediately enacted decisions

These types of decisions were made and enacted immediately. This was for small every day choices where decisions were often not even recognised as decisions, as Susan articulated:

> *"I don't know, decision making, it all comes natural. You don't know you're doing it when you're doing it"* [C:Christopher, I3, Susan, L:10].

It appeared that the need for continual decision making had become so integrated into families' daily life, that they often did not recognise that they were making and implementing decisions.

Other immediately enacted decisions were more significant and involved a cognitive process. Daisy's case provided an example of this, where Catherine administered antibiotics immediately on recognition of Daisy having a cough. Catherine explained:

> *"I'll just start her on that bottle, it's always Augmentin that we usually have, just start her on that...if it's a wet cough and I can see she's got a runny nose, she's got a wet cough, it's on her chest, then I'll just start it and then ring them while she's on it*" [C:Daisy, Catherine, I2, L:98].

Similarly for Christopher, when the professionals recognised his poor feeding skills and signs of aspiration, they made the decision, which was immediately enacted, to discontinue oral feeding.

Many immediately enacted decisions were however, observed to be immediate refusals of a treatment that may or may not be required in the future. Often such decisions arose when professionals highlighted a concern about an aspect of the child's health, alongside a recommendation for how this could be improved, which was considered by the family as unnecessary at that time. Tube feeding represented the primary example of this where families often refused this at many subsequent appointments, each acting as separate immediately enacted decision point. In some cases the immediate refusal was accompanied by a statement that they would revisit their decision in the future, in light of the child's condition, as alluded to in the extract from Elena's field notes:

> *"Pauline stated that there had been issues with Elena's feeding at the start of the holiday…[but] that she is eating well now, although she is eating slowly, she is still eating. Dr Price stated that this was good, but that Elena will need a PEG at some point, to which Pauline said she doesn't think she is ready"* [C:Elena, O5, L:159]

In many cases the professionals enabled these treatment refusal decisions, which acted as a mechanism to delay the treatment, until the family recognised that it was necessary. This demonstrated the complex and often protracted process of decision making in which, for example, one decision may encompass many decision points where a treatment is rejected multiple times before it is accepted.

6.2.2. Future planning decisions

The other type of decision which was identified within this study was the future planning decision. Future planning, as a consideration of future possibilities, occurred frequently, both formally and informally.

Internally declared decisions

Informal future planning often occurred through a process of internally declared decisions, which were conceptualised simply as internal thoughts about what would happen in the future. If parents were alerted to the probable need for a future treatment they discussed having resigned themselves to the fact that this was necessary, suggesting that they had decided that they would accept this treatment. Such an internal decision was alluded to by Susan, in relation to her awareness that Christopher may need to commence NIV:

> *"I'm thinking might have to have one of these CPAP machines, so…but probably when he's got one I'll probably think "oh wish I had one ages ago""* [C:Christopher, Susan, I1, L:312].

Susan's statement that "when he's got one", suggests that she viewed this as a decision which had already been made and it was just a matter of time before it was implemented. For this type of decision, given that it has been internally made but not actually implemented, the final outcome may be different to the one that was initially internally declared, as was the case for Christopher's CPAP. In addition to Christopher's case, this type of decision was noted in two other cases. In Charlotte's case, the family had made the decision that she would undergo splenectomy and gastrostomy surgery, but this did not occur, due to changes in her condition. In Michaels' case, Kate had made an internal declaration of the criteria which would make her agree to a gastrostomy for him. Such examples signal to the internal thought process which occurs in relation to decision making and were articulated by individuals' as though they were decisions which they had already made.

Formal future planning

Formally decided and documented plans, such as emergency care plans, detailing the actions to be taken in emergency situations also signalled to decision making. These decisions, however, appeared to hold a greater

significance for professionals than families. Alice commented about how professionals value these plans:

> *"So the good thing about the [plan] is we have it in key locations, so Grace will have it with her, we'll for instance have one in our office. Transport will have one and [Children's Centre] will have one. It's easy to grab, you've got it with you and it's very quick sort of reference right you know "full compressions, bag and mask, hospital" you know and you can kind of break it all down"* [C:Grace, Alice, I2, L:187].

However, Pauline spoke about how she felt that the plan did not hold a great deal of relevance or significance for Elena currently:

> *"It's a bit like the suction machine, I've been carrying it around for 2 years and never needed it. And I know it sounds awful, every now and again I need to jog my memory as to if this happens, this is what I need to do"* [C:Elena, Pauline, I2, L:528]

The other interesting aspect about future planning decisions was the acknowledgement that although these decisions had been made, that they were liable to change, either through regular updating, or based upon relevant aspects of an emergency situation. Fiona expressed this to the doctors during a hospital admission, as outlined in the following field notes:

> *"Dr Simon said that there is an [Emergency Care Plan] in place and that he understands that the plan is to do CPR if Maisie collapses. He went on to ask if there is any other thing in place and Fiona said that it is what she decides and repeated this assertion, saying "what I decide is best at the time".* [C:Maisie, O7, L:187]

The changeable nature of the formal future plans may explain their perceived lesser significance for the families, who felt that they would be present with their child to make important decisions during an emergency situation.

Immediate decisions not immediately enacted

Finally another type of future planning decision was the immediate decision which was not immediately implemented. Such decisions were often discussed

in clinic appointments, for example, the possibility of further investigations or surgery and sometimes these decisions were immediately made during the appointment. Primarily these decisions were regarding elective surgery and families were sometimes asked to sign a consent form during the appointment. Given the elective nature of the procedures, they would not be conducted until a later date and therefore despite having signed a consent form, would be able to withdraw their consent and alter their decision before it was enacted. This type of decision was observed in Hassan, Omar and Maisie's cases in relation to surgical procedures or investigations under general anaesthetic. The timescales between the decision and the treatment/investigation ranged from a few months in Maisie's case, to 8 months in Omar's case and in Hassan's case the investigation had not been carried out by the end of the study, despite having been decided on almost 6 months earlier, thus demonstrating the temporal nature of decision making and future planning.

The study data highlighted that there are numerous types of decisions and at times these overlapped, or transitioned from one type to another along the process towards enactment of the decision. It was also observed that different stakeholders perceived the same decision as a different type, for example, in Michael's case, Kate had many times made immediate decisions to refuse a PEG. However, for Dr Price, this same decision had been internally and externally declared as a definite future treatment and therefore appeared to be conceptualised by her as a future plan. A consideration of these decision types provides an understanding of the complexity of the concept of decision making, prior to an examination of the features of the process in action.

6.3. Moral accountability in decision making

6.3.1. Parents as the ultimate decision makers

In all but one case, parents identified that they were the ultimate decision makers. Chapter 5 highlighted that primarily mothers were the main decision makers for the child's care within the family, and furthermore, as the parent they wished to portray that they had the overall decisional responsibility for the child's care and all aspects of their welfare. Salma identified that she was the ultimate decision maker and felt that as Hassan's parent, she was ultimately responsible for all his

medical decisions, and that even decisions to refuse major surgery would be within her realm of responsibility. She explained:

> *"what you always have to remember is yeah sometimes you don't have a choice but then you're still given that consent form aren't you, so ultimately it's up to you, because I could just say "no, he's not having it" and I'm within my rights to say that, even if it's a big bowel surgery, "just leave him, let him be, let's see what happens", but it is down to you…sometimes parents might feel "OK we've got no choice". In my eyes I'm a parent who really thinks about things, everything, oh just because a medical professional has given, it's not going to be OK, I'll sit and I'll take my time and say "no, I've made that decision". It's not me making myself feel special, it's me thinking hang on "how's this going to impact my son and my family*" [C:Hassan, I1, Salma, L:527]

In this extract Salma articulated her feeling that as a parent she has a right to decide what happens Hassan, but her statement also acknowledged the responsibilities attached to those rights, such as educating herself about the procedures and a wider consideration of the needs of her family. This quote also signalled to her beliefs about her moral accountability in decision making and her desire to be perceived as a morally responsible parent.

Parents described their responsibility in terms of their expertise and detailed knowledge regarding their child, which they believed meant that they were best placed to make such decisions. Fiona articulated how she felt that parental expertise trumps professional expertise and that this needs to be considered in decision making:

> *"I'm the parent, she might know textbooks and everything else, I know Maisie best, I've always said that from the beginning, she's mine, I know her. I'm the one that grew her, I'm the one that made her, I'm the one that looks after her, with great help and support, but she's still mine and I know her best"* [C:Maisie, I3, Fiona, L:188].

Other parents implicitly and explicitly referred to their own intricate knowledge and expertise regarding their children, as a way of legitimising their role as their

sole protector, and decision maker. Fiona's discussion implied that the practical knowledge, which only she has through her unique experience of carrying and caring for Maisie, is superior to the textbook knowledge, which anyone could acquire.

Professionals also expressed a belief in the importance of parental expertise in decision making, often stating that they would be instrumental in making decisions. Field notes for Hassan demonstrated how Dr Martin valued Salma's expertise:

> *"Dr Martin then said that they should start to think about weaning the ventilation at home…and went on to tell Salma that "you'll drive that" and that they would "hear from you how long he can manage""* [C: Hassan, O1, L:98].

Such commitment to parental expertise was frequently observed; in the majority of instances when a treatment was recommended, professionals accepted the parental decision, except in cases where they believed that this put the child at risk (see Section 7.3).

Michael's was the only case in which a parent did not identify as the ultimate decision maker, rather his mother, Kate, stated that decision making was shared with the professionals:

> *"I think it's shared, I think it's a joint thing, you've got to work as a team with the specialists, you've got to understand where they're coming from though. As a parent, for me, I've got to understand where they're coming from for why they want to make this decision. I think, see this is the thing where if a specialist made a decision on something and I didn't agree with it, I want to be able to say "I don't agree with it because""* [C:Michael, I2, Kate, L:322].

Despite Kate's assertion that decision making was shared, she wanted to be able to disagree. In both of the significant decisions in Michael's case, Kate had been observed to be the one to make the final decision. Both times her decision was in contrast to the one that the majority of professionals favoured. Additionally through reference to the medical notes, it appeared that there had been many

other decisions where Kate had disagreed with the medical recommendation, such as whether or not to start prophylactic anti-biotics, suggesting that despite her assertion that decision making was shared, she mainly acted as the ultimate decision maker. Kate's self-portrayal as a team player and a parent who sought to understand why recommendations were made, appeared to enable her to reasonably justify her disagreements with the professionals' recommendations. Consequently she could maintain the appearance of a morally accountable and grateful parent. Her apparent role as the ultimate decision maker, as contrasting with her portrayal as a shared decision maker, may have resulted from her need to retain control over her and her son's life, something which she identified was continually being removed from them by the disease process.

Conversely, in some of the other cases where parents identified themselves to be the ultimate decision maker, for some more significant decisions, they were at times observed to be making shared decisions, or agreeing with the professional recommendation. Even in such instances parents described themselves as ultimately responsible for having made the final decision.

Parents' portrayal of themselves as the ultimate decision makers was often utilised as a way of ascribing the child's positive outcomes to their decisions as a parent. This alludes to the moral work involved in making and living with their decisions and reveals the importance of a feeling of agency for parents, particularly when an improvement in the child's health is demonstrated. Hassan's case exemplified how parents' may perceive that they are morally accountable for decision making and how their need to act as a 'good parent' results in them seeking treatments to improve their child's quality of life. Her account of starting Hassan on CPAP, fully attributes that decision, and his subsequent development, to her having requested this treatment. Salma explained:

> *"I said "what are those machines called? Can we have one of them", you know not knowing anything and being quite naïve and they were like "well that's, he's not on ventilation, he's on CPAP" and I was like "well can he have that?" Then it was an elective procedure, he went, I really pushed for it but it's been brilliant, he has thrived. Ever*

> *since he's been on that CPAP, going from a baby who just lay there, he's so interactive, he's so inquisitive, he's thrived"* [C:Hassan, Salma, I1, L:443].

Despite Salma's need to claim responsibility for the decisions which had had a positive impact on Hassan's care and development, she also asserted her responsibility for decisions which did not have such favourable outcomes. An example of this was the decision to try him on a new feed, which resulted in a number of days of diarrhoea and discomfort for Hassan. This aspect of claiming sole responsibility for decisions with unfavourable outcomes was rare and primarily families tried to distance themselves from responsibility of decisions with unfavourable outcomes.

Such behaviour was demonstrated in Charlotte's case regarding the decision to try the NGT. Charlotte had been resistant to it and her parents thought that there were likely to be negative consequences to NGT feeding. Given these concerns, the family portrayed the decision to have the NGT as a reluctant agreement, rather than an active decision as autonomous agents. The negative consequences of NGT feeding for Charlotte may have resulted in the portrayal of the decision as led by professionals and reluctantly agreed to by the family. Such an account enabled the family to detach any blame for the negative outcomes from them and thus potentially relieve any feelings of associated guilt. Jamie's account regarding the NGT explicitly highlighted his feeling that the decision had not been the family's:

> *"We conceded"* [C:Charlotte, I1, Jamie, L:183].

Guilt was an emotion which was implicit in many of the parents' accounts, particularly when discussing terminating oral feeding or commencing tube feeding, as parents referred to their child's love of food and the desire to not deprive them of this. The explicit reference to guilt in decision making however, was only made by one parent. Salma stated:

> *"you do have that, I think with a lot of decision making in this particular circumstance there comes a lot of guilt, as to whether you've done the right thing or not"* [C:Hassan, I2, Salma, L:108].

In this extract Salma refers to concern as to whether she has done the right thing, suggesting that at times she may experience decisional conflict which may result in a sense of guilt. Additionally competing goals of care may mean that feelings of guilt are experienced by parents even when they consider that they have made the right decision. For example, with Hassan, he may require numerous normalisation surgeries to correct his facial disfigurement. Salma will therefore be required to choose between the desire for him to appear normal and the desire to avoid surgery, which are competing goals and therefore likely to precipitate guilt.

Joanna, Elena's physiotherapist, articulated how such conflicting goals are often present in any elective surgical decisions, thereby making them difficult and partially characterised by guilt. She explained:

> *"If you electively do surgery on your child, it's very different from if you get a phone call from A&E that says "child's arrived at school, broken leg, we need to do surgery", you actually as a parent have no option. Whatever we choose to do with our children, other than those circumstances are elective…But those conversations will have to be revisited and they're the hard ones because if what happens is that she mentally deteriorates…if you lose the essence of who that person is and you have electively done that surgery, I think that's a really hard decision"* [C:Elena, I1, Joanna, L:519].

The moral work of parents to avoid feelings of guilt may signal to why they may at times be reluctant to claim sole responsibility for some decisions, even when they have provided an account suggesting that they are the ultimate decision maker for their child. Despite the longitudinal nature of the research, there was no data to suggest whether claims of decisional responsibility were only made once the outcome of the decision was clear or whether their claims of decisional responsibility underwent narrative reconstruction and changed over time, in relation to the changing outcomes of each decision.

This section highlights the need for parents to feel accountable for the decisions made for their child, albeit with the need to distance themselves from this at times when their child suffered negatively as a result of decisions. Doing so

appeared to be a way in which parents could spare themselves some of the guilt associated with such outcomes. The following sub-sections aim to provide an overview of the types of decisions which parents took or delegated responsibility for.

Examples of parental responsibility for decision making

Parents were happy to make decisions which impacted on the child's schooling or housing modifications without seeking advice from professionals. In some situations, advice or recommendations were given, but families felt confident to discount this due to their knowledge and expertise regarding what would be appropriate for their child and family.

Aspects of care which had become integrated within the sphere of everyday life for the family, came under the responsibility of the parents, even if they would usually (for children without complex needs) be considered within the realm of medical professional expertise. For example, in Maisie's case, she was continuously on oxygen in the daytime and as a result of Fiona's extended experience in managing this, the decision whether to increase or decrease the amount of oxygen delivered was made solely at her discretion. This was observed to be the case even when Maisie was admitted to hospital, where the professionals deferred to her expertise regarding how much oxygen was appropriate for Maisie at that time. Decisions regarding some medications also provided examples where parents accepted primary responsibility.

Examples of parental delegation of responsibility

Despite parents' need to feel ultimately responsible for their child's care, at times they explicitly delegated decisions to professionals. This occurred particularly for decisions which held more gravity in relation to their child's physical health, required a degree of technical understanding which parents felt they did not possess, or for decisions where there was some degree of uncertainty.

In Hassan's case, Salma was adamant that she was the ultimate decision maker. However despite these assertions, she was at times observed to defer to professionals for certain decisions, not only asking for advice and accepting recommendations, but requesting professionals to explicitly make decisions for her. During one clinic appointment Salma articulated that she felt that Hassan's

gastrostomy button needed changing, but requested Mr Abraham to make the decision and tell her to change it. It appeared that contextual factors, such as the stress of caring for Hassan at home, alongside his behaviourally challenging sister meant that Salma felt that she perhaps no longer trusted her judgement to make certain decisions. In this situation it appeared that being given instruction by the surgeon, particularly given that this procedure was new to her, provided her with the moral justification for her actions, in case anything went wrong. Such delegation did not appear to detract from her feeling of ultimate responsibility however, but was an integral part of making morally responsible decisions.

In Charlotte's case, certain technical decisions were observed to be delegated to professionals, whilst other medical decisions remained within the family's realm of responsibility. The decision for Charlotte to have a stenting procedure to reduce her portal hypertension was an explicit example of a decision made by the professionals and agreed to by her family. Jamie stated:

> *"The other decision was this functionality of the stent issue that we had the other month which was again a medical decision you know. The ultrasound indicated that the velocities were high and that it was starting to narrow…they decided to intervene, but no decision on our side really, I mean just informed rather than being part of the decision making"* [C:Charlotte, I2, Jamie, L:114].

In Jamie's narrative around this decision he acknowledged that the family could have had greater involvement into the decision, but felt that they did not have the necessary knowledge to meaningfully participate and were therefore happy to delegate the responsibility for this decision to the medical professionals. In Jamie's account he portrayed himself as a morally responsible parent through the need to at times push the boundaries and challenge medical decisions, but also an awareness of the limits to this and when deference to the medical plan was necessary.

The data indicated that many parents felt that the ultimate responsibility for decision making was theirs. However, this often occurred alongside instances where they explicitly deferred to professional recommendations. Parents did not

appear to find this contradictory, rather they continued to declare that they had ultimate decisional responsibility. It appeared that their decision to defer to professionals was integrated into their feelings of ultimate decisional responsibility, such that even in cases where the professionals made the decision, enacting this had been facilitated through the parents' deference. Regardless of whether parents deferred to professional advice in decision making, unless the decision was associated with unfavourable outcomes, on the whole parents were keen to assert some degree of responsibility for the decision. This appeared to be related to their need to portray agency and demonstrate that they have the final say, thus being able to validate their role as a morally responsible parent.

For professionals, there was an implicit commitment to sharing decisions, however there was very little explicit discussion of decisional responsibility, except in cases where there was a concern about the child's best interests (see section 7.3).

6.3.2. Shared Decision Making

Alongside parents' portrayal as the ultimate decision makers, they often discussed the benefits of sharing decisions with professionals. Some of them felt that this was facilitated through requesting advice from a trusted professional, particularly for decisions which were out of the parent's sphere of experience or knowledge and therefore they required more guidance regarding the possible impact of the decision. Seeking such advice was one way in which parents continued to position themselves as morally responsible. Such advice seeking was exemplified in Maisie's case, when Fiona sought advice from Dr Price, a trusted professional, regarding her view of Maisie's potential surgery.

A commitment to SDM was often inferred by professionals in the study. Dr Brett provided an insight into how it may be easier for SDM to occur when a professional peripheral to the decision shares the information, as they do not have a vested interest in the decision. She explained:

> *"What I try and do is to try and navigate between the difference between can and should…And I think that's my role in helping parents to understand that and then getting their views, because I probably I haven't got a best interest in either operating or not operating, I can*

look at it in a slightly different way" [C:Christopher, Dr Brett, I1, L:245].

In such situations where the professional sharing the information is not immediately connected with the decision, as Dr Brett alludes to, they can present the information in a neutral way and elicit the family preference, as a way of ensuring SDM.

SDM was observed during appointments and was exemplified in Ella's appointment with Dr Price, regarding the treatment of her chronic constipation. The field notes detail how Dr Price gave an overview of the medications that could be used and together she and Jill arrived at a compromise:

"Dr Price stated that what Ella needs is something to give the bowels a push and she gave some examples: Senna, prunes, Picosulphate or Movicol. Dr Price highlighted that she would want Ella's bowels opened once a day ideally…[and] to give the push, she stated "I'd prefer to try the Movicol again" and to "try a tiny dose but frequently". Jill was quite forceful here and said "we've discounted Movicol" and that she thought that the Picosulphate and Senna "causes irritation with the stomach" when they had tried it previously…Dr Price said to give Picosulphate another try and Jill responded by saying that they "will be happy to try that but have discounted Movicol"." [C:Ella, O2, L:76]

This extract shows how, for some decisions, the range of options and professional preference can be shared and this can then be combined with the parents' experience of the child to arrive at a decision. Despite Jill's reluctance to try the Picosulphate again due to concern about irritation of the stomach, she was happy to agree to this, but fully rejected the recommendation of Movicol, suggesting that both opinions and preferences had been integrated together to arrive at a decision.

Despite a commitment to SDM either explicitly stated or implicit in professionals' accounts, it was not always observed to occur in congruence with the policy definitions and appeared that for professionals, parental acquiescence or lack of dissent with their stated plan, signalled that SDM had occurred. This

was a regular feature in appointments where professionals would state what they felt the plan should be and often parents would agree, or would fail to disagree and the plan would be implemented. One extreme example of this was noted in Ella's spinal appointment where despite mentioning that her parents would be involved in the decision of when to operate, Mr Machin provided information and his opinion to Jill, but did not invite her to provide her opinion. However, reference to the field notes suggest that he believed that a shared decision had been made:

> *"Mr Machin narrated a clinic letter into his Dictaphone, whilst looking at Jill intermittently. He narrated Ella's clinical history and then said that "we've decided to leave matters alone for the moment" regarding the scoliosis surgery"* [C:Ella, O1, L:59].

Mr Machin stated that "we've decided", signalling that he believed that they had jointly made the decision, integrating Jill's non-dissent of his preference into the decision, rather than inviting her to meaningfully participate. Jill did not appear to feel that surgery was appropriate at that time and therefore may also have perceived that SDM had occurred. Similarly clinic appointments for both Omar and Hassan where surgery was discussed also demonstrated this type of decision making, where a professional recommendation was given and a parental agreement assumed, rather than discussion of the pros and cons to each of the viable options. This suggests that the professional conception of SDM may be in contrast with that suggested in national policy.

Professionals were keen to clarify however, that SDM does not mean unrestricted choice and that they are acutely aware of their role in facilitating reasonable choice. Dr Price indicated to the moral work that is undertaken in such instances of SDM, saying:

> *"There can be several reasonable pathways that I can live with, but there will be some pathways that I can't, that would be too bad for the child…so in my mind I've got a best pathway that I would say is the number one choice and I've got a worst case scenario which I'm not going to go down at all…And then there's generally a range of different things, options in between and I try to negotiate as far as I can toward*

what I think is the best pathway, but there may be ones that I can live with in the middle, that I can say aren't best care, but they're good enough and they're not harmful" [C:Maisie, Dr Price, I2, L:247]

This demonstrated how choice may be constrained through professionals' perception of what is appropriate for the child (see section 6.4). However, at the other end of the spectrum full choice was provided to families and such situations occurred where there was significant uncertainty. These situations were in relation to Maisie's cardiac surgery and Ella's gastrointestinal surgery. In both cases the families were given the information by the specialists and told that the difficult decision was theirs to make. This consideration of SDM demonstrates that in practice it exists on a continuum from family led decisions, to professional led decisions, with non-dissent from the family.

6.3.3 The child's role in decision making

Within this study only two cases involved children who were of the age and cognitive ability to participate in decision making[23]. For 11 year old Michael, Kate spoke of him being involved, particularly in relation to the refusal of the gastrostomy and appeared to feel that this was something that he had the capacity to decide on. Dr Price disagreed and tried to explain to Kate, that given Michael's age, it would be hard for him to understand the long term implications of any decision he made. It seemed that given Kate's awareness of how ongoing discussions about Michael's weight and the potential for a gastrostomy made him upset, she drew upon his refusal to have the gastrostomy as a way to avoid further discussions and cause any further upset, as well as somewhat absolving herself of the sole decisional responsibility.

As a mature 14 year old, Charlotte was competent to participate in decisions regarding her care, and this was something that her parents promoted. Professionals also involved Charlotte in decision making and often made decisions about the timing of her treatments based on her assessment of her own health, suggesting that they had confidence in her decision making abilities. Charlotte, although she was under the age of 16 years old and therefore not

[23] Elena was able to communicate verbally, albeit very slowly, but her parents felt that she would not understand enough to participate in interviews and did not consent for her participation in interviews.

legally responsible for making decisions, felt that she was afforded the ultimate decision making responsibility already, as outlined in the extract below:

> *"Emma: when you go to the [Adult Hospital] what are you thinking in terms of, so you'll be 16, you'll be like legally the person that makes decisions about yourself. Do you feel that the process of you making decisions will be the same in terms of checking, involving your family and involving the doctors and things as well or do you think it will be different?*
>
> *Charlotte: I'll consider what the doctors say and what everyone else says and then I'll make my own conclusion.*
>
> *Emma: Is that how you feel you do it at the minute? Or do you feel like other people have more input at the minute?*
>
> *Charlotte: I think so. No I think that's how I do it now."* [C:Charlotte, I3, Charlotte and Rachel, L:406].

This extract demonstrates that although Charlotte's parents acknowledged that Charlotte was heavily involved her decisions regarding her care, Charlotte appeared to feel that she was the one that held the decisional responsibility. Given her age and level of intelligence, she may have been aware that she could refuse treatments if she was able to demonstrate that she had the capacity to make the decision. Charlotte's case in particular suggested a close family relationship, wherein decisions were discussed openly. This possibly resulted from the fact that her parents were separated and her father was heavily involved in negotiating her medical care, such that open discussions were needed and therefore the family appeared to arrive at a preference for major decisions together. Jamie discussed how in some situations he would be inclined to support Charlotte's view, even if he held an opposing one, citing the importance of her psychosocial wellbeing in view of her limited life-expectancy.

6.4. Communication of choice and options

6.4.1. Communication when there is uncertainty

A significant factor in the process of decision making was how the options were communicated. In many cases treatment options were initially presented as

possibilities for the future, providing families' choice in whether or not to accept the offer of treatment, or regarding the timing of acceptance of the treatment. These discussions generally signalled to the professional's preferred option and were presented to the families in such a way which demonstrated this preference as being in the child's best interests. However, professionals also appeared to be undertaking moral work in situations of uncertainty, where the professional perceived that there was no clear 'right' option. In such cases different options were presented for the family to choose between and professionals appeared to see it as their duty to provide the family with a choice which would best fit in with their family life and values. This was explicated by Dr Brett regarding the possibility of Christopher commencing NIV:

> *"I think if he would benefit from that, it's not, it's harder to see clear, clarify what the benefits are and I think Dr Martin is wanting us to do, you know he's not going to rush into it but he may go down that route. But it's very much about what the parents want, we don't have to do it"* [C:Christopher, I1, Dr Brett, L:304].

Similarly, discussion with Dr Price demonstrated how circumstances exist where, despite the professional recognition of an optimal course of action, there may be numerous other acceptable options which, although not optimal, do not pose a risk to the child. In such situations the family would be informed of the acceptable options and enabled to choose between these. The decision by professionals to allow families to make such decisions also comprised some moral work, as professionals were required to undertake continual judgements about whether or not the child's health or quality of life was being put at risk. Observations demonstrated that in cases where there was no obvious best treatment option, professionals believed that it was their obligation to provide the family with the choice regarding what course of action they would prefer. This seemed to signal to a need to portray a culture of SDM, something identified by policy as best practice.

However enabling choice and SDM sometimes presented professionals with two concerns: decisional burden and best interests. Despite the need for professionals to feel that they were providing choice to families and empowering them to make

decisions impacting on their lives, it was highlighted that this may at times present a huge burden for the family, such that professionals felt conflicted about the extent to which they should help share this burden with the family. Dr Chase stated:

> *"I think there is a point there that I think we should be trying to take away the burden or that the parents are making the decision so that they don't feel that they've made the right or the wrong decision for their child. This is, it's difficult because you don't want to be too paternalistic and say "that as a medical team, this is what your child should have, do you agree", because it's just, that kind of whole counselling thing I really struggle with because, I think, do, you can see what I'm getting at is…that they push, it's just so difficult that how much do they engage I suppose and how much do they not engage and how much do they feel that that decision making is on them"* [C:Maisie, I2, Dr Chase, L:420].

The way Dr Chase articulated the provision of choice and whether this is solely placed on the family or shared in relation to decisional burden is something that she reported having struggled with. It demonstrates the moral work that professionals undertake when assessing the extent to which they should place decisional responsibility on the family.

6.4.2. Communication when there is only one viable option

However, for many decisions it was outlined explicitly that professionals perceived there to be only one right treatment option and therefore this was presented as the only choice, thus rendering the ideal of shared decision making unrealistic. The moral work undertaken by professionals in the presentation of choice in such situations was expressly demonstrated by Dr Price in the below extract, where she identified a moral imperative on professionals to be paternalistic and only offer options which they believe are in the child's best interests:

> *"Parents will always say "try". If you say "well I could", they'll say "oh try"…so you mustn't say "well I could but you can't have it".*

> *You have to say "this is the best thing""* [C:Maisie, I1, Dr Price, L:453].

In this extract Dr Price also alludes to the moral work undertaken by parents, with the understanding that often they feel the need to try everything to keep their child alive, even if it may result in some suffering. She therefore concluded that the professional has a moral responsibility to provide direction to families and try to ensure that they agree to what the professional perceives to be in the child's best interests.

In many of the cases where there was only one option presented, families understood the need for the treatment and that there was no other acceptable option. This understanding facilitated the alignment of the parental and professional view of the morally responsible choice, as explained by Catherine:

> *"it's obviously that's what she needed so it was just, we just took obviously all doctors' advice and did what [was needed]"* [C:Daisy, Catherine, I1, L:47].

In the above extract Catherine described doing what was needed, suggesting she felt a sense of ultimate decisional responsibility, even in this situation where there was no other choice. This further reveals the need for parents to feel a sense of agency in relation to their child's care, even when they have little control over the options.

6.4.3. Communication when the only presented option is rejected

In some situations families were presented with little or no choice, but nevertheless sometimes demonstrated a feeling of moral responsibility to reject the 'only option'. This was demonstrated in a number of cases regarding parental rejection of gastrostomy insertion for their child. In these cases, the gastrostomy was presented by professionals as the only feasible option, but the families were aware that they also had the option to do nothing and maintain the status quo, at least for the time being.

However, in relation to housing modification and equipment provision, parents demonstrated defiant rejection of the only presented option, instead seeking out alternatives on their own. In Evie's case, her parents worked incredibly hard to

identify that the only choice that they were presented with in relation to her wheelchair provision, was not actually the only choice. They were told that Evie was only eligible for one wheelchair, which upon assessment they recognised would not be suitable to meet her needs and would therefore alienate her from her peers. Her parents discussed how they were prepared to instigate a challenge in order to advocate for Evie. Their self-identified educated background meant that they questioned this provision and exerted pressure on the service to ensure that Evie got what they felt she needed. Anthony said:

> *"we asked them for a copy of their policy, which they went "oh, what you want that for?" "well we want a copy of your policy, hand it over", so they did and then we went "OK well that's a bit of a poor policy" so we went to the Clinical Commissioning Group (CCG) and we said "this is their policy" and they said "hmm OK" and I said "can I see your copy of your contract that you have with wheelchair services or the NHS" and they went "course you can" and they took it out for us and they went "oh it doesn't match"...So we went back to wheelchair services and they said "oh yes you are right, you can have a wheelchair with a riser on it""* [C:Evie, I1, Anthony and Ruth, L:418].

Their actions highlighted that what they had been offered was actually not the only choice, but the one that was in the organisation's interest to provide. Anthony described the energy they invested, to ensure that they were aware of the full range of options. They were also aware that the CCG had not had this experience of families coming to them with such questions before, demonstrating that most families were unlikely to challenge the assumption that the only choice presented to them was in fact the only choice available.

This demonstrates that the way choice and options are communicated impacts on the decisions that families make and this case demonstrates that families questioning the choices communicated to them is rare, particularly when service provision issues are involved. It demonstrates the power held by professionals and organisations in the provision of choice, particularly for faceless structures such as Wheelchair Services and the CCG, which may seem out of reach for

families to communicate with. Anthony's assertion that this was the first time a family had gone to the CCG with such a concern, suggests that the enactment of this power presents a significant barrier to informed decision making for families.

Evie's parents underlying values of ensuring integration for her also drove them to recognise the lack of appropriate choice in relation to housing modifications and similarly reject these, despite their presentation as the only option. Ensuring that she was fully integrated in family and social life, and had access to all the normal opportunities of a child her age guided their decision making and appeared to be how they could feel, and demonstrate that they were good parents. Ruth stated:

> *"There's no way in this world that you know I'm, we're ever going to exclude her from anything. If your home can't work for you, God only, God help you because you know out there the world, goodness knows the world is not set up for people who have to use wheelchairs"* [C:Evie, I1, Ruth and Anthony, L:750].

6.4.4. Proactive searching for options

In Omar's case, Nadia appeared very aware that professionals do not always have the time to search for or think about other treatments or options for Omar, and when he is stable, they may not suggest any further investigations or treatments. This perception was verified through reference to his medical notes after Nadia had asked about further investigation regarding airway management:

> *"Ask ENT to r/v re: airway as parents concerned re: NPA and whether any further airway input needed, although we have explained that if managing well with NPA, unlikely to undertake any surgical management."* [C:Omar, Medical Notes, Entry dated 04/08/15].

Nadia articulated that her awareness that professionals lack the time to focus on Omar and what would be best for him and therefore felt the need to be proactive, searching for new treatments and researching options for Omar which she then discussed with the professionals in order to gain their opinion. This strategy appeared to have been instrumental in Omar having a tonsillectomy. This

enabled Nadia to attribute the positive outcomes of such treatments to herself and consolidate her feeling as a good parent.

6.4.5. Acceptance of recommendations

Despite the proactive seeking of treatment options, if professionals presented a treatment option for Omar, Nadia almost always accepted this without question, due to her underlying belief that the professionals know best and that as a good parent, she ought to agree with their recommendations. In Hassan's case, despite not always agreeing with professionals, Salma identified that when surgery is concerned, she generally accepted the professionals' recommendations because she believed that they would not offer futile surgery, possibly signalling to the moral work that the organisation undertakes in the allocation of resources. She explained:

> *"There's been times where you know the surgeons have just come round, given me the forms to sign and I'm just like "does he really need this?" You know clearly he does because the NHS have got better things to do than carry out futile surgeries"* [C:Hassan, I2, Salma, L:100].

The way in which choices were presented influenced the extent to which they were accepted or rejected. However, families who appeared to have greater health literacy were more able and willing to research these choices and reject them if necessary. Professionals' moral work was influential in how they communicated choice and positioned themselves as morally responsible agents.

6.5. Achieving agreement

During decision making, often disagreements between stakeholders were encountered regarding the best course of action. In such situations there were a number of strategies which were used in order to try to solve the disagreement, or justify the individuals' preferred option. Negotiating disagreements appeared to involve a great degree of moral work for all stakeholders, during which they challenged others' perspectives of the right choice and in turn their own perspectives were challenged and required justification. Both families and professionals undertook similar amounts of moral work during disagreements as they both simultaneously worked towards achieving agreement through

persuading the other of their perspective. For many of the cases, disagreement was related to the initiation of tube feeding, but was also identified for other decisions, as outlined in Table 5. Daisy's case was the only case in which disagreement did not feature and may have resulted from the early stage of her illness in which her parents were still developing confidence in their expertise and in which contentious decisions had not yet been encountered.

Case	Tube feeding	Other topics of disagreement
Daisy	N/A	[None identified]
Maisie	x	Cardiac surgery
Michael	✓	Schooling, Prophylactic antibiotic use
Christopher	✓	[None identified]
Grace	✓	[None identified]
Charlotte	✓	Portacath, Schooling
Elena	✓	[None identified]
Omar	x	Resuscitation plan and going home from the NICU
Hassan	✓	Eye surgery
Evie	N/A	Wheelchair provision and need for specialist referral
Ella	x	Medication

Table 5: Topics of disagreement

[Key: x-no disagreement identified, ✓-disagreement identified, N/A-no discussion of tube feeding]

6.5.1. Persuasion

Tube feeding

For the nine cases in which discussion of tube feeding was identified, six of them had experienced historical or ongoing disagreement about this decision. Discussions regarding tube feeding often occurred over a number of years, during which time the professionals, who believed that such methods of feeding would be in the child's best interests, tried to persuade the family of their view. Families discussed occasions where they felt under pressure by the professionals to agree to their child commencing tube feeding and similarly the professionals identified having a role in trying to overcome family reluctance and in ensuring the family recognised the need for this. The mechanism of persuasion incorporated a range of strategies from simple repetition of information over an extended time period, to discussion of the limits to parental decisional responsibility in cases where professionals felt that the child's health was being put at risk.

Dr Price identified that she believed that she had a duty to try and persuade families of what she saw as the option which was in the child's best interests and in the example below she discussed how she felt the need to ensure that Kay agreed to a gastrostomy for Grace. Dr Price said:

> *"I have a duty to do that and of course I want to. But on the other hand I've got a mummy who isn't seeing what I'm seeing and has a different view. So what I need to help her to see what I'm seeing and help her to understand why I'm saying what I'm saying. A) I need to help her to hear what I'm saying and I need to help her to believe what I'm saying and I need her to then agree with me or come on the same, come on my path"* [C:Grace, I2, Dr Price, L:226].

The use of language in this extract conveys the sense of moral duty that Dr Price felt in relation to persuading Kay to agree to the gastrostomy for Grace and how this had been incorporated into her own sense of morality, such that she felt a personal motivation to act in accordance with her professional duty. This sense of duty in relation to persuading families of a decision, which the professional believed to be best for the child, was observed in numerous clinic appointments, albeit in more obvious or subtle forms.

Although persuading occurred in relation to the gastrostomy for Elena, it was somewhat more subtle than in the other cases presented. Given Elena's adequate weight and her lack of any further choking episodes, the family were supported in their decision to delay any action towards initiation of tube feeding. During one appointment, Elena's parents stated that they did not believe that the upcoming surgical appointment was necessary yet and Dr Price agreed to the family's decision to postpone this, but was quick to highlight that the gastrostomy was a certainty in the future for Elena, as shown in the field notes extract below:

> *"She highlighted that they can use the tube in the nose if needed, but that it has its disadvantages and went on to say "I'll let him know that her feeding skills have improved", but that the "PEG will be required eventually""* [C:Elena, O5, L:171].

There were numerous times throughout the study when Dr Price similarly highlighted that the gastrostomy was a certainty for Elena, rather than a possibility and this appeared to be more a way of acclimatising the family to the need for a gastrostomy eventually, through repeated exposure to the idea, rather than persuading them that it was necessary. Many times throughout the study, Pauline stated that she knew that a gastrostomy would be inevitable for Elena, but her concern was to delay this for as long as possible. Given that the gastrostomy was not being presented as something which they ought to agree to imminently, the family appeared to feel a moral obligation to delay until it was absolutely necessary, but in doing so, regularly calculate the risks of delaying a procedure that they knew she would eventually need.

Perhaps Dr Price's lack of direct persuading in Elena's case was related to her knowledge that the family accepted it as an eventuality, but were trying to keep some semblance of normality for as long as possible and would agree to it at the time when they identified it as necessary. It appeared that she was aware that putting any additional pressure on the family to agree to the gastrostomy earlier, may result in unnecessary stress to the family and damage to their relationship, which would not be in Elena's best interest in the long term.

Persuasion did not appear to frequently result in agreement, although trusted professionals entering the discussion and seeking to persuade families caused parents to reconsider their position. In Michael's case, Dr Patel had been discussing a gastrostomy since Michael was five years old and had sought to persuade the family over the following six years to agree to the gastrostomy, using the evidence of his static weight as a mechanism to try and persuade them. However, Kate identified that the involvement of Dr Price in the discussion had a greater persuasive effect on her, clarifying that her involvement in the discussion led her to question her decision more:

> *"Kate: You argue in your head about the pros and cons all the time. Now that Dr Price's on the case as well, and it also depends how they approach it, you've got to think that have they got the understanding of what we've got and if they have then you're probably more likely*

be on their side as well, "yeah I see where you're coming from". Which I do see where they're coming from.

Emma: So do you mean now that Dr Price is on the case you're questioning it a bit more?

Kate: Oh definitely." [C:Michael, I2, Kate, L:393].

The addition of Dr Price into the discussion appeared to have a persuasive effect, due to the trust that Kate had in her, which suggested the need for Kate to revisit her previous decision.

Persuasion also occurred within the family, as demonstrated in Michael's case. His age and cognitive ability meant that he was able to participate in decision making but was extremely resistant to the gastrostomy, and despite being able to articulate that he felt that he would be healthier if he had one, did not want it because it meant having a hole into his stomach. In his case, the process of persuasion occurred during appointments with Dr Price and also in discussion with his mother, where she appeared to feel the need to 'sell' it to him in different ways. Kate explained:

"He's just so against it. I've done the nice way, I've done the positive way talking about it to him. You know I've said that if he ever gets poorly, meds can go in there. I've done the nasty way "you're gonna have to have it, it's getting ridiculous now". You know you've tried that way, so now it's got to the point of, right we've tried everything, I've said everything to him, it's him now what needs to get his head round it… I can't do it to him anymore, I've had enough, he's had enough, it's the topic of his life" [C:Michael, I3, Kate, L:346].

This extract demonstrates the need for parents to assist with the strategy of persuasion, but also highlights the stress and concern that acting in this capacity could cause. Kate's participation in persuasion had caused Michael distress and therefore she no longer wanted to participate in that process. The moral work of being a good parent and trying to persuade Michael to agree to the gastrostomy had begun to make Kate feel like a bad parent and that consequently she was unprepared to continue to participate in persuasion. She subsequently wished to

take up the role of, or at the least portray herself as, a parent who would vehemently support her child's choices.

In addition to the persuasion outlined above, it was simultaneously employed from the parents to professionals and aimed to persuade the professionals that their child did not need tube feeding, or at least not yet. Parents provided evidence to back up their claims, and this strategy of appealing to evidence will be discussed separately (see Section 6.5.3).

Other decisions

Although persuasion was most often identified in relation to tube feeding decisions, it also occurred in relation to other decisions and there was evidence to suggest that it was utilised not only in the professional to family direction, but within the family, within the medical team, and also from families to professionals. Such instances occurred in five cases.

In Omar's case, when he was an infant, owing to the complexity of his health conditions, Nadia was reluctant to take him home from the neonatal unit. She described how when he was first born, the doctors would try to persuade her to take him home to die and this may have resulted in her continued reluctance to take him home when he was ready, as she had previously connected the idea of going home with Omar's death. Omar's cousin Hafisa, who had just started training to become a midwife, felt that she had played a key role in the process of overcoming these issues and persuading Nadia to take Omar home. She explained:

> *"I feel like my auntie would have never been ready to take him home, if we could have we would have delayed it and delayed it and delayed it and she I think would have. But then I had to explain the practicality of it is that he is always gonna be like this and it's just something that we've got to go home and het him used to it"* [C:Omar, I1, Hafisa, L:134].

Hafisa's explanation of her role in persuading demonstrates her perception that what was best for Omar was being at home with his family and that she felt some degree of moral duty to support Nadia to come to this same realisation. Persuading in this case perhaps played a role in providing assurance to Nadia

that she would be capable of caring for Omar at home with the support of the family. Within family persuasion also occurred in relation to Charlotte's portacath, where it was identified that Jamie's mother acted to persuade him of the need to agree to the procedure for Charlotte.

In Maisie's case, parent to professional persuasion occurred through Fiona's repeated requests for a second opinion regarding further surgery and was also evident within the medical team, where Dr Chase identified it as a necessity:

> *"People can be very fixated on what they think should be happening to that child, or which effectively is what should be happening to that anatomy, because they don't know the background as such. And so then we'll try and be very persuading to the rest of the team and we saw exactly that with her, is that you know the team are so persuading to the fact that we actually have to take it to the ethics committee to say you know actually "essentially we don't agree with what the team are saying, we want to just explore this a bit further""* [C:Maisie, I2, Dr Chase, L:280].

Dr Chase's account here demonstrates the moral work that professionals undertake for patients who they have knowledge of in relation to quality of life and who they feel emotionally involved with. She highlighted how the case can become about a disembodied anatomy and discussed the importance of trying to be persuasive to the rest of the team, inferring the need to demonstrate that the patient under discussion is a whole person who deserves to have their fate considered as a person, rather than as a series of constituent body parts. It also signals to the extent of the disagreement in Maisie's case, such that she considered it part of her moral obligation to refer to the ethics committee as a strategy for persuading her colleagues that further investigation could be considered ethical.

Hassan's was the only other case where a parent stated that they had tried to persuade the professionals to conduct surgery, and was described much more explicitly than in Maisie's case. For Hassan, there were two treatments that Salma felt she had participated in the process of persuading the professionals to undertake. She stated in her interviews that she had, for many months, shared

her concerns with the professionals that Hassan had been vomiting and was therefore not growing and her accounts suggested that she felt that she played a major role in the process of persuading the professionals to conduct surgery. In her account, she suggested that she initiated the consideration of the operation, and despite the risks that were identified, pushed for it to be done. Salma said:

> *"I was like "come on, he keeps vomiting, what is holding you back?" "Well there'll be extensive scar tissue from his duodenal atresia". I was like "but he's not thriving, he's not growing, why don't you just give him the Nissens [fundoplication]?" there was a lot "OK let's see, let's see if he vomits, let's change his milks". I said "my son is going through so much, you're going to give him [the surgery]""* [C:Hassan, I1, Salma, L:587].

Salma's account above, through her use of forceful language signals to the conflict which she experienced in trying to persuade the professionals to act in accordance with her wishes. Within Salma's accounts she articulated a feeling of responsibility for many of the positive outcomes associated with Hassan's care and development to date, and this may be why she portrayed herself as having tried to persuade the professionals of the need for Hassan to have a fundoplication. Reference to the medical notes suggested that the professionals favoured a 'wait and see' approach, as Salma identified, but no reference to Salma's preference for the surgery was documented. The medical notes suggest that eventually the clinical evidence demonstrated to the professionals the need for Hassan to have a fundoplication and jejunostomy and that this was the deciding factor for the professionals, rather than Salma's attempt at persuading.

A similar narrative was also told in relation to Hassan's eyes, with Salma insistent on the need for surgery to protect his eyesight. Salma's concerns and chasing of the plan for this was documented in the medical notes, however, again according to the notes, treatment was only given when it was considered a medical necessity, that is, when there was evidence of a corneal abrasion. Salma described the moral work that such persuading involved for her, including the need to justify her role in this process to other parents, who she felt were judging her decisions (see Section 6.6).

The other case in which persuading in the direction of parent to professional was observed was Michael's and was in relation to his schooling. Kate was keen that Michael experience true integration and did not want him to be treated as though his physical disability had any kind of impact on his mental abilities. Although the education professionals were keen to start Michael in Nurture (supported learning) groups on his transition to secondary school, Kate disagreed and worked hard to try to persuade the professionals that this was not appropriate. She appealed to the need to challenge Michael academically rather than enable him to achieve the bare minimum, which she believed would occur if he were to start in the Nurture classes. Kate tried to ensure her preference was enacted through the use of persuasion during transition meetings and during one disagreement indicated that she would be prepared to send Michael to a different secondary school if the professionals did not comply with her request:

> *"Kate appeared to be upset and said that he "doesn't necessarily have to go there""* [C:Michael, O2, L:113].

Although Kate's attempts at persuasion may have impacted on the decision to allow Michael to enter mainstream classes, he had also demonstrated ability to cope with the pace in these classes during his insight visits and this evidence may have resulted in Kate's preference being enacted. The professionals demonstrated some residual concern about how Michael would cope in mainstream classes, suggesting that there may have been some degree of acquiescence to Kate's preference, rather than feeling that this would be the best course of action for Michael and highlighted that the decision would be overturned if necessary:

> *"The Teaching Assistant identified that there are concerns about Michael managing the pace, but that he will be starting in the mainstream classes and form group at first and then they can "adapt and accommodate as and when we see fit""* [C:Michael, O4, L:18].

Although it appeared that the professionals felt that Michael starting in the Nurture classes would have been better for him, they did not perceive that it would be significantly detrimental to him to start in mainstream classes and were therefore prepared to acquiesce to Kate's preference. Additionally Kate's threats

of sending Michael to a different school and her visible distress at the idea of putting Michael in Nurture classes may have also acted to persuade the professionals that it would be better, and perhaps easier, to agree with her preference and *"adapt and accommodate"* when necessary. This demonstrates that for some issues, perhaps those that relate to more ambiguous social outcomes, the process of persuading may actually result in professional agreement with a parental decision. It also indicates the difficult moral terrain that parents tread in resisting professional recommendations; they act to achieve what they believe is in their child's best interests, but may in doing so be perceived as a 'difficult' or 'obstructive' parent, which threatens their moral identity as a good parent.

Despite the presence of persuading and the frustration that this process frequently caused families, it appeared that on the whole they did not harbour any resentment to the professionals who used this strategy. Overall parents recognised persuasion as stemming from a professional concern for their child's best interests and the need to ensure that the parent had the knowledge to make an informed decision. Salma explained:

> *"I don't think it's a bad thing because I think ultimately the decision is yours, you're going to sign that form as a guardian or as a parent of your child. They're not going to force you, they will put the pros and cons forward…but I think it's good for them to persuade, to make parents aware of what you know the pros are for this thing"* [C:Hassan, I2, Salma, L:625].

Persuasion was a frequently employed strategy whereby professionals 'drip fed' families information and persuasive comments over time, with the aim of reaching agreement and if this did not occur by when they perceived it to be necessary, eventually (albeit very rarely) they resorted to discussions of legal challenge to parental responsibility (see Section 7.3). Persuasion was identified to occur in the opposite direction also, from family to professional and within the family, at times occurring simultaneously in all directions. However, the participant accounts suggested that persuasion was not frequently effective in changing their mind and resulting in agreement. Rather they articulated it in a

way which suggested that agreement was achieved when the individual observed the sufficient evidence to persuade them of the need to agree.

6.5.2. Agreeing to treatments only when necessary

As highlighted in the previous section, the decision to commence tube feeding was controversial and families often resisted this as far as possible. Professionals viewed these decisions as being beneficial earlier in the course of the child's illness, partially as a result of their previous experience. Dr Brett explained:

> *"So something like gastrostomy placement is one of those areas where it's, I've got a couple of patients where it, for me it seems the obvious thing to do and for the professionals it seems the right thing to do, but the parents are being hesitant about it… when you put it in, you know their lives are transformed and it's really difficult because we know that that's likely to be transformative"* [C:Christopher, I2, Dr Brett, L:354].

Dr Brett articulated that for professionals, previous knowledge of the transformative effect of gastrostomies means that they recognise the need for and benefit of this earlier than families. She inferred that professionals experience moral conflict through parents' resistance to something that they claim to know will be of great benefit for the child and therefore feel compelled to push for this treatment early. Such early discussions often signal the start of the protracted discussions and strategies of persuasion. Families' lesser exposure to medical procedures, such as gastrostomies, meant that the benefits of these were much less obvious for them than for professionals. In addition parents' drive towards normality for their child meant that they would resist the treatment for as long as possible, and only agree when they determined, through their intricate personal knowledge of their child, that it was necessary.

Kay identified that despite having had discussions regarding gastrostomy insertion for 8 years, she only accepted it when she identified that the time was right, saying:

> *"I think the time was right. Might be that's why my mind never give me to do it because it, she go on life and death at the time when she's doing the surgery and when them they do want give her she was just*

> *a little teeny tiny baby at the time, so when she really get it this time her body was more bigger and stronger for it and even when she got 2 lungs collapsed, life and death, at the time because they resuscitate her in the surgery and everything, might be if she did it younger, I would lose her…So I think I'm doing this decision, this time, I'm happy"* [C:Grace, I2, Kay, L:138].

Similarly in Elena's case, the decision to insert the gastrostomy was on hold over a period of a few years. Despite numerous problems having occurred in relation to Elena's feeding in the months prior to this interview and an awareness that early insertion of the gastrostomy may have prevented these issues, Pauline identified that she was glad that she had resisted the gastrostomy previously, as it had enabled Elena to live a more normal life up until that point. She explained:

> *"I just think she wasn't ready for it and they suggested it 2 years ago. 2 years on she has fed lovely and you could argue it was a dip and she's feeding lovely again now. But yes I suppose we took it to emergency point but it just deteriorated that quick, we couldn't have foreseen this. And I actually still think it was the right decision because Elena in all of it, yeah you can argue everything's gone wrong in the last month, but she's had a normal life up to now"* [C:Elena, I3, Pauline, L:554].

For Pauline, maintaining normality for as long as possible appeared to be a key issue underpinning her decision making. She had previously discussed the progression of Elena's disease, with the full awareness that it would render her unable to do anything independently or unable to communicate. It appeared that Pauline felt that it was her duty to enable Elena to live as normal a life as possible for as long as possible and delaying any intervention which may compromise this goal was something that she felt morally compelled to do.

Treatment or investigation requiring surgery or general anaesthetic were avoided by parents wherever possible due to worry about the risks, particularly when they were not fully persuaded about the necessity. Despite any persuasion which is encountered, the data suggested that a decision will only be made when an

individual recognises that it is unavoidable, which often occurs only when sufficient evidence is available.

6.5.3. Appeal to evidence

The appeal to evidence as a strategy in the process of decision making was present in eight cases. This section seeks to explore the significant role which evidence played, as a mechanism of decision making and how it related to the moral work of decision making. It will consider specific instances where evidence appeared to be the most important mechanism in reaching agreement, particularly in cases where there had been disagreement between stakeholders previously.

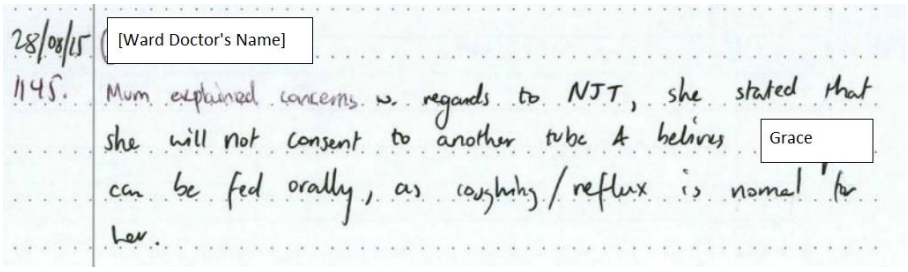
It was identified that during the process of persuading, individuals would make reference to evidence in their quest for the agreement of the other stakeholders. However, despite this, frequently such evidence was not sufficient to ensure agreement. This demonstrates a difference in the threshold of evidence which was considered sufficient for each individual to believe that the treatment was necessary. In Maisie's case, the offer of further surgery made by Dr Alexis, her initial surgeon, who the family explicitly expressed trust in, was enough to meet their threshold of evidence that something should be done. However, it appeared that this was not sufficient initially for the other professionals involved and the cardiology team required additional evidence to suggest that further investigation or intervention was appropriate. For them, such evidence was provided in the form of Maisie's quality of life and development, which was inexplicable given her anatomy and prognosis at birth. This evidence exceeded their threshold and demonstrated that further investigation may be appropriate, as explained by Dr Chase:

> *"Maisie's quality of life was the, I think she surprised everybody in having a good initial year of life and then I think as her second year of life kind of continued…I think during that second year of life, actually she, her cardiac status is not in the same place as we thought it was going to be and that actually up to date information was needed"* [C:Maisie, I2, Dr Chase, L:33].

The difference in perceived significance of evidence and therefore the evidence threshold, which signals to each individual the need for the decision, appeared to be the cause of disagreements in decision making.

Differing interpretations and privileging of the evidence

At times disagreement was not solely due to the threshold of evidence, but also differing interpretations of the same evidence. In both Grace and Elena's case, tube feeding was discussed following concerns regarding aspiration and reference to interview transcripts and the medical notes suggested that their parents did not interpret signs and symptoms in the same way as professionals. Kay felt that the signs that professionals considered signs of aspiration, were normal for Grace, as outlined in the extract from her medical notes below:


28/08/15 [Ward Doctor's Name]
1145. Mum explained concerns w. regards to NJT, she stated that she will not consent to another tube & believes Grace can be fed orally, as coughing / reflux is normal for her.

[C:Grace, Medical Notes, entry dated 28/08/15].

Similarly for Elena, the initial discussion of a gastrostomy occurred following a choking incident at school and staff were concerned that she presented an aspiration risk, however this evidence was interpreted differently by Pauline, who felt that the school had been scared and escalated their concerns too quickly. She explained:

> *"It was the first time it happened, I wouldn't class it as a choke, it was a cough and splutter what I'm more than capable of doing, you know you have to, now every child does it, we felt she was eating lovely at home, this was a bit of a drastic decision"* [C:Elena, I2, Pauline, L:40].

The escalation of concern by the school staff to Dr Price demonstrated that the evidence they had seen meant that they felt Elena was at risk of harm through oral feeding, however, Pauline disagreed and referred to her evidence of how Elena ate at home and her perception that this had been a normal coughing

episode, which she felt negated the school's concerns. Furthermore, the way in which Pauline described the incident, referring to a cough and splutter which every child does, appeared to demonstrate her need to maintain the normality which Elena had at that point, but which she knew she would eventually lose.

Similarly Kate interpreted the evidence regarding Michael's weight differently from the professionals, appealing to the fact that he hadn't lost weight, whereas professionals appealed to their concern that he hadn't gained any weight in five years. Such differing interpretations led the families to appeal to additional evidence to support their decision to delay the PEG further. Pauline's additional evidence was that Elena was eating lovely at home and Kate appealed to evidence regarding Michael's overall health, saying:

> *"you've got to look at everything, it's not just one thing that he's not putting on weight and height, it's is he active? Is he ill? In what way is it affecting everything else? You know you've got to look at the bigger picture"* [C:Michael, I2, Kate, L:377].

Kate also appealed to the family genetics as a reason for Michael's low weight and portrayed that his weight was 'normal' for their family, again demonstrating the importance of decisions which promote normality. Specifically for Elena and Michael, as a result of the degenerative nature of their diseases, it is possible that their parents were keen to retain as much normality as possible, for as long as possible, in order to delay the processes that they knew were inevitable.

The difference in interpretation of evidence provides an account of why at times there was disagreement about appropriate treatments or interventions, and consequently why persuading was employed. Predominantly when persuasion existed, it was bi-directional or multi-directional and evidence was used in various directions as stakeholders simultaneously used their evidence to support their claim and justify their opinion.

At times it was a different interpretation of the same evidence that was appealed to, but at other times different types of evidence were appealed to during attempts at persuasion. In one encounter in Elena's case, Pauline appealed to anecdotal evidence of cases from the international disease community, whereas Dr Price was seeking the source of any scientific medical evidence, demonstrating how

different types of evidence are differently prioritised depending on the stakeholder. This was characteristic of most appeals to evidence, where professionals sought out research evidence or results of clinical investigations and families appealed to anecdotal evidence or the child's behaviour.

Evidence as persuasion

Decisions regarding tube feeding were those which most clearly demonstrated the process of evidence reaching the threshold and persuading the individual of the need for action. Given the protracted nature of tube feeding decisions in the cases studied, there was a wealth of data which illuminated the mounting evidence being collected by families and professionals, and even a consideration of what future evidence would reach the individuals' evidence threshold and persuade them that the treatment was necessary.

It appeared that the need to avoid surgery was carefully weighed up by families, against the current care plan and a decision made as to which option (agreeing to surgery or not) would be best for their child at that point in time. This appeared related to their sense of what suffering they considered their child was currently experiencing and what suffering they would experience as a result of the surgery.

Two of the children had required nasogastric tube (NGT) feeding from birth and it appeared that this decision had been largely unproblematic for their families as this had always been their child's normal. Omar was fed via a NGT from birth, due to an unsafe swallow and discussions around gastrostomy insertion occurred when he was semi-electively admitted to hospital to try to stabilise his condition. Nadia stated that she had recognised that the NGT was not sufficient, given the problems with Omar's poor weight gain, vomiting and frequent need to repass the tube and that this prompted discussion about a gastrostomy. Nadia stated:

> *"The dietician came to see us, and that's when we told her that, "look, what's going on with his milk? Because he's not going to put on no weight, no nothing because, this is just not working" and Omar has a tendency to go like that [demonstrates swiping her hand near her nose] and he kept bringing it out and I was always forever putting one back in. And that was when I spoke to the dietician about it and*

> *she said that somebody would come and talk to me while we were in hospital, about having a gastrostomy"* [C:Omar, I1, Nadia, L:543]

Nadia here discussed her awareness of the need for a different approach to Omar's care even before the gastrostomy was ever mentioned, because of the problems associated with his NGT. Following discussion with the Stoma Nurse Specialist and some reassurance about the reversibility of the gastrostomy, Nadia identified that:

> *"We didn't really think much about that, we thought "yeah go ahead and do it""* [C:Omar, I1, Nadia, L:595].

This suggests that the evidence of Omar's condition and current care plan, and the reassurance from professionals, provided her with sufficient evidence to reach her threshold and demonstrate to her that the gastrostomy was necessary and would be better for Omar. It appeared that a similar process had occurred for Hassan, given Salma's lack of discussion of the NGT or gastrostomy as a significant decision for her. The moral work which Salma undertook in decision making for Hassan was aimed at preserving his life and ensuring his optimal development, regardless of the surgical and technological implications. Hassan in particular required a number of invasive procedures during his infancy and it appeared that the gastrostomy was not a decision that Salma viewed as significant or difficult to make, which was likely due to the evidence of his gastrointestinal anatomy, lack of a swallow reflex and the risks posed for oral feeding by his cleft palate.

Continual re-evaluation of evidence

For the other children in the study who tube feeding was relevant to, there had been significant resistance to the decision expressed by the families. It appeared that this related to the need to maintain normality and engage in social bonding through eating together and time spent feeding the child, in addition to emotional factors and lack of understanding about the potential medical consequences of resisting tube feeding. In these cases, time to agree to tube feeding was extended whilst families waited for a sufficient amount of evidence to demonstrate to them that the decision to commence tube feeding was necessary.

Christopher's case demonstrated how continually mounting evidence persuaded his family of the need for tube feeding. The initial evidence of Christopher aspirating, had been observed by a professional and was unrecognised by Susan, but resulted in the order to discontinue oral feeding. Following investigations, which demonstrated that Christopher had an unsafe swallow, he was discharged home with a NGT. Susan described that initially she was resistant to the idea of tube feeding, but that the evidence that Christopher wasn't eating well, alongside the results of the investigations provided her with sufficient evidence to understand the need for the NGT. A prolonged period of NGT feeding, caused her to reassess the evidence and even though she had been dreading the operation, enabled her to recognise that a gastrostomy was necessary. Susan explained:

> *"we knew it had to be done, because of the NG tube, we noticed what a difference it had made, because he hadn't had a seizure, he hadn't been coughing, he hadn't had a chest infection, and his weight, he put a stone on in a week, you know it was just like, "ah wow", so there was big like positive and we thought well we can't go back now to, and I didn't want to keep this in his nose, cos it just, cos it was a nightmare"* [C:Christopher, I1, Susan, L:202].

Susan had recognised that the NGT had reduced Christopher's suffering through hospitalisations due to seizures and chest infections, but that the NGT was not a viable long term solution and had its own complications resulting in some degree of distress for Christopher and the family. Susan's use of language articulated her inability to return to oral feeding for Christopher and implied a moral imperative for her to agree to the gastrostomy, as something which would be in his best interests. In doing so she was able to portray herself as a good parent in championing Christopher's overall best interests.

This continual process of re-evaluating the evidence was observed to occur in different ways in different cases and for different decisions. This linear form of continual assessment culminating in the enactment of a decision, as evident in Christopher's case, appeared to be the most common form and was also observed in relation to tube feeding in Elena's case. Elena's family had initially

resisted the gastrostomy, because the evidence suggesting it would be beneficial had not initially met their threshold. However, during the study, the evidence suggesting that Elena required tube feeding mounted and by the end of the study Elena had had an NGT inserted. The longitudinal nature of the study meant that the mounting evidence over time was visible, as was the moral distress experienced by her parents when deciding whether to further delay or accept tube feeding. Following a virus and overnight stay in hospital, Elena's feeding skills further declined and persuaded Pauline that tube feeding was now a requirement. She said:

> *"Then she had that bout in hospital when she went in with stomach pain and stuff and to be honest she never, I think she took a big dip then. Cos she came back and she was only eating 3 mouthfuls at a time and it just seemed you were finishing one feed when you were preparing the next one. She was hardly drinking and it just went on for weeks. And it just got to the point where it was, I hated feeding her, it was, I hated it, it was just so stressful, so I spoke to Tracy [Community Nurse] and we worked out that she wasn't really getting enough anyhow, so we went for the nasal [tube]"* [C:Elena, I3, Pauline, L:63].

Pauline was very honest about her own feelings about the difficulty of feeding Elena, but was quick to clarify that Elena also needed tube feeds for her own benefit, so as not to appear to be agreeing to this for selfish reasons. Then subsequent to the insertion of the NGT, issues associated with its management, and her awareness of the degenerative nature of Elena's illness, appeared to provide sufficient evidence to demonstrate to Pauline of the need for a gastrostomy, which Elena is currently awaiting. The extract also alluded to the emotional and moral work that parents carried out during their continual re-evaluation of the evidence relating to their child's treatment.

For Michael, consideration of the gastrostomy was still being firmly resisted at the end of the study, but appeared to also take the linear form with regards to evidence re-evaluation. Unlike any of the other cases, Kate shared her predefined threshold of evidence which she believed would act to persuade her

of the need for Michael to have a gastrostomy. It appeared that the process of re-evaluation in this case would continue linearly until this threshold of evidence was reached. Kate recounted an appointment where a doctor had asked her what would make her think that Michael needed a gastrostomy and she told me that she replied:

> *"if he was ill often, if he was becoming more ill, if he stopped being as active as he is and if he wasn't gaining any weight or height, then yes definitely" but he is gaining weight and height, it's not very good but he is doing it, he eats really well he's so active still and he's hardly ever ill"* [C:Michael, I1, Kate, L: 427].

Michael's overall health and activity levels were continually appealed to in her justification for refusing the gastrostomy for him and followed the linear process. At the end of the study Kate's predefined evidence threshold was not reached and therefore the gastrostomy was still refused. This example demonstrates that even when a treatment is being refused, continual assessments may be being made against the family's criteria for agreement, in order to determine if they are making the right decision at the right time.

Although this linear form of re-evaluation was most frequently observed during the study, instances where it took a more dynamic form, moving in one direction and then back in the other, were noted. Charlotte's case was the one in which this was the most explicit and occurred in relation to NGT feeding. Mounting professional concern, clinical results and medical evidence eventually provided sufficient evidence for her family to accept the NGT. However, subsequent to this decision being enacted, the evidence of how Charlotte was coping and the negligible impact that it was having on her nutritional status demonstrated to all stakeholders that a re-evaluation was required and the decision was reversed. Rachel and Charlotte explained:

> *"Rachel: it was horrendous, but not only that, when she had it in, one night it came out, so that traumatized her.*
>
> *Charlotte: Through my mouth so that was so gross…*

> *Rachel: And it was restricting you clearing your secretions wasn't it when you were doing physio and it got to the point where Jamie said "right you got to tell them this isn't alright for her" and also, what was also happening was Charlotte wasn't eating food. So she was having the overnight feed, but then she was full…So she wasn't gaining anything because the calories that she'd been having previously, with food in the daytime, were just replaced by the overnight feed and she wasn't eating in the day so she wasn't gaining any more calories. So we'd swapped real food for overnight food. So we said this just isn't making sense at all and the hospital completely agreed"* [C:Charlotte, I1, Rachel and Charlotte, L:279].

This extract demonstrates the nature of evidence as a mechanism in decision making, which is continually evaluated. In this case, the significance of the medical argument, which had served as evidence to persuade the family to try the NGT, was outweighed by the physical evidence of the impact of the NGT on Charlotte, from a holistic and medical perspective. The re-evaluation of the evidence demonstrated that the NGT should be removed. Rachel explained that they felt that they had a moral duty as parents to prevent this from continuing and to ensure that the decision was reversed in order to prevent any further distress.

The appeal to evidence in decision making has been demonstrated as a complex weighing of a variety of different types of evidence which all hold different levels of significance for the individual stakeholders. Therefore it appeared to be difficult for the stakeholders to understand the others' perspectives, when the evidence that each valued, was not recognised as so valuable by others.

6.6. Family justification of decisions

Justification appeared to be an important process in the moral work associated with decision making and often entailed a reference to best interests. Predominantly it was identified in relation to parental decisions and the reasons for agreeing to or refusing treatments. It was also identified in the professionals' accounts, but only when reference to best interests was made. Professional justification will therefore be discussed in the next chapter (see Section 7.3.1).

Justification appeared to be an important mechanism used by families as a way of feeling at ease with their decision and was potentially more prevalent when some degree of guilt was experienced. At times participants formed a particular narrative which they used as a means to justify their decisions to me as the researcher, other stakeholders, family members, and also perhaps to themselves.

Although parental justification was used in nine of the eleven cases, it appeared most prominent in Michael's case in relation to Kate's refusal of a gastrostomy for him. She used justification frequently and had developed a narrative which was relayed to me almost word for word multiple times during the study, about the family genetics and Michael's good general health. For Kate, it appeared that she felt conflicted by the decision not to agree to a gastrostomy for Michael yet, as she recognised the benefits it may have for his health, but experienced Michael's frustration about the topic and his absolute refusal to agree to the surgery. Kate herself identified:

> *"Now I do question myself now big time now on the PEG, but I think Michael's doing very well, he is very active and it's not just one factor you know. He's not putting on much weight and he's not growing very tall. Well the whole family on this side are all midgets but as they've gotten, they've all been lean, if you saw my, you've seen [Michael's Sister], you could count her ribs if she stretched, it's just our build, none of my family are big. Michael's side of the family, Dad's side, they're small, ours are thin. They become quite big when they're older, so I'm thinking maybe that will happen with Michael"* [C:Michael, I2, Kate, L:371].

The beginning of this extract demonstrates Kate's uncertainty about the decision she has made, but then immediately transitions onto her rehearsed justification of why she thinks Michael does not need to have a gastrostomy yet. Given the conflict which Kate herself identified she was experiencing, it appeared that alongside the need to justify her refusal to the professionals, the narrative displayed above appeared to also act as a justification to herself at times when she was unsure as to whether she was making the right decision. Kate also appeared to appeal to best interests as another justification for her

resistance to the gastrostomy, speaking about the life-limiting nature of Michael's condition and hence her desire for him to maintain a good state of emotional and psychological wellbeing, something which she believed would be impacted upon if he was forced to have the surgery. It may be that her conflict between her need to meet his physical health needs and protect him emotionally, required her to justify this decision; especially as she began to recognise the potential benefits that the gastrostomy may have for him.

Acknowledging the child's emotional wellbeing as a justification in decision making was also used by Jamie in Charlotte's case around her request to have her spleen removed. He explained:

> *"I don't like her going under the, under a general anaesthetic. I'd like as little intervention as medically necessary, would be my starting position, from a purely risk management position really and obviously her immune system is better served with the spleen in than out, but…that's balanced with if Charlotte really wants it out and it's making her unhappy because of, you know, she's gonna get interested in boys…and you know having a tummy that sticks out and a huge bloody spleen is not a great thing. And I think my view is you know her life expectancy is not what it would be with a normal child, I mean you know I want her to have the best life possible whilst she can…you know I want her to have the best quality experience she can and if she's just unhappy with her body image all the time then that's not great. So if, so my view would be tempered, the medical, the risk, sort of risk view would be tempered by what Charlotte really wants, and if I felt that she really wanted it and the medics said "yeah it's OK, we're OK with it" then I'd support her"* [C:Charlotte, Jamie, I2, L:97].

The reference to emotional or holistic wellbeing of the child as a justification was used in many of the cases, but was most apparent in Charlotte and Michael's cases as they were able to effectively communicate their wishes, desires and feelings. In these cases where the child's view was different from the parents' or professionals, it appeared that justification was a big part of the moral work undertaken by parents in making, and living with, their decisions. It spoke to the

need to enable the child to develop choice and agency and have some control over their bodies, which was, to some degree, being removed from them by their diseases.

Justification was also observed in Maisie's case as a way to overcome the fear of a decision to be made. Fiona discussed feelings of conflict between her head and her heart for decisions regarding further investigations and surgery and appeared conflicted about what was the right thing to do. The longitudinal nature of the study enabled the fluctuating nature of decision making to be witnessed and it was only once surgery became a real possibility that Fiona began to use justification in her decision making accounts. Her justification referred to her moral imperative as a parent to make decisions which promoted life. Fiona stated:

> *""I have to give her a second chance at life", saying that if she didn't do it who else would"* [C:Maisie, O5, L:47]

She also made significant reference to her trust in Dr Alexis as a justification for consenting to a surgery which carried risks. The changing offers made to Fiona by professionals (see Figure 8, p.113) made the environment for decision making difficult and despite being able to initially justify her agreement to surgery in relation to trust in Dr Alexis, the plan changed and he was no longer able to be present. This required an alternative justification and despite having previously stated that Dr Alexis would be the only doctor she would allow to operate on Maisie, she was forced to accept that another surgeon would have to carry out the operation. The trust in Dr Alexis that she had previously used as a justification, had been partially withdrawn from her, but she was able to modify this to still enable her to accept the offer of surgery and continue to justify her decision with reference to that same trust. Fiona said that:

> "*she would rather the catheter next week as initially planned and acknowledged that Dr Alexis would not be here but that she knows that he would trust the person that he was leaving to do the procedure on Maisie"* [C:Maisie, O6, L:116].

This extract appears to demonstrate Fiona's need to justify her decision in relation to trust, such that she was able to place her trust in the surgeon who

would operate, using her trust in Dr Alexis as a proxy. This prospective justification through reference to trust enabled Fiona to agree to the surgery for Maisie, which was something which she wanted, but felt conflicted about due to the risks.

Justification predominantly occurred within the child's network of care, however in Hassan's case, Salma identified the need to at times justify her decisions to others outside of this network. She identified that parents whom she had got to know during Hassan's hospitalisation at times questioned her decisions and she felt the need to provide them with a justification. Salma said:

> *"A few of the parents were quite yeah abrupt with me but I said "it's not your decision and it's not your child", "why are you putting him under the knife again?" "because he needs it, it's not your problem and it's not your child" and then once he's done really well after that and I'm like "look, I told you so, look at him now, look how well he's doing". What's the lesser of 2 evils, do you let your child suffer for so long?"* [C:Hassan, I1, Salma, L:652].

Salma's justification in this case made reference to the fact that Hassan was her child and it was therefore her decision. Her justification also made reference to the outcomes of her decisions, which had been positive and therefore enabled her to accept her decisions which may not have been ideal, but which she perceived to be the 'lesser of two evils'.

Justification by the family appeared to be an important part of the process of decision making, which involved a great degree of moral work and implicit reference to the child's best interests. Parents were required to make significant decisions for their life-limited children and needed to be able to live with those decisions. Justification was a mechanism through which they were able to feel comfortable with the decisions that they had made and were able to portray themselves as good parents acting in their child's best interests, particularly in situations where they felt conflicted regarding the best option.

6.7. Conclusion

This chapter has presented a novel insight into the concept and process of decision making. It has outlined the different types of decisions, articulating how

decision making takes on different characteristics and is differently manifest in different situations, over a long period of time. It has demonstrated issues around how choices are communicated and the perceptions of families and professionals in relation to their feelings of moral accountability for decision making. The strategies for achieving agreement have been discussed, with persuasion often used, albeit not recognised by participants as a factor which significantly influences their agreement. Rather re-evaluation of the evidence was determined to be the factor which led to agreement for a previously disputed decision, however relevant and persuasive evidence took different forms for different stakeholders. Although evidence was seen to lead to agreement, at times protracted disagreements were visible and an appeal to the concept of best interests was made, as will be discussed in the following chapter. Justification was a significant factor for families during the process of decision making and subsequent to it, and it enabled individuals to make decisions which they felt conflicted about and to live with decisions which might cause them to experience guilt. This justification was a way in which the individual maintained their own sense of moral responsibility and displayed this to others.

Chapter 7. Findings Theme 3 - 'Best interests' and professional use of moral justification

7.1. Introduction

This chapter will consider the concept of best interests and how it is perceived by the various stakeholders involved in decision making and discuss their views on who is best placed to make assessments of best interests. It will consider how appeal to the concept of best interests is used as a strategy in decision making when there is protracted disagreement and growing concern about harm. It will show that discussions of best interests signal to a feeling that compromise may soon be no longer possible and therefore reference to the legal boundary of parental decisional responsibility is made. It will also consider how relationships are maintained when difficult discussions, referring to the possibility of escalation to the courts, are had. Finally the chapter will consider how justification is used by professionals and how this signals to the moral work they undertake in decision making.

7.2. The concept of best interests

7.2.1. Understanding best interests

The previous chapter alluded to the role of best interests in decision making and appeared to be a concept which was referred to during disagreements, but was only formally invoked when these disagreements were protracted and professionals grew concerned about harm to the child. It became clear that the concept was different for families and professionals, with professionals conceptualising it both as a health and wellbeing issue, but perhaps more so as a legal concept, particularly when the potential for harm was highlighted. Families' discussion of this concept however, more frequently involved reference to the family values, child's overall health and a consideration of their psychosocial wellbeing.

Families' understandings

Families did not often use the terminology of 'best interests' but more frequently referred to decisions which were best for the child, in relation to their values and

goals. Pauline's assertion in relation to tube feeding for Elena demonstrated how the family considered her overall interests in relation to this decision:

> *"It was going to be a big thing for Elena, and also she loves eating, that's her thing. So to us it was, actually we felt it was disabling her before her time."* [C:Elena, I2, Pauline, L:47].

Her discussion of eating being Elena's 'thing' shows how as a family, Elena's best interests were assessed with significant focus on the enjoyment that she derived from the normal things in life. Similar foci were present for many of the other cases when there was any discussion of best interests. For Ella, best interests were described in relation to the management of her abdominal discomfort and reflux. During a discussion when the possible treatments were discussed, Jill felt that the option of continual jejunal feeds would not be in Ella's best interests, as described in the following field notes extract:

> *"Jill said that she did not think it would be in Ella's best interests to be hooked up to food all day and that she thought that they needed to tackle the bowel "rather than rushing to" surgery. Both Jill and the carer then had a discussion that the doctors missed the day to day understanding of Ella in relation to this decision and that they see a narrow field, not holistic picture of how the decision may impact on Ella and the family"* [C:Ella, O3, L:86].

This extract from a clinic appointment demonstrated that families believe that they consider best interests in a much more holistic way than professionals do.

Despite most families being relatively unaware of the legal conceptualisation of best interests, Jamie appeared to be acutely aware of this and the boundary of their decisional responsibility as parents and was the only non-professional participant to allude to this. When asked about who had the ultimate responsibility for decisions for Charlotte, Jamie replied:

> *"You know, in terms of the law, I mean she's under 16 so I, my understanding of the way this works is that ultimately the medical profession will act in her best int-, will do what they think is in her best interest and if they believe that the parents are trying to take*

> *decisions or stopping things happening that are jeopardising her health, then there's recourse through the courts, right? For the doctors, for the hospital to get those decisions taken"* [C:Charlotte, I1, Jamie, L:186].

This extract demonstrates Jamie's awareness of the limits to the family's decisional responsibility and the concept of best interests. Interestingly during this comment he appeared to rephrase it half way through and went from stating that the professionals would act in Charlotte's best interest, to stating that they would do what they thought was in her best interests. This comment suggests that he may not always agree with the professional assessment of Charlotte's best interests and may signal to why disagreement occurs when various stakeholders all simultaneously claim to be acting in the child's best interests.

There were only two other cases in which families demonstrated awareness of the professional concept of best interests. Maisie's family were aware that professionals felt the need to discuss her case at an ethics committee, to be sure that it was ethical and in her best interests to offer surgery. In Omar's case, his cousin Hafisa, who was studying to become a midwife and therefore had some medical understanding, alluded to best interests. She highlighted that the knowledge about the child's best interests and the emotions that these present for the family cause conflict in decision making. Hafisa stated:

> *"I know that they know, the doctors, normally when they come up with these plans they know and they know what's best for that individual person. But I think when it's your own member of the family you still feel like "no actually I want you to try"...But in Omar's condition I did know that it probably wouldn't be the best thing for him to keep resuscitating him and doing all that because of how fragile he was and how poorly he was"* [C:Omar, I1, Hafisa, L:258]

This demonstrates that for family members, they may be aware of what the 'best interests' decision would be, yet may struggle to agree to it, or at the very least feel conflicted in doing so, due to emotional factors.

Professionals' understandings

Professionals often referred to the concept of best interests in their interviews, particularly in relation to decisions for which they believed there was one best course of action, or in situations where the family had requested a course of action which they felt was inappropriate. The predominant professional view, articulated in this excerpt by Dr Price, was that in order to make best interests' decisions, they need to constantly consider:

> *"Benefits, burdens, risks to the child and risks to my relationship with the family"* [C:Maisie, I2, Dr Price, L:276].

This was exemplified in relation to tube feeding by Dr Price who explained its relation to physical health and the risk of harm:

> *"To me the clinical picture is saying this child is not safe to feed this is distressing the child, this is wrong and not in the child's best interests and that the child needs an operation to keep her healthy and safe. So I've got a lot of evidence and a lot of other people's opinions and the evidence with my own eyes and my medical knowledge, interpretation all of the clinical signs and features which say to me this is the best thing to do"* [C:Grace, I2, Dr Price, L:218].

Similarly the best interests' concept was invoked in relation to emergency health care plans, when there was consideration of limiting resuscitative efforts. Dr Price discussed this in relation to Maisie's emergency health care plan:

> *"When I am in a situation where Maisie is not able to run and walk anymore because she is that breathless, she's just in bed on as much oxygen as I can get her and on regular Furosemide to try and keep her heart from filling, keep her lungs from filling up with fluid, then I will say to her, "I'm not going to do that to her anymore". "We're not going to do that"…But I take it step by step and I live with what's not the ideal plan as far as I'm concerned, as long as it's not going to do Maisie any harm. So I can only negotiate or compromise to a certain extent and I can live with the that we are going to do, going to try cardio-pulmonary resuscitation, that's actually the least of my worries, it's not going to work, the kiddie's going to be dead, it's not*

> *going to. You know I would like for the family to have a different ambience around a baby's passing, I would like them not to keep bringing her back to die again if we can you know, I don't want to prolong her death, but that's not a big deal to me, if they want to try, that's fine. But I don't want her to die on a ventilator."* [C:Maisie, I1, Dr Price, L:425].

Again the appeal to harm is raised in the professional discussion of the child's best interests. This extract from Dr Price provides an insight into the morally complex issue of best interests, as indicated by her apparent conflict in whether it would or would not be appropriate to resuscitate Maisie. She stated that such an attempt at resuscitation would not be in Maisie's best interests, but simultaneously accepted that she would be unlikely to come to any harm, as she would already be dead and she believed that resuscitation would be unsuccessful. It appears in such cases, she may permit resuscitation attempts in the knowledge that this may be comforting for the family to know that everything had been done to try to save her. In this way, it appears that when the child is not at risk of experiencing harm or suffering, the concept of best interests' may be applied to the wider family, rather than just the child.

The foregoing discussion serves to demonstrate that professionals and families consider the concept of best interests differently, and at times the data demonstrated that even when having joint discussions about what was in the child's best interests, professionals and family members came to different conclusions. It appeared through reference to these cases, that families considered best interests' decisions more holistically than professionals who assessed the concept primarily in relation to the child's physical health.

7.2.2. Who is best placed to assess best interests?

The difference in family and professionals focus in best interests' assessments at times meant that different conclusions were made by different stakeholders. It was identified by professionals and families that best interests' decisions should be made jointly. Jamie identified the importance of the family understanding the purely medical view, but also highlighted the importance of the professionals

recognising and valuing the holistic and patient specific aspects and consequences of the decision. He said:

> *"If I think it's a purely medical, technically medical decision I would feel very uncomfortable getting too stuck into it, if I think it's got a much broader range of issues then I'd be happy to get stuck into it and weigh the pros and cons. I think with those kind of decisions, I think equally the medical profession need to take a, need to not just take a purely medical black and white view, they need to take a balanced approach. So using nasal feeding tubes in Cystic Fibrosis patients, you know I read a couple of research papers that came out of the States because the previous debate was the nasal stuff. And there was a very black and white medical research paper that came out of one of the US doctors, it basically said this is the research, we've done, we've had a couple of hundred patients and there's a clear clinical improvement and you know it should be mandated that you use nasal feeding tubes in Cystic Fibrosis patients with these conditions…Well I can tell you with absolute clarity that using a nasal feeding tube with Charlotte had negative effects, not positive effects. She didn't put any weight on, she didn't it didn't have any benefit, I mean it was horrendous. The whole experience, including the medical outcome was rubbish. So that medical factual black and white position in that scenario in her instance just wasn't correct. And all the broader issues that we'd raised were valid…the broader issues outweighed the pure medical issues with the nasal feeding tube. It's not just a clinical sort of decision, it's got to be a balanced decision you know looking at the psychology of the patient, and its patient specific as well and it depends on the individual"* [C:Charlotte, I2, Jamie, L:225].

This extract demonstrated Jamie's awareness that each stakeholder comes to the decision with different knowledge, experience and expertise and that these all need consideration in decision making. He also made reference to the importance of focus on the individual, something that all families referred to as essential. Many parents felt that, as Jamie highlighted above, research evidence or professionals' clinical experience meant that they tried to treat all patients with

the same disease or symptoms similarly, but that each child is an individual and decisions need to be made with this in mind.

Professionals also highlighted the need to consider a variety of family factors in assessing the child's best interests and that this should be done jointly with the family. Dr Price recognised the need to combine professional and parental knowledge when making best interests' assessments, saying:

> *"I have to try to share as much as I can with them but because they've not trained as a doctor and they're not scientifically thinking, then some of it they won't ever understand or know. They know Maisie better than me, they know their family resources and their resiliencies and what they can deliver and what they can't deliver as a family…They know their feelings and how much they're willing to give or what they can give to the whole thing, which I can't ever know fully. So they bring what's right for our family and also "what we believe is right for Maisie based on our cultural background and our knowledge of the child" that they will, can try and explain to me but I will never fully know"* [C:Maisie, I1, Dr Price, L:292].

This demonstrated Dr Price's awareness of the need to communicate and share such assessments and decisions with the family, and coming to a conclusion which incorporated their views, cultures and values. However, despite this commitment, there were limits to which the family views, cultures and values would be incorporated into best interests' assessments and therefore decision making. Considering the professionals' accounts, it appeared that these views would only be taken into consideration in situations when the professional was uncertain about the best course of action or when the family view was commensurate with the professional view. In cases where the professional felt that there was only one viable option which protected the best interests of the child, from a physical perspective, the professional commitment to integration of family culture or values appeared to be in principle only. In such cases persuasion occurred through continual communication and discussion, during which professionals aimed to ensure that the family eventually agreed with their assessment of the child's best interests. Dr Price explained:

> *"A lot of it is just about not having all the information and it takes time and when you're not on the same vision at all it's generally because you haven't really understood where they're coming from or they haven't really understood where you're coming from. It's the same with any kind of stale mate or whatever is that there's, you've then got to just listen some more to that other person's point of view, try to understand why it is that they've made up their mind that, or have they made up their mind then or. And sometimes just take them on a little journey step by step to get to where you want them to, so you know just negotiate and say "well we can't agree today but what we'll do is just carry on in this way because I can live with it, you can live with it and we'll look at it again a bit further down the line when you've had a chance to see what I can see, which you can't see at the moment""* [C:Maisie, I2, Dr Price, L:303].

This extract demonstrates that professionals are able to compromise in relation to some best interests' decisions from a time point of view, until the family eventually agree with their assessment and decision. It appeared to involve significant professional attempts at persuasion and seemed to suggest a professional opinion that once the family come to assimilate the information provided to them by the professional, they will arrive at the same conclusion. However, the families' accounts suggested that this was not the case. Nevertheless, despite Dr Price's earlier statement that she could never fully understand what the family know, her reference to the need for persuasion indicates that when professionals have a conviction that something is in the child's best interests this becomes their priority. Consequently any commitment to integrating the wider family values into best interests' assessments become of secondary importance.

Although professionals discussed their assessment of best interests differently from families, Dr Chase articulated that she believed that the assessments were fairly similar:

> *"I don't think it is that different, it's quality of life"* [C:Maisie, I1, Dr Chase, L:122].

Quality of life can be a nebulous concept however, and as previously stated, families may prioritise psychosocial aspects of the child's quality of life, whereas professionals prioritise physical health, something which Dr Chase did not identify as problematic. Her reason for discussing best interests in this way may have been due to the focus on Maisie's case, where there was significant uncertainty, no one best course of action and therefore the decision would be determined by the family's assessment of Maisie's quality of life and their goals and values.

This section has shown that professionals and families feel that assessments of best interests should be made together, but that disagreements may occur. Participant accounts suggest that families prioritise the child's overall wellbeing, whereas professionals often focus more on the child's physical health. When professionals make an appeal to the child's best interests, they often invoke the concept of harm and perceive a moral duty to act on behalf of the child and protect them from harm.

7.3. Invoking the concept of best interests and the legal implications

In three cases it was identified that professionals invoked the legal concept of best interests and inferred the possible legal implications of the family's continued disagreement. These cases were all in relation to discontinuing oral feeding and/or initiating tube feeding. In Elena, Michael and Grace's cases, professionals had felt that the child's health may be compromised through sustained disagreement and therefore the legal limits to parental decisional responsibility were highlighted to their families.

In Elena and Michael's cases, reference to the limits of parental decisional responsibility were made in the midst of stressful discussions. In the initial discussion about a gastrostomy for Elena, Pauline described Dr Price's assertion around the decisional responsibility:

> *"I did get the thing "look, if you don't agree and I do agree, I will be making the decision". We did come up against that. It was quite a strong intense meeting that time, cos I think she felt it was right"* [C:Elena, I1, Pauline, L:265].

This demonstrates Pauline's feeling that this challenge stemmed from Dr Price's sense of doing the right thing, suggesting an awareness of the moral work that Dr Price was undertaking in the process of decision making. Similarly for Michael, despite Kate's initial assertion that Dr Price's involvement in the discussion had some persuasive effect, her continued resistance since then meant that Dr Price apparently needed to intensify her persuasive attempts and subsequently drew upon the legal implications of an enduring disagreement. Kate felt that she had been made aware of these legal implications in an appointment with Dr Price when they were discussing the PEG. She explained:

> *"There was a bit of pressure there because she said "right well we're going to have to have a meeting about it". So that offended me a bit because I thought, you know what, we've been through this for so many years, so I just turned round and said "what is it then, will it be court?" a bit like that…But the thing is. She was a bit shocked, because I don't think she realises how serious we are about it, we're not taking it lightly"* [C:Michael, I3, Kate, L:311].

Kate's assertion that Dr Price was a bit shocked may be because Dr Price had not meant it in the way which Kate interpreted it or alternatively Kate's growing awareness of Michael's static weight and probable need for a gastrostomy in the future may have resulted in this defensive stance where she boldly challenged Dr Price. This demonstrates Kate's awareness of the legal implications regarding such disagreements and potentially a recognition of her perceived moral obligation to go to court and act in what she believed was Michael's best interest.

It appeared that such legal references were only highlighted when the professional had significant concerns, demonstrating their need to act in any way necessary to protect the interests of the child. It appeared that such references, despite not resulting in immediate agreement in two of the three examples in this study, perhaps served to act as another factor to persuade families that the need for the treatment was approaching. This may have occurred through the recognition that the issue was becoming more serious, as their trusted professional was prepared to challenge their decisional responsibility, something which was perceived to be antagonistic.

For Grace however, legal means had been employed in decision making, with Kay having been forced to sign a contract agreeing to no longer feed Grace orally. Her account demonstrated an awareness of the possible outcome of her refusal to sign the contract, which was not an outcome that she was prepared to accept and she was therefore persuaded that signing the form was the only option. Kay explained:

> *"Glenn was threatening me and said he's going to get solicitor and if I don't sign the paper and if him leave here today I'm gonna sign it, I'm getting solicitor to look into it. And I know what it's coming down to, to taking my child from me"* [C:Grace, I2, Kay, L:244].

This example demonstrates the threat of legal action as a means of persuasion or perhaps coercion, but also again about the communication of choice and a perception of being powerless to act in a different way. Given Kay's minimal level of education and knowledge of the health and social care system, it is likely that she felt powerless to challenge this command. Additionally, in order to maintain overall parental responsibility for Grace, Kay appeared to recognise that she was required to defer to the professionals in this instance. Kay was aware that not agreeing may have resulted in removal of Grace from her custody and the subsequent lack of power to make further decisions on Grace's behalf, something which was unacceptable to her.

7.3.1. Professional justification when invoking the concept of best interests

As in the family accounts, justification was a key part of the moral work in decision making for professionals. However for them, justification primarily occurred in relation to conflicted cases where the legal implications of continued parental refusal were being discussed. Justification in these cases explicitly involved moral work, as professionals referenced a duty to safeguard the child, regardless of the values or preferences of the parent or wider family. Grace's School Nurse, Alice justified her role in the termination of Kay's ability to feed Grace orally, through reference to an overall professional consensus that this was necessary and thereby appeared to dilute her feeling of responsibility for this decision. Alice explained:

> *"Mum has got to be able to know when that [oral feeding] is appropriate and when it is not and the consensus I think for all professionals is that she doesn't and that's the difficulty…Whether it'll change in the future, I hope it does, but from working with Mum for a long time with a lot of other professionals and trying to get her to understand that, I don't think it's ever going to be safe really"* [C:Grace, I2, Alice, L:272].

In this case Alice justified her actions through reference to Kay's lack of understanding potentially jeopardising Grace's health. Given the trusting relationship that Alice had built up with Kay over the years, it appeared to be as important for Alice to feel that she had not betrayed Kay, as it was for Kay to avoid feeling betrayed by Alice. This was achieved by Alice diluting her feeling of responsibility, through reference to a professional consensus, and Kay transferring all the responsibility to Glenn for the decision, thereby sparing Alice from this.

For Dr Price, justification was used in relation to best interests. For decisions where she perceived the child's best interests were at risk of being compromised and they were being placed at risk of harm, she referenced a duty to intervene:

> *"I can't do harm to a child and I can't allow a child to come to harm, because that's as a paediatrician I can't, I'm not allowed to. If a child is under my care and the family are harming him, I have a responsibility. If the child is under my care and I do harm to that child, I have a responsibility*" [C:Maisie, I2, Dr Price, L:254].

Dr Price believed that primarily such situations come from a lack of understanding and that perhaps is why the process of persuasion occurs over such a long period, so that this lack of understanding can be overcome and conflict subsequently be avoided. Despite the reference to legal proceedings as a possibility, it appeared that these were mainly made to make the family aware of the final route for the professional, but all of the professionals interviewed identified that they had never had to take a case to court[24].

[24] The contract which Kay was forced to sign in relation to terminating oral feeding Grace was a step prior to needing to take the case to court.

Dr Price also justified her behaviour in working with Kay to convince the surgeons to agree to perform the gastrostomy surgery as something which will be beneficial for Grace in the future. She stated that the relationship which had been built up through doing so, may mean that in the future, Kay may be more likely to go along with her recommendations:

> *"It was the mummy and the me pushing the surgeon to do what we wanted and so I think and having been through that kind of experience where you're supporting her to get what she perceives her child needs, makes me in an even better position to persuade her to do the next thing that I want to persuade her to do if that's the case"* [C:Grace, I2, Dr Price, L:132].

At first glance this statement may appear manipulative, however, in relation to moral work, Dr Price believed that this would produce the best result for Grace and would therefore be prepared to act in a way which objectively seems immoral (due to the manipulative nature), but comes from a true concern for the welfare of the child. She expressed a strong moral imperative of professional responsibility to prioritise the child's needs and ensure that they do not come to harm. Justification for professionals appeared to be an important aspect of the moral work that they undertook. Their primary concern was for the child, who was their patient, and any challenge to the relationship which they had built up with the family, was justified through reference to their primary duty to safeguard the child's best interests, even if this meant application to the court or placing a ceiling on resuscitative efforts, in order to prevent harm.

7.3.2. Containment within relationships during best interests challenges

Despite the pressure often felt by parents in the process of persuasion it was mostly not negatively evaluated by families. Even in the cases where professionals had referenced the possibility of escalation to the courts, parents excused the professional's actions, enabling them to maintain the trusting relationships that they had developed. In Elena's case, Pauline provided her interpretation of the discussion that Dr Price had had with her, saying:

> *"Well I think as well, I think I personally caught Dr Price on a bad day because what happened is we got called in within 24 hours so it*

> *was an emergency appointment, Richard was away. It was something that I knew this was going to be a serious chat with Dr Price, I brought a friend with me and my friend was sort of like saying the same as me "Elena's fine" and I don't know whether Dr Price took a dis-, but them two didn't get on and it was a real tense meeting and we came away and she went "I don't like her" and I went "well actually she's a real nice doctor and I really like her and I don't know what that was all about", so I don't know if she didn't like or I just caught her on a bad day or if it was a difficult subject for her or I don't know"* [C:Elena, I2, Pauline, L:100].

In this quote it is evident that Pauline felt that this discussion and the manner in which Dr Price conducted it was out of character for her and therefore excused this behaviour, so much so that she defended her to the friend that had accompanied her to the appointment. The occasion which Pauline was talking about had taken place prior to their recruitment into the study, and throughout the duration of the study, interview and observational data suggested that despite the tense meeting that Pauline had described, they had maintained a good quality relationship, which involved mutual trust. A similar sentiment was also expressed by Kate regarding Dr Patel and Dr Price in relation to the discussions around Michael's need for a gastrostomy.

The provision of excuses by the family may be related to their understanding that the professional has their child's best interests at heart, and potentially their knowledge that despite their resistance to the treatment, it may actually be best for their child in the long term. The awareness that such discussions had come from a good place appeared to make it difficult for families to harbour any negative thoughts about the professional, despite the tense discussions that had occurred. Additionally, the importance of having good relationships with their professionals, as outlined in Chapter 5, may have meant that families were determined to preserve the quality of these relationships as far as possible. This may explain why they were prepared to provide excuses for behaviour that they felt was not in keeping with their previous experience with their trusted professional.

For Kay, although she did not make any comments to excuse Alice's role in getting her to sign the contract to state that she would not feed Grace orally, she was observed throughout the study to have a good relationship with her, something which was further demonstrated through her nomination of Alice to take part in the study. As previously discussed (see Section 5.3.2), Glenn had only recently been allocated to Grace's care and therefore did not have the previous relationship with Kay that Alice did. It appeared that Kay may have attributed all the blame onto Glenn as a mechanism of containment, which enabled her to maintain a trusting relationship with Alice. This again highlights the need for strong and trusting relationships, as they can seemingly withstand incredibly tense and difficult discussions.

7.4. Conclusion

For decisions where disagreement existed between parents and professionals, at times the concept of best interests was invoked. Families and professionals were shown to conceptualise best interests' differently, with families considering a more holistic view and professionals focusing on physical health as the main determinant. There was a sense that assessing best interests should be undertaken jointly, but with an awareness by professionals that in situations where they perceived the child was being placed at risk of harm, their assessment was privileged. In situations where there was discussion of the possibility of escalating to the courts, families undertook moral work in maintaining trust in the professionals. Justification enabled professionals to demonstrate their actions as morally acceptable, even when employing antagonistic strategies, through reference to their professional codes of conduct and human desire to do what they believed was best for the child. Through justification, professionals were able to maintain their own sense of moral responsibility and display this to others.

Chapter 8. Discussion

8.1. Introduction

This chapter presents a discussion of the findings of this study in relation to previous research and the theories relevant to the topic. It considers the novel insights which this research has provided and the contribution which it makes to the wider body of literature. A reflexive account of the process of conducting the research and the challenges faced in doing so is presented. The strengths and limitations of the current research will then be provided, followed by a discussion of the implications for practice and further research.

8.2. Overview of findings

Previous studies of decision making have been largely cross-sectional in design (Erby et al., 2006, Nicholson, 2012, Rapoport et al., 2013, Carroll et al., 2012, Popejoy, 2015b, Zaal-Schuller et al., 2016b), however the longitudinal element of this research has facilitated a nuanced investigation of decision making, illustrating its complex and often protracted nature. Through elicitation of multiple stakeholder accounts, the intricacy of integrating often disparate views in the practice of decision making was acknowledged. The findings highlighted that individuals' considered that they had made a decision, even when this was not implemented and this insight was facilitated by the longitudinal feature of the study. Had the study captured data retrospectively, it is possible that decisions would only have been discussed with reference to the decision implemented, rather than any choices which had previously been considered and subsequently overturned. The serial interviews enabled the development of a rapport between myself and the participants, facilitating the disclosure of sensitive and private issues which impacted on individuals' decision making capacity and beliefs.

This study aimed to understand the process of decision making and future planning for children with life-limiting conditions by exploring:

- The experiences and preferences of families, professionals and 'significant others' for engaging in decision making and future planning.

- The communication between families, professionals and 'significant others' about decision making and future planning.

- The networks of care that exist for children with life-limiting conditions and how decisions are negotiated and distributed through these networks.

The overarching aim of this study was achieved as a result of the research design, facilitating novel insights into the process and nature of decision making for children with life-limiting conditions. Chapter 4 provided an overview of the cases, depicting the complex and varied nature of the networks of care, and included timelines which acted to illuminate the dynamic nature of decision making. Chapter 5 provided a consideration of the role of communication and relationships in the process of decision making. This was followed by a presentation, in Chapters 6 and 7, of the strategies involved in containing disagreement, working towards decisional agreement within the network of care and the significant feature of moral work within these strategies.

8.3. The nature of decision making

Findings from this study have shown that decision making is often a complex process, involving numerous stakeholders and evolving over time. This is in contrast to the concept analysis of client decision making, in which Noone (2002) identified the essential attributes to be: *"an intentional choice between two or more discrete options, based on a recognition of a stimulus for action, commits a person to a course of action and expects to accomplish a specific goal or goals"* (p.27). This conceptualisation portrays decision making as simplistic, deductive and unequivocal, resulting in a clear outcome. The focus of Noone's (2002) concept analysis however, was client decision making, in which an individual with capacity weighs up the available options and choses an option which best fits with their goals and values. The concept analyses regarding clinical decision making also discuss the process of decision making as an individual pursuit (Simmons, 2010, Johansen and O'Brien, 2016), which is in stark contrast to the findings of the present study where the process was identified as highly relational. The applicability of these concept analyses in paediatrics, where multiple individuals are involved in the process and often

multiple proxy decision makers exist, is therefore limited and may explain the difference in findings between the present study and the concept analyses outlined.

The concept analysis of SDM in paediatrics accounts for the multiple proxy decision makers involved, (Park and Cho, 2017), although it presents these processes as having a clear consequence, that is, an enacted decision. This decisional consequence was also found in the analyses of clinical decision making (Simmons, 2010, Johansen and O'Brien, 2016). Starting a new medication, consenting to surgery and insertion of an NGT were examples of enacted decisions in the present study. However, the current study also suggested that some decisions occur through an ongoing process, meaning that there was not always an immediate clear decisional outcome. For example, decisions around gastrostomy insertion followed a process over time, where the decision was delayed, demonstrating the lack of a clear decisional outcome, given that the decision topic was often still 'live' and ambiguity existed about the outcome. The difference in the findings from the present study and these concept analyses may have resulted from the different decisions being made. Clinical decision making often involves time critical decisions in response to patient deterioration and therefore a clear decisional outcome is required. Within the context of decision making for children with life-limiting conditions however, the decisions were frequently not time critical and therefore a clear outcome was not immediately required.

Simmons (2010) and Johansen and O'Brien (2016) also allude to the cyclical and recursive nature of decision making. This ongoing and processual nature of decision making was also highlighted by Lipstein and Britto (2015) who found that for children with chronic illnesses the boundaries of decision making were often blurred, such that the end of one decision leads into the beginning of the next, *"with no clear beginning or end"* (p.7). This was similarly identified in the present study and was particularly noticeable for decisions around tube feeding. Lipstein and Britto (2015) also found that decision making in all of their cases occurred over time. Whilst the present study has similarly identified that decision making often occurs over time, it has also elucidated that in some

situations, decisions are made as one off events, such as initiating a new medication or taking the child to the emergency department.

The concept analysis of SDM in paediatrics provides an understanding of decision making in its natural context, whereby it involves participation of parent, child and professional, collaborative partnership, compromise and common goals (Park and Cho, 2017). Such participation, collaboration and compromise was noted in the present study. However, the concept analysis appeared to portray the participants involved in decision making as one professional, the child and their parents, whereas the current study highlighted that in many cases, multiple professionals may be involved in making decisions and discussing these with the family. The literature which the SDM concept analysis (Park and Cho, 2017) was derived from focussed on simple chronic conditions, such as asthma or ADHD, or oncology, where often only one team is involved. Few of the included papers discussed SDM in the context of vastly complex or life-limiting conditions and therefore the wider context within which decision making for such children may occur, may have been overlooked. Discussion of the concept of decision making was considered important in order to understand the process being studied. These findings however, have identified that the current concept analyses regarding health care decision making available, fail to adequately capture the complexity of the process due to their focus on the individual, as with clinical decision making, or lack of attention to the wider literature regarding children with more complex life-limiting conditions.

8.3.1. Types of decisions

It has been recognised that decision making occurs longitudinally through a relational and distributed process (Rapley, 2008). Although it often occurs over time as this study identified, some decisions were ‘immediately enacted’, in which they did occur as a one off event and were implemented immediately. Such decisions were predominantly time critical, such as attendance at the Emergency Department, or considered as routine, such as how much physiotherapy to do, whether to commence a course of oral antibiotics, or altering the flow of oxygen. Rapley (2008) has previously recognised the immediate nature of some critical decisions in acute care, which is in keeping

with the current findings. However, the immediately enacted nature of routine decisions has not previously been reported and may be due to the foci of previous investigations, such that routine decisions were overlooked or not considered worthy of reporting.

The other type of decision identified through the present research was 'future planning'. This type of decision encompassed three sub-types: formal future planning, immediate decisions not immediately enacted and internally declared decisions. 'Future planning' alluded to the unfolding trajectory of decision making which, as Rapley (2008) stated, occurs through a series of encounters, with both medical and non-medical professionals. Formal future planning occurred through multiple encounters with numerous significant others over time, which has previously been recognised by Rapley (2008).

The immediate decisions not immediately enacted sub-type, appeared initially to be an immediate decision rather than a future plan, but given the critical nature and often the greater perceived significance of these decisions, on analysis they were observed to be different. Decisions such as consenting to elective surgery were at times made immediately, but had the potential to be reversed prior to a date for the surgery being arranged. Additionally, although the decision often appeared to be made immediately, on discussion with the families, it was evident that some aspect of the decision had been under consideration within the family previously and that therefore these decisions had been made through a previously unfolding trajectory.

The final sub-type of future plan was the internally declared decision, where families had mentally decided upon a treatment, but for some reason this treatment was no longer required. The making of these internal decisions was facilitated through previous encounters in which the need for the decision was discussed, and through which information about the treatment was gained. Such findings demonstrating a perception of having made a decision which was then overturned has, to my knowledge, not been identified within the literature. The findings from this study suggest that even when decisions are not implemented, participants consider the process of arriving at that choice, as decision making. The cross-sectional nature of the majority of decision making studies may be the

reason why this perception of decision making has previously been absent from the literature.

Although some immediately enacted decisions occurred in the present study, 'future planning' occurred much more frequently and therefore a detailed consideration of the process of 'future planning' will be presented.

8.3.2. Future planning

Findings from the present study revealed that the term 'future planning' was not widely recognised by the non-professional participants and when asked about future planning, many said that they had not engaged in this. This is in keeping with previous research which suggests that families are unfamiliar with the concept of future planning (Wharton et al., 1996, Liberman et al., 2014, Dellon et al., 2010, Erby et al., 2006), although such research has been specific to end of life care plans, rather than future care planning more generally. The findings from the wider literature which suggest that families are unfamiliar with future planning may therefore be due to the barriers experienced by professionals to engaging families in end of life care planning (Lotz et al., 2014, Durall et al., 2012). Within the current study however, despite families indicating that they had not engaged in future planning, many provided examples where they had considered future care, such as resuscitation plans, possible surgery or other future treatment options. This data suggests that they conceptualised future plans simply as decisions, or at the very least articulated them in such a way. When families described decisions that they had made (which included plans for the future), they conveyed similar levels of emotional and cognitive effort for both. This suggests that they experienced little practical difference between decision making and future planning and may signal to why they failed to articulate a difference between these two concepts.

It could be argued that it is misleading to combine decision making and future planning, as the latter does not necessarily result in an outcome, a consequence of the concept of decision making as outlined by Noone (2002). However, decision intentions have previously been acknowledged as important to understand in relation to the process of decision making (Ranyard, 2014). In the present study future plans appeared to be an important part of the process of

making and enacting decisions, which was a core goal of this research. Discussing these terms as though they represented different concepts, when this was not articulated within the participant accounts, would not be in keeping with the underpinning constructionist philosophy of this research. The realities expressed by the participants demonstrated that, for them, decision making and future planning represent part of the same process. Although others may disagree that future planning is part of decision making, the purpose of this research was to explore individuals' experiences in making decisions for children with life-limiting conditions. It is therefore their accounts of the process and concepts involved which are presented.

It appeared important for all participants in the current study to have the ability to change and revise any future plan that they had made, thereby declaring their intention for future action, but with the ability for this to be altered at a later date. Previous literature has similarly alluded to the need for future plans to be changeable. Such literature often uses the term advance care planning (ACP) and primarily focuses on end of life care decisions. Beecham et al. (2016) investigated ACP, with reference to all aspects of the child's future care, identifying that *"for parents ACP is not just about withholding treatment or resuscitation status, but rather an on-going conversation to create and revise a tailored, flexible plan considering daily life and end of life, to optimize the length and quality of life of the child"* (p.8). In concordance with the findings by Beecham et al. (2016), the results of the current study suggest that it is important for formal future plans to be changeable and overturned at a later date, given different circumstances. Other research investigating end of life care planning has similarly highlighted parents' need to know that any plan they make can be revoked if they change their mind (Liberman et al., 2014, Lotz et al., 2017). The current study suggests that this need to change their mind about future plans applies to a broader range of decisions than solely end of life care or resuscitation decisions.

Professionals in the present study referred to future planning more explicitly and appeared aware of the term, something which may be linked to the increased policy drive towards ACP (National Institute for Health and Care Excellence, 2016). Although ACP as outlined by the National Institute for Health and Care

Excellence (2016) refers to care planning more generally, within the literature, traditionally research investigating future planning for children with disabilities relates to end of life care planning.

This study showed that rather than just end of life care planning, any decisions which were deemed likely by professionals to be necessary in the future, served as the basis of future plans and discussions were often initiated with families early. Such early discussions, particularly for tube feeding decisions, often received parental resistance initially and this early introduction appeared to be a strategy employed by professionals to promote families' familiarity with the need for the decision in the future, as has been previously recognised (Edwards et al., 2017, Mitchell and Dale, 2015). Early introduction of future decisions however, may not always be favourable to families (Erby et al., 2006, Nicholson, 2012, Mitchell and Dale, 2015, Delany et al., 2017) and the present study demonstrated divided perspectives on the matter. Some families wished to be prepared for future decision-making, for example, an awareness of organ transplants, however in relation to gastrostomy discussions, most families felt pressured as a result of continued discussions. Despite ongoing conversations acting as the basis for future plans, each time a discussion was had, this served as a decision point. For example, even if a parent knew that the decision would be needed in the future, not agreeing to the treatment yet was seen to be a decision, that is, they made a decision to delay agreement to the treatment.

Within the present study the accounts of non-professional participants did not make any distinction between decision making and future planning, suggesting that they considered future planning to be equivalent to decision making. Given the intertwined nature of future planning and decision making, in the interest of brevity any discussion of decision making within the rest of this thesis will, unless specified, be referring to decision making and future planning.

8.4. Conceptual framework of decision making

This research has provided an understanding of the complex and nuanced nature of decision making. It has uncovered future planning as an integral aspect of decision making from the perspectives of those involved and clarified the impact which time has on decision making in different circumstances. The variety of

relevant relationships, both within the family and professional contexts, which impacted on decision making and the essential role of trust and communication within these was identified. The data illuminated the strategies used in decision making which facilitated collaboration and SDM, and enabled an understanding of the factors specific to the individual or decision which influence these strategies. Decision making and future planning occurred collaboratively most of the time. However, the data highlighted the contextual boundaries to such collaboration, with particular reference to the legal context. Such contextual factors were identified as posing possible tensions to SDM.

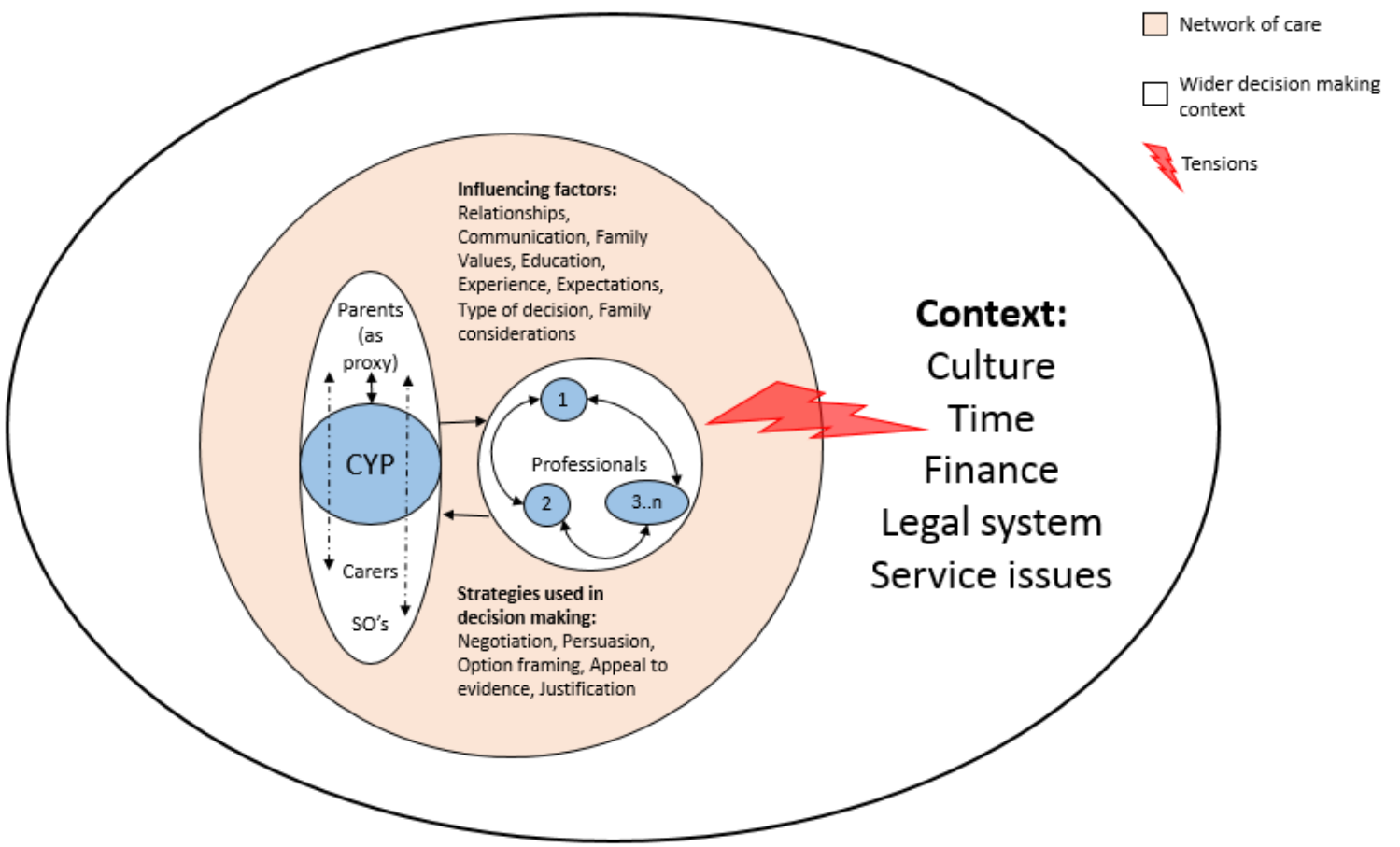
Although some models have illuminated the relational and temporal nature of decision making (Nicholson, 2012, Hinds and Kelly, 2010, Noseworthy et al., 2013), the longitudinal case study design of the present study has revealed the complexity that these factors pose within this process. Furthermore, it has enabled an understanding of the limits to SDM given aspects of the wider societal context.

A conceptual framework of decision making has been developed from the study and is presented in Figure 9. It aims to depict the network of care within which decisions are made and the factors influencing, and the strategies used in, the process of decision making. It also illustrates the tensions which can occur between the individual and relational aspects of decision making, and the wider context in which it takes place. Given the factors influencing decision making for the family, this framework problematizes the singular nature of professional 'best interests' discussions which, in the rare circumstances in which they occur, the child is seen as the sole consideration. It also accounts for the wider considerations in decision making, including the implications for each stakeholder who may be impacted by the decision, in relation to their culture or any financial considerations.

The conceptual framework presented is a two dimensional representation of a vastly complex process, which evolves over time and through the interaction of various stakeholders, and the resulting diagram is necessarily over-simplified. It does however, intend to represent the challenges that have thus far been overlooked in the literature pertaining to decision making for children with life-

limiting conditions, particularly the potential role that the context may play in constraining the process of decision making and the tensions that this may create. The remainder of this chapter will discuss the aspects of this framework in relation to the wider literature.

Figure 9: Conceptual framework of decision making



8.5. The network of care: Relationships and communication

The nature of decision making for children with life-limiting conditions is highly relational, due to the numerous stakeholders involved. As illustrated in Chapter 4 at least one parent and at least two professionals were involved in every case, although most cases involved significantly more stakeholders. As described in Chapter 5, most relationships between families and professionals were described as good and involving mutual trust. A consideration of the role of these relationships in relation to decision making is essential, as they provided the relational context within which the decisions were made.

8.5.1. Trust

Trust was a key feature in good family-professional relationships. Findings from the present study indicated that this developed over time as a result of previous experience and the outcomes of decisions made. This is largely commensurate with the existing literature which illuminated the role of trusting relationships in facilitating decision making and conversely how relationships which lack trust pose a barrier to the process (Nicholson, 2012, Welsh et al., 2014, Ling et al., 2016). In certain cases it appeared that this trust was continually re-evaluated and ongoing interactions served to consolidate or degrade this trust. This was particularly evident in one case where the family started any new relationship with professionals from a stance of trust in them, yet this trust was not invulnerable to damage as a result of experience. Dissolution of trust through experience has also previously been acknowledged (Wood et al., 2010, Konrad, 2008, Todd and Jones, 2003, Iversen et al., 2013) and may lead to significant conflict in which collaborative decision making is inhibited (Forbat et al., 2015). Such conflict was not observed in the present study and signalled to the importance of the work which was being undertaken by all individuals to maintain good working relationships. In situations where trust was damaged, strategies such as calling upon others' for assistance in decision making or requests for second opinions were made, in order to 'contain' any damage to essential relationships. The absence of significant conflict in the present study illustrated the effectiveness of the strategies of 'containment' in maintaining good working relationships, either out of desire or necessity.

8.5.2. 'Containment'

In the current study, although somewhat antagonistic strategies were occasionally utilised in decision making and tense conversations had, episodes of significant conflict were not observed. The antagonistic strategies used however, at times appeared to impact on the relationships which they occurred within, but rather than damage them irreparably, strategies of 'containment' were used by families to ensure that good working relationships were maintained. 'Containment' has previously been described by Davies et al. (2017) as a mechanism through which parents of children with cancer maintained relationships with professionals in the presence of conflict.

Many of the narratives within the current research in which 'containment' appeared to occur were within pre-established trusting relationships and most frequently occurred when professionals challenged the family's decision and referenced the possible limits of their decisional responsibility. Families 'contained' such events through reference to a belief that the professional was only doing what they believed was right. 'Containment' also occurred in one case in relation to the professional's skills. The family had previously had a bad experience with their Orthopaedic Surgeon in administering Botox injections and 'contained' this experience solely to his skill at administering this treatment, rather than to his overall expertise in his field (p.168, Chapter 5.4.2). The importance of trusting relationships in facilitating difficult discussions, as previously identified (Erby et al., 2006, Delany et al., 2017), alongside their role in making specialist referrals appeared to be the reason why families felt the need to 'contain' any challenging encounters and maintain trusting relationships.

Although many of the accounts of 'containment' were presented by the family as occurring in the context of ongoing trusting relationships, some occurred within new relationships. When this occurred it appeared that 'containment' was not a strategy used out of desire to maintain a previously good relationship, rather a necessity for the child to receive care locally, or access specialist care. These rare narratives presented by families, described a perception of uncaring professionals which the family would have to work with for the sake of their child, and they therefore 'contained' the difficult start to their relationship. It is possible that such narratives, rather than presenting an historical picture of

events, aimed to promote the moral character of the family, by contrasting their behaviour with that of the professional. This type of narrative of the self has previously been recognised in the accounts of mothers of children with intellectual disability, where they positioned themselves as good, against a backdrop of bad professionals (Todd and Jones, 2003). Viewing these narratives as polarized accounts, rather than events in which professionals behaved unacceptably, may help to explain how families were able to 'contain' their experiences and maintain reasonable working relationships with professionals.

Containment appeared to be a strategy, used successfully by families, which prevented conflicts from escalating to the point at which trust between the family and the whole service could be damaged, such as is seen when conflict is unresolved (Forbat et al., 2015). This work of containment appeared to be a way in which parents maintained relationships which they perceived were necessary for their child to receive the best care and was therefore part of the moral work that they engaged in. 'Containment' however, was not recognised to be a strategy used by professionals. Although 'containment' was not described in relation to poor inter-professional communication, this issue was described in terms of frustration for families and disappointment when it occurred in relation to professionals whom they trusted.

8.5.3. Inter-professional communication

On the whole, decision making communication within family-professional relationships was reported and observed to be caring and effective. However, the present study also highlighted effective communication in inter-professional relationships as highly relevant to decision making, although such communication was frequently reported to be suboptimal. The present study concurs with the results from previous research (Odeniyi et al., 2017, Stille et al., 2003b, Harlan et al., 2009, Ranade-Kharkar et al., 2017) and found that professionals experienced deficiencies in communication with their colleagues in different teams, particularly between the hospital and community.

Many families in the present study expressed some concern regarding the risk to decision making for their child when poor inter-professional communication occurred. Previous research involving families and professionals has reported

similar concerns (Stille et al., 2003b, Cady and Belew, 2017, Dewan, 2017) and research in the adult field has recognised the role of inter-professional communication failure in significant medical errors (Moore et al., 2003, Arora et al., 2010) or uncertainty during decision making (Arora et al., 2005). Although within the present study significant medical errors resulting from poor inter-professional communication were not identified, at least two cases were plagued by uncertainty in decision making which was articulated to have resulted from poor inter-professional communication. Despite most cases not having experienced errors or difficulty in decision making as a result of poor inter-professional communication, there was a real awareness among other families of the potential for such consequences.

In order to overcome these problems with inter-professional communication, the present study found that parents often took on communication and co-ordination roles, and perceived that they represented the primary mechanism of communication between different professionals involved in their child's care. This was sometimes reported to negatively impact on their experiences of decision making, but the extent to which this role of primary communicator was considered problematic was individual to each family. This need for families to act as the primary mechanism of communication has also previously been reported, as has the burden that this places on them (Kirk and Glendinning, 2002, Gulmans et al., 2009, Golden and Nageswaran, 2012, Stille et al., 2007, Dewan, 2017). Although professionals' accounts detailed the problems with communication with their colleagues, discussion of how this was overcome and who assumed responsibility for communication was absent from their accounts.

Ensuring that an environment existed in which trusting relationships and decision making were facilitated, represented significant work undertaken by all stakeholders. Strategies of decision making aimed at reaching agreement between stakeholders occurred within the context of such relationships and environments.

8.6. Strategies used in decision making

Stakeholders utilised a variety of communication strategies in decision making, through which they negotiated towards an agreeable outcome. These strategies

enabled the sharing of each other's views and helped to avoid the need for more adversarial or confrontational approaches. The most passive and common approach was that of option framing.

8.6.1. Option framing

Option framing as discussed herein, refers to whether choices were framed in relation to their potential benefits or risks, whether the option was presented as one of many possibilities, or whether a strong recommendation, indicating the preference of the professional, was given.

In the present study when treatment and care options were discussed, such conversations were primarily commenced by the professional and their preference was often clear in the way in which these were communicated. Research with professionals caring for patients with Muscular Dystrophy has shown that their own beliefs and opinions influence the way in which they communicate the option of NIV with their patients, with some providing recommendations and others being paternalistic (Gibson, 2001, Edwards et al., 2017, Kinali et al., 2006). This finding was recognised in the present study and professionals were observed to make strong recommendations, particularly when one option was deemed by professionals to be far superior to any other. A recent study has suggested that decision making communication and outcomes in the PICU may be influenced by the professional's gender and parental status (Bateman et al., 2017), suggesting that information may be framed differently depending on individual characteristics of the professionals involved. Likewise variability in doctors end of life decision making in PICU has been reported (Wilkinson and Truog, 2013, Richards et al., 2017), which suggests that individual factors impact on their decision making and the extent to which they are directive or neutral in their presentation of options to the family (Richards et al., 2017).

Information framing may be valued by families as previous research has suggested that when information regarding end of life decisions was presented in a neutral manner, parents felt as though a positive outcome was unlikely (Allen, 2014). In vignette studies of neonatal resuscitation decision making, framing choice in relation to percentage chance of survival, or chance of death

or disability, has been shown to be significantly associated with the choice made (Marteau, 1989, Haward et al., 2008). Similarly framing the choice in relation to the hospital's default option has also been shown to influence the decision made (Haward et al., 2012). In the present study it was not possible to identify the extent to which families' choices were influenced as a result of the way options were communicated.

Nicholson (2012) also alluded to the presence of directive communication in decision making and that perceived choice is influenced by whether professionals *"present a recommendation for technological support as a choice between two options, or a decision that parents must agree to for their child"* (p.170). The extent to which parents in the present study perceived there to be choice available to them appeared similarly related to how information was communicated to them and was predominantly seen in relation to clinical test results indicating the need for action. However some contrasting cases existed where parents did not accept the professionals' representation of available choice and sought out their own solutions; these primarily related to equipment provision or educational decisions. Such rejection of the option which had been presented as the only one available, appeared to be related to parents' feelings of expertise regarding that aspect of their child's care.

Encounters in which options are communicated contain a significant amount of moral work. Professionals are required to identify how much choice they wish to convey to the family and simultaneously the family assess whether they believe the choice conveyed is accurate and represents their child's best interests. Within the present study, in cases where professionals deemed that there was one clearly superior choice, they even articulated feelings of moral imperative to communicate choice in such a way to facilitate parental agreement. It has been argued within bioethics, that medical professionals have an obligation to try to persuade patients to accept their advice (Barilan and Weintraub, 2001, Maclean, 2006, Swindell et al., 2010, Smith and Pettegrew, 1986) and this obligation appeared to be keenly felt by many professionals in the present study.

8.6.2. Persuasion

Persuasion is a strategy used by stakeholders which aims to influence the other individuals involved and work towards agreement with their preferred option. Within the present study it was observed to be a strategy employed by various individuals, often simultaneously.

Professional to parent persuasion

Strategies of overt professional to parent persuasion were observed to occur frequently. My own clinical experience meant that I was familiar with this strategy and considered it a routine, natural, and possibly even necessary aspect of the professional role in relation to decision making. Within the existing literature, persuasion has been reported in relation to decision making in the NICU and PICU (Verhagen et al., 2009, Power, 2012), oncology and general paediatrics (Linnard-Palmer and Kools, 2005), and gastrostomy insertion (Thorne et al., 1997, Craig, 2004). In these studies, professional persuasion was described as an ongoing process in which professionals aimed to influence families to agree with their recommendations, or at the very least not to disagree with them. Such persuasion was recognised by parents in the present study and in keeping with the previous literature (Brotherton and Abbott, 2012), some families perceived this to be coercive. In the present study persuasion was highlighted as a key feature of the decision making process, occurring over many years for some treatment decisions where protracted disagreements occurred.

Delaying decisions whilst persuasion occurred has been identified in the NICU and PICU literature (Verhagen et al., 2009, Power, 2012), albeit in this context decisions were delayed for a number of days, rather than protracted over months or years, as observed in the current study. Delay in decision making has also been identified by Land et al. (2017) who reviewed the conversation and discourse analytic literature and found that when resistance to a recommended treatment was presented, decisions were deferred to a later date. In the present study, enabling decisions to be delayed appeared to be in keeping with parents' need for more time to adjust to changes in the child's care and recognise that the decision was unavoidable, particularly in relation to gastrostomy insertion. This has been previously described by Morrow et al. (2008) and demonstrates that decision making for children with life-limiting conditions may frequently be a

protracted one, with ongoing discussions involving persuasion, prior to an agreement being reached and the decision enacted. This was particularly the case for decisions which were not time critical but which were considered significant by the family.

In one instance in the current study, a professional was observed to utilise another professional who had a good relationship with the family to persuade them of their recommendation (see p.125), something which has previously been found in relation to NICU decision making (Verhagen et al., 2009). Although only one such instance had been discussed in the present study, it signalled to professionals' awareness of the importance of relationships in decision making. Such examples may not have occurred more frequently in the current study due to the good relationships which most of the professionals had with the families, meaning that enlisting the assistance of others for persuasion was deemed unnecessary.

Parent to professional persuasion

Persuasion in the direction of parent to professional was also observed in the present study. Although some aspects of parent to professional persuasion have been previously acknowledged in the literature, this has primarily been termed fighting and has been recognised both in relation to health (Nicholson, 2012, Lindblad et al., 2005) and education (Ryan and Cole, 2009, Duncan, 2003, Hess et al., 2006). Although fighting was a term used by five families in the present study, it was used infrequently in their narratives and was only referred to when frustration about system constraints was being expressed. Persuasion as a more collaborative strategy however, was much more frequently employed. Parent to professional persuasion in the present study occurred at the micro-level of individual parent-professional relationships, whereas fighting, as defined in the literature (Ryan and Cole, 2009, Duncan, 2003, Hess et al., 2006, Nicholson, 2012, Lindblad et al., 2005), appeared to occur at a much more macro-level where the fight was against the system as a whole.

Fighting has been described as a much more energy intensive and antagonistic strategy than that of persuasion which was identified within the present study. Fighting has been noted to result in emotional and financial resource depletion

(Ryan and Cole, 2009, Duncan, 2003), such emotional resource depletion was only observed in one encounter in one case in the present study. The intensive energy investment required for parents to fight may result in them only taking this approach if absolutely necessary and maintaining good working relationships with professionals by using less antagonistic strategies, wherever possible. The predominance of good relationships between families and their main Consultant, appeared to ensure that conflict did not escalate to the extent to which fighting was necessary and therefore it did not feature heavily in this research.

The role of parent to professional persuasion appears to have been absent from previous research. The difference in terminology used in the literature, that is, that professionals persuade and parents fight, may signal to the decisional responsibility parents feel for their children. In the infrequent instances where parents discussed fighting in the present study, they were describing a moral battle against the system and the term fighting may have therefore come across strongly in previous research when parents articulated their perceived responsibilities. Professionals conversely articulated their responsibility in terms of advising parents and imparting their medical knowledge to them, such that the collaborative strategy of persuasion was identified. The different terminology may also suggest a power imbalance in the parent-professional relationship, where professionals are only required to persuade, but parents are required to undertake a more active approach of fighting.

Mutual persuasion

Persuasion was frequently observed to have occurred in both directions (from parent to professional, and from professional to parent) simultaneously. This has previously been recognised by Thorne et al. (1997) in relation to gastrostomy insertion, where professionals tried to persuade parents of the need for a gastrostomy and parents tried to simultaneously persuade professionals that the treatment was unnecessary. Smith and Pettegrew (1986) argue that *"mutual persuasion"* is a necessary process in medical decision making:

> *"involving both physician and patient in both the roles of persuader and persuade. Each is attempting to generate messages which will*

> *influence the choice-making behaviour of the other. Neither is passive or controlled; neither is immune to effective arguments and good reasons. Decision-making is shared via the process of mutual persuasion"* (p.140).

This mutual persuasion, in which arguments for and against the available option are shared, was observed to take place in many occasions in the present study. It appeared to be related to feelings of moral duty and a shared commitment to trying to do the right thing for the child. Reference to a professional moral duty regarding persuasion has been made in the bioethics literature (Barilan and Weintraub, 2001, Maclean, 2006, Swindell et al., 2010, Smith and Pettegrew, 1986) and similarly parental feelings of duty are implicit in the literature discussing 'fighting' (Nicholson, 2012, Ryan and Cole, 2009). In the present study, parents occasionally made reference to their awareness of professionals' moral duty in relation to persuasion, however professional accounts did not similarly demonstrate any awareness of parental moral duty.

Despite persuasion being a significant strategy used in the process of decision making, the results of the present study suggested that this was not the factor which led to agreement. Guerriere et al. (2003) similarly found that parents experienced persuasion (which they perceived as pressure) in relation to gastrostomy insertion and argued that there was no indication that this assisted with or influenced the parents' decision making. Similarly in the present study, for cases in which gastrostomy insertion was considered, families indicated that persuasion had not impacted on their eventual decision regarding the gastrostomy. This suggests that despite the frequent use of persuasion in this study, it was not perceived to be the strategy which was responsible for achieving agreement.

8.6.3. Appeal to the evidence

Another important strategy in decision making, which was often an integral part of attempts at persuasion, was making reference to evidence. Appealing to the evidence was used within persuasion, but also as a mechanism of justification, both in terms of justifying a choice to others and at times internally justifying a

choice to oneself. Evidence was also appealed to when re-evaluating a previously made decision.

All stakeholders in the present study utilised evidence during attempts at persuasion. Craig (2004), in her study of gastrostomy feeding, identified that parents used evidence of their child's physical presentation as a means of challenging the results, or disputing the relevance, of clinical investigations. Parents in the current study similarly appealed to evidence that they believed meant that the results of clinical investigations were less valid. In the present study parents primarily referred to the child's physical health or emotional wellbeing as evidence when attempting to persuade professionals. This has previously been reported in the literature (Craig et al., 2003, Craig, 2004, Nicholson, 2012, Morrow et al., 2008). Appeal to this type of evidence appeared to be related to parents' moral work in maintaining 'normality' for their child.

Parents, when attempting to persuade professionals of their opinion, appealed to evidence which they believed demonstrated their child's normality[25], such as feeding well at home, and tried to make decisions which protected this normality as far as possible. A thematic synthesis by Nelson (2002) identified that the quest for normality is a significant goal for mothers of 'other-than-normal' children. Nelson et al. (2012) also found that parents equated making decisions which promoted their child's normality, with being a good parent, and Bluebond-Langner (1996) discussed that families engage in normalisation work, through the routinisation of care tasks. Such routinisation may lead to families perceiving prolonged and challenging feeding practices as 'normal' for their child, thereby interpreting this evidence differently from professionals, as appeared to be the case in the current study. In contrast to this interpretation, mothers in the study by Sleigh (2005) were aware that their child's feeding was not normal, but perceived that feeding via a gastrostomy posed a greater risk to their normalisation practices and therefore resulted in a resistance to the gastrostomy. The moral work that parents undertake in promoting their child's normality was key for families in this study and their accounts suggested that they wished to avoid any options which they perceived to violate their beliefs about normality.

[25] In this context normality either referred to similarity to peers without a disability, or the situation which had become normal for the child and family over their lifetime.

This moral work of normalisation may explain why they appealed to evidence about the child's physical presentation, over evidence of investigations. Evidence of clinical investigations may be more likely to highlight the child's physical problems and therefore pose a challenge to the parents' work in maintaining normality.

Additionally, parents undertook significant moral work in making decisions about the child's best interests and often cited quality of life and emotional wellbeing, as more relevant than physical health alone. In the study by Nicholson (2012), she described a similar finding, highlighting overall quality of life as important in non-life-threatening decisions:

> *"For children and young people whose medical symptoms are not life-threatening, quality of life is sometimes the key factor in considering the appropriateness of a gastrostomy or ventilation, and parents describe the need to underpin decisions by their child's current quality of life at the time of making a decision"* (p.176).

In contrast to families, professionals however did not refer to emotional wellbeing aspects of quality of life, or maintaining 'normality' for the child. Rather professionals' accounts focussed on the physical health of the child and particularly at times when they expressed concern regarding harm to the child, reference to wider psychosocial concerns were absent from their narratives. My background as a professional may have meant that professionals constructed their accounts based on a belief that we shared a common duty to improve the physical health of the child and therefore may have presented this as the primary focus of their narrative. The literature suggests that professionals most frequently refer to research evidence, or the results of clinical investigations (De Vos et al., 2015, Lotz et al., 2016, Heath et al., 2016), which can be "*verified by objective 'factual' evidence"* (Heath et al., 2016, p.189). Such findings were supported by the present study, as professionals referred to results of investigations to demonstrate their belief that the physical health of the child provided evidence that a treatment was necessary. These findings in consideration with the existing literature suggest that a mismatch between the types of evidence that is valued by each stakeholder exists. Given their different

priorities, the threshold for evidence convincing each stakeholder of the need for action, will be different.

Despite the use of evidence in attempts at persuasion, differences in how the respective evidence was privileged by professionals and families meant that such attempts at persuasion were portrayed as largely unsuccessful. The present study suggested that the greatest factor in changing an individual's decision was the re-evaluation and recognition of new evidence (in the form of more severe symptoms or deterioration for parents, and additional test results for professionals) which convinced the individual that the decision was now required. Such re-evaluation of the evidence by parents has previously been recognised by Craig et al. (2003) in relation to gastrostomy feeding, suggesting that even when significant resistance has previously been present, re-evaluation can result in a change to their decision. Previous research has also reported that parents assimilate evidence of the child's condition and their physical symptoms into their decision making, and that any changes can result in the recognition of the need for action (Nicholson, 2012, Rapoport et al., 2013, Zaal-Schuller et al., 2016a). Wood et al. (2010) also identified that any event or decision representing a transition from normal to abnormal were significant milestones for families. The current research suggested that such significant milestones require significant evidence for the family to re-evaluate their decision and recognise the necessity of agreeing to such treatments.

Professionals have previously recognised the need of parents to see sufficient evidence of a plan of care for them to agree to it (Birchley et al., 2017). However, professionals in the present study did not appear to demonstrate awareness of this parental need, rather they appeared to believe that persuasion alone would facilitate agreement. The difference in these findings may be due to the differences in focus. Professionals in the life and death situations in the study by Birchley et al. (2017) may be acutely aware of the need for the evidence to demonstrate to parents the 'right' course of action. Such 'right' choices regarding contentious decisions, such as tube feeding, however may be less explicit and therefore absent from the accounts of the professionals in the present study. This study suggested that appeals to the evidence when employing persuasion may not be effective, due to the different types of evidence which the

different stakeholders privileged. This meant that the type of evidence used by the persuader was not considered persuasive by the recipient.

Justification

Evidence was also used as a mechanism for justifying decisions to oneself and to others. This need for justification and the use of evidence for it was similarly highlighted by Craig (2004) in relation to how mothers portray themselves as morally responsible in relation to tube feeding decisions. It has similarly been recognised in paediatric oncology (Kars et al., 2011) during the transition to palliative care. Kars et al. (2011) highlighted how guidance from professionals helped parents to understand the necessity of making treatment decisions and acted as evidence for their justification. In the present study, justification appeared to be a strategy in which individuals could feel content with the decisions they had made and that they were in congruence with the morally responsible identity they wished to assume.

Mahant et al. (2011) and Craig (2004) have previously recognised that lack of professional consensus contributes to parental uncertainty in decision making. Although a rare occurrence, when parents in the present study received conflicting opinions from professionals, they incorporated this lack of consensus into their justification of their decision. Professional justification in the present study solely referred to their professional and moral duty to escalate to the courts in situations where they were concerned about harm to the child.

8.6.4. Reference to best interests

During disagreements, particularly those which were protracted or in which the previously discussed strategies failed to result in agreement, reference to the child's best interests was made by professionals. In the current study whenever the concept of best interests was invoked, parents and professionals each referred to their own specific expert knowledge to justify their assessment of the child's best interests. Consequently they often arrived at different understandings of what constituted the child's best interests, something which has previously been described as a *"fundamental ideological difference"* (Thorne et al., 1997, p.92). Their different perspectives resulted in conflicting opinions, something which Moore and Kordick (2006) identify can result in conflict. Although previous

research has demonstrated the presence of conflict when different perspectives exist for end of life care decisions (Archambault-Grenier et al., 2018, Verhagen et al., 2009), conflict was not experienced in the present study. This may have resulted from the effectiveness of the strategies employed, or alternatively due to the difference in emotional implications of decisions such as tube feeding and NIV, and those around end of life care.

In the present study, tube feeding represented the decision for which disagreement most frequently occurred. This decision has previously been identified as a contentious issue (Thorne et al., 1997, Guerriere et al., 2003, Rouse et al., 2002, Craig, 2004, Morgan et al., 2014) and therefore the fundamental ideological differences (Thorne et al., 1997) between professionals and families became clear. It appeared that parents perceived a moral duty to protect the child's normality and that this drove them to avoid tube feeding. However, the professionals' perceived moral duty to ensure the child maintained optimal physical health, meant that they promoted it. Consequently for this decision when parents and professionals referred to the child's best interests, they came to very different conclusions about what this constituted. Therefore, even when the concept of best interests was invoked, an agreement was not necessarily reached.

In best interests' discussions in the current study, professionals' accounts were prejudiced towards the child's medical needs, with discussion of their wider psychosocial wellbeing being absent. Parents did not often refer to the concept of best interests, but when such discussions were initiated by professionals, they appealed to evidence of the child's wider psychosocial wellbeing to demonstrate how they assessed what would be best for the child. Similar findings in relation to tube feeding (Morgan et al., 2014) and goals of care in paediatric oncology have been reported (de Vries et al., 2013). Whilst initially agreeing on what was in the child's best interests, parents and professionals in the study by de Vries et al. (2013) diverged in their views over the course of the child's treatment, with parents becoming more focussed on holistic considerations and professionals remaining focussed on physical concerns. This is in contrast to the findings by Zaal-Schuller et al. (2018) who found that parents and professionals agree on what constitutes the most important aspects of quality of life. However, the latter

paper investigated decisions regarding end of life care and therefore overall quality of life may be more important to professionals than considering the child's physical health as the primary concern, as was found in the current study.

Despite the bioethics literature being vast, empirical research on best interests in decision making is sparse (Birchley et al., 2017), limiting any comparisons between professionals' assessment of best interests and the assessments of families. One study suggests that professionals are committed to incorporating family values and opinions into best interests' assessments (Birchley et al., 2017) and this commitment was similarly expressed by professionals in the present study. However, despite professionals vocalising this commitment, in cases where they voiced concerns about harm to the child, reference to their professional duty (General Medical Council, 2007) to place the child's needs as primary over any family values, was made. The professionals in the present study utilised the concept of harm as a justification for their actions and appeared to conceptualise it as self-evident, rather than the subjective concept which it has been argued to be (Opel et al., 2013, Birchley et al., 2017).

Within the bioethics literature, best interests and the harm threshold are presented as distinct concepts which professionals can draw upon in their decision making. However, the present study demonstrated that the concept of best interests was only invoked when professionals were concerned that the child was at risk of harm, suggesting that in practice these concepts are linked. The bioethics literature has highlighted these concepts as ambiguous (Birchley, 2016b, Rhodes and Holzman, 2014) and indeterminate (Diekema, 2004, Birchley, 2016a, Shah et al., 2017). Despite this, in the current study, professionals' accounts suggested that these terms were accepted as unproblematic, and as though they were self-evident. Within some disagreements where professionals noted concern, these terms were used unsparingly as a way of justifying their moral work in persuasion and occasional use of confrontational approaches.

8.7. The network of care: Shared Decision Making

The strategies used by all stakeholders in decision making demonstrated how numerous stakeholders work together to negotiate and compromise when

making decisions for children with life-limiting conditions. SDM has been identified as an ethical imperative (Coulter and Collins, 2011) and its promotion within national policy (Department of Health, 2010) demonstrates it to be considered as essential for decision making. This study demonstrates that the process of decision making is often collaborative, even in cases where there is disagreement. Despite this collaboration, the way in which decisions are made collaboratively in practice does not reflect the conceptualisation of SDM in the literature.

8.7.1. Presence of SDM

Zone of parental discretion

Professionals in the present study voiced a commitment to SDM and such findings have previously been reported in a systematic review of physician attitudes (Pollard et al., 2015). However, in the present study SDM was only truly facilitated in certain situations where professionals viewed there to be a range of viable options, whereas when professionals perceived that *"there is one clearly superior path…patient preferences play little or no role"* (Barry and Edgman-Levitan 2012, p.780). Professionals in the present study appeared to agree with this, as they actively engaged families in SDM only when they viewed there to be more than one appropriate option, suggesting that collaboration and SDM existed solely in the presence of professional acquiescence.

One professional's account of shared decision making alluded to the zone of parental discretion, an ethical tool which can be used by clinicians to identify when and whether a parents' decision should be followed. It is premised upon the importance of avoiding harm to the child and the moral claim of parents as decision makers for their child (Gillam, 2016). It accounts for *"the ethically protected space where parents may legitimately make decisions for their children, even if the decisions are sub-optimal for those children"* (Gillam, 2016, p.2) and allows parents to make the choice, as long as it is deemed 'good enough'. Within the present study, for some decisions such as commencing a new medication or some emergency care planning discussions, the zone of parental discretion appeared to be acknowledged and families were enabled to participate in a process of SDM. In such situations, although professionals sometimes voiced their preferred option, they allowed families to choose

between a range of acceptable options, so long as they did not pose what they perceived to be a risk of harm to the child.

This highlights the presence of professional control over parental collaboration in their child's care and has previously been recognised (Knox, 2000, Paliadelis et al., 2005). It signals to some degree of power imbalance in the family-professional relationship and raises the question of whether decision making can ever truly be shared with families if professionals have the power to control their involvement. Family involvement appeared to be dependent on the professional's perception of whether more than one viable option existed.

Previous research has suggested that in situations with only one viable option, rather than SDM, a process of shared problem-solving or simple agreement occurs (Smith et al., 2013, Pentz et al., 2012). Within these studies however, it appeared that there was a shared perception between all stakeholders that there was only one viable option, thereby enabling a process of shared problem solving or agreement (Smith et al., 2013, Pentz et al., 2012). Within the present research, for some rare decisions, for example, emergency surgery, this process of shared problem solving was observed. However, in other instances, particularly for tube feeding decisions stakeholders disagreed about whether there was in fact more than one viable option and therefore a process of shared problem solving or agreement could not occur.

When professionals and families disagreed about whether more than one viable option existed, professionals narrated the decisions in terms of allowing families to delay the treatment initially, whilst they employed the strategy of persuasion. Fiks et al. (2011) found that professionals perceive SDM to involve persuasion, which is in contrast to the attributes of the concept espoused in the literature (Park and Cho, 2017) and national policy (NHS England, Undated). The current study concurs with these findings and suggests that professionals believed that they were engaging in a process of SDM even when they employed persuasion as a strategy. In such situations where professionals employed persuasion whilst enabling families to delay treatment, families appeared to perceive this process of decision making as shared, as their decision to refuse or delay the treatment was, at the very least, being considered by professionals. The present study

suggests that both professionals and families often perceived decision making to be shared, even when the situation did not demonstrate the attributes outlined in the concept analysis by Park and Cho (2017). These findings suggest that a reconsideration of the attributes of SDM to encompass persuasion may be beneficial and more representative of how the process occurs in practice.

Assertion of the parental role in SDM

Despite the stated professional commitment to SDM, at times families felt the need to take an active approach to ensuring that their views were heard and they were able to share in the process of decision making. Parents discussed how they were most appropriate people to make decisions for their children, referring to their expert knowledge, and has previously been recognised in the literature (Wood et al., 2010, Nicholson, 2012, Smith et al., 2015a, Gibson, 1999). Professional narratives suggested that they valued the parental expertise in decision making, yet when they were concerned about possible risks of harm to the child, the value that they placed on parental expertise appeared reduced. This led to parents occasionally feeling the need to assertively claim their role in decision making and consequently feeling more involved.

When parents claimed their role in decision making, often this was through co-ordination of their child's care or contacting hospitals to obtain results and care plans. Some parents found this approach difficult and therefore employed other strategies, for example, utilising advocates (such as other professionals or family members) or very tactfully negotiating their position. The literature recognises parental advocacy roles (McKeever and Miller, 2004, Ryan and Cole, 2009, Todd and Jones, 2003, Hess et al., 2006, Moore and Beckwitt, 2003, Lindblad et al., 2005, Dewan, 2017, Giambra et al., 2018) and how undertaking such roles enables parents to act as they believe a good parent would (McKeever and Miller, 2004). McKeever and Miller (2004) highlighted that different parents use different approaches, either advocating themselves or, for those who felt relatively powerless in relation to professionals, utilising others to do it on their behalf. Both approaches were evidenced in the current study, albeit rarely. More common was a collaborative approach in which families sought to work with, rather than against professionals. McKeever and Miller (2004) state that fear of retribution for their child resulted in parents following the rules of the healthcare

encounter and modifying their behaviour to get along. Although fear of retribution was not identified by families in the present study, it was clear that they predominantly behaved in a way which enabled collaboration. This may either signal to their perceived lack of capital in the encounter or simply that, as appeared to be the case, good working relationships had been established with their main consultant.

For particular decisions where parents believed that they had a greater level of knowledge and expertise, they felt able to make and implement decisions without reference to professionals. In certain circumstances, families made a decision which was against the advice of professionals; these were regarding home schooling, housing decisions, tube feeding and medications. Previous research has found that parents are prepared to override professional advice in relation to decisions regarding nutrition and diabetes management, when they believed that their relevant knowledge was superior to that of the professionals (Thorne et al., 1997, Kirschbaum and Knafl, 1996). Feelings of expertise regarding the child has also previously been associated with parental confidence in decision making (Wood et al., 2010, Nicholson, 2012, Smith et al., 2015a, Gibson, 1999). Within the present study, families most commonly asserted their opinion in relation to tube feeding decisions, and in keeping with the existing literature, such instances were often accompanied by discussion of their detailed knowledge and expertise of the child.

The role of certainty

In the present study it appeared that the level of decisional involvement of stakeholders was determined by different individuals' evaluation of uncertainty. Families were at times less inclined to accept professional recommendations when they viewed the decisional outcome as uncertain, although often asked for advice in such situations. Austin et al. (2017) found that for deep-brain stimulation decisions which have somewhat uncertain outcomes, parents extensively considered and weighed up the options, but also referred to the professionals' advice. In their study, professionals were also observed to influence families' decisions to accept surgery. This was in contrast to the present study where, when professionals voiced uncertainty about a decision,

they appeared willing to delegate decision making to the family and were less likely to try to influence their decision.

Leonhardt et al. (2011) conducted a study of proxy decision making under conditions of uncertainty using hypothetical vignettes. They demonstrated how choosing an uncertain option in loss framed scenarios, may reduce feelings of responsibility and self-blame. This mechanism appeared to be manifest in the present study in relation to gastrostomy insertion. Parents of children facing gastrostomy insertion, appeared to frame this decision in terms of loss: loss of social bonding and normality. In such situations parents appeared to favour the status quo, the consequences of which were uncertain, rather than the decision to insert the gastrostomy, which was perceived by them as a certain negative outcome. Choosing the uncertain option, of the status quo, may have facilitated parents to feel less responsible for the outcomes and may help to explain this decision making approach.

Elwyn et al. (2000) discussed that uncertainty and equipoise represent two different concepts in relation to decision making and these impact on the extent to which decisions are shared, and that the way in which these are communicated impacts on patient choice. In the present study when equipoise existed, SDM was facilitated, with mutual information sharing and agreement being facilitated. In situations where there was significant uncertainty, professionals were observed to offer the decision over fully to the family, devolving the decisional responsibility to them. Previous research regarding life-sustaining treatment decisions in the neonatal and paediatric ICU has recognised that in situations of significant professional uncertainty, families are more actively engaged in decision making and their views given more weight (Richards et al., 2017, Payot et al., 2007). In such uncertain situations it seemed that professionals felt able, and perhaps compelled, to include the family values and expertise regarding the child in decision making, as was indicated in the present study. van Zuuren and van Manen (2006), highlight in relation to neonatal end-of-life decisions that discussing uncertainty appears to be integrated with the option which has the greater weight of evidence behind it. Most of the research into decision making in situations of uncertainty are about life-sustaining measures and therefore may not help to understand decision making in relation to non-critical decisions.

Nevertheless, it appeared to hold true in the present study that in cases where there was little evidence to guide decision making and substantial uncertainty existed, the uncertainty was incorporated with the family's preference and served as a justification for that choice.

8.7.2. Ultimate responsibility for decision making

Parental sense of ultimate responsibility

The policy drive towards patient autonomy (Department of Health, 2010) is in keeping with the findings of this research, in which parents portrayed themselves as having a moral duty to make decisions on behalf of their child. This feeling of moral duty has previously been recognised in relation to parents' knowledge of and expertise regarding their child (Lindahl and Lindblad, 2013), with parents of children with palliative care needs feeling that making informed decisions is an important attribute of being a good parent (Hinds et al., 2009, Feudtner et al., 2015). The parental narratives regarding their decisional responsibility in the present study were constant and remained stable over time, as reported in a longitudinal study of children with gastroenterological and rheumatological conditions (Lipstein and Britto, 2015). Although the feeling of responsibility appeared to be a constant, the feelings of expertise, and therefore confidence in decision making was alluded to have evolved over time for the majority of the cases in the present study, reflecting the findings of previous research (Wood et al., 2010, Nicholson, 2012, Smith et al., 2015a, Gibson, 1999, Reeves et al., 2006).

Previous research into preferred parental decisional roles has largely been conducted in relation to end-of–life care decisions and preferences ranged from autonomous, to shared, to paternalistic, which was largely dependent on culture. For example, western countries are reported to mainly adopt an autonomous or shared decision making style, and Middle Eastern countries a largely paternalistic style (Pyke-Grimm et al., 1999, Mack et al., 2011, Kilicarslan-Toruner and Akgun-Citak, 2013, Sullivan et al., 2014, Madrigal et al., 2012, Birchley et al., 2017). Carroll et al. (2012) found that parents in their interview study of decision making felt responsible for the decisions regarding their child, but did not delve into how this responsibility was perceived in terms of autonomous or shared decisions. In the present study, it appeared that

prospectively, families narrated a feeling of ultimate decisional responsibility for all decisions. The introduction of the study to families may have inadvertently encouraged them to present a position of assuming ultimate decisional responsibility, as I had asked them if they wished to nominate anyone who helps them make decisions. Families may have inferred that I saw them as solely responsible for decision making, with other people merely 'helping them' and they may have therefore wished to present this stance in their accounts. However, parents' portrayal of ultimate decisional responsibility appeared to be revised retrospectively and the outcome of the decision subsequently determined the extent to which families portrayed themselves as being responsible for the decision (see p.269).

Relational autonomy

Although parents presented accounts in which they were the ultimate decision makers, their narratives also suggested that when they felt they lacked decision specific expertise, or other factors such as stress prevented them from feeling confident in their decision making, they decided to defer to professionals' decisions. Nevertheless, parents were able to maintain the perception that they were autonomous agents and held the view reflected by Maclean (2006) that *"the autonomous nature of the decision is unaffected by reliance on the professional's advice"* (p.328). Baruch (1981) recognised how parents were able to maintain feelings of, and portray, moral adequacy through accepting responsibility for decisions within the 'parental reality', but discharging responsibility for anything which occurred within the 'medical reality'. In doing so they were able to maintain their accounts of moral adequacy (Baruch, 1981). Within the present study, parents were happy to delegate decision making for some aspects of care within the 'medical reality', yet simultaneously maintain their overall feelings of decisional responsibility.

Additionally, as articulated by McKeever and Miller (2004) when parents comply with medical recommendations, they are not absolving themselves of responsibility, but their compliant behaviours act to demonstrate their belief and trust *"in the value of the treatment ''game'' for their children, and were a public and personal measure of belief in…the abilities of professionals to improve their children's conditions or sustain their lives"* (p.1183). This appeared to be how

parents in the present study were able to maintain a belief that they were ultimately responsible for decisions concerning their child, yet simultaneously discharge this responsibility to, or share it with, professionals when they lacked the relevant expertise and knowledge. Ownership for such decisions however, appeared to remain with the parents in the present study.

Role of the child

Within paediatrics, the role of the child is important in decision making and further highlights the relational nature of autonomy, given the developing role of some of the children in managing their care. It was clear through the accounts of the two children interviewed in the present study that they had involvement in the decisions concerning them, but referred to their parents and professionals for guidance. This desire for assistance in decision making for children and young people has previously been recognised (Skyrme, 2016, Derman, 2008, Day et al., 2016) and has taken into account the perceptions of some children and young people with learning difficulties (Mitchell, 2012). Children have been shown to value being involved in decision making, even if they are not the ones who ultimately make the decision (Miller, 2009) and this appeared to be the case in the present study. Similar to parents, research has also shown that children and young people appeared to desire, and achieve, differing levels of decisional responsibility in relation to their expertise and the significance of the decision under consideration (Derman, 2008, Miller, 2009). Given the lack of young people interviewed in this study, these findings from the literature were neither supported nor refuted. It did, however, demonstrate the different ways in which children may be engaged in the process of decision making and consequently the extent to which they claim responsibility for these decisions. The two children interviewed in the present study displayed decision making behaviour at very different ends of the spectrum of engagement, although the reasons for these different behaviours was not identified. Level of involvement in decision making may be related to a desire to accept or avoid decisional responsibility in relation to the possible outcomes of the decision.

Reducing feelings of responsibility for negative outcomes

As highlighted previously, families desired to be perceived as the ultimate decision maker, whether or not they had made the decision themselves or

delegated it to professionals. The exception to this was in situations where families felt that the child had been negatively impacted by a decision. In all but one case (see p.180 in Chapter 6.3.1), they subsequently distanced themselves from such responsibility in order to avoid feelings of guilt and self-blame. However, it was not possible to identify whether attributing the responsibility to professionals was a prospective decision or whether it was made retrospectively once the outcome of the decision was clear.

Direct involvement in decision making has been associated with feelings of self-blame for the negative outcomes of decisions in the NICU (Botti et al., 2009). This was likewise a core finding by Atout et al. (2017) who identified that sharing, or delegating decisional responsibility onto professionals assisted mothers in avoiding any feelings of self-blame. The need to avoid feelings of guilt may help to explain the disparity between parent's accounts of responsibility and their observed role in decision making. Likewise these different accounts of responsibility are in keeping with the constructionist epistemology upon which this research is based, in that individuals presented different accounts of decisional responsibility in different contexts.

Professional perception of ultimate responsibility

Where professionals in the present study were confident that one treatment was superior to another, their sense of professional and moral duty to the child appeared akin to the ultimate decisional responsibility that parents felt. Professionals are held accountable for their actions (Swallow et al., 2016) and feel a duty to act in accordance with their professional bodies' guidance and safeguard the child (General Medical Council, 2007). This professional moral duty was observed to transcend their commitment to embedding the family values or preferences within decision making, and reference to the limits to the parent's authority for decision making was made. This occurred in relation to gastrostomy insertion in three cases, where professionals mentioned the possibility of escalating contested decisions between parents and themselves to the courts. In such cases the court would then be the arbitrators of the child's best interests (Children Act, 1989) and therefore recognised as the legal ultimate decision maker.

Despite parental accounts portraying an overall feeling of ultimate decisional responsibility, interviews with the professionals described the zone of parental discretion (Gillam, 2016) and therefore identified ultimate decisional responsibility as much more complex. This resulted from the different and equally valuable expertise of, and the moral duty felt by, all stakeholders involved, highlighting that the ultimate decision maker for each decision is at least somewhat based upon the certainty of the outcomes of the decision in question. Professionals held significant control over the ultimate decisional responsibility through their use of option framing and reference to the legal system. Such professional control is alluded to in the terminology of the zone of parental discretion (Gillam, 2016). Such decision making frameworks are juxtaposed to the current rhetoric around SDM and patient choice, and highlight the complexity of decision making for children with life-limiting conditions, where the court may act as the final custodian of the child's best interests, thereby subverting the parental decisional responsibility. Empirical research into professional perspectives of ultimate decisional responsibility has not been identified and therefore it is not possible to compare the perceptions of professionals in the current study to those held by the wider professional population.

8.7.3. Relational frameworks for SDM

Previous work has acknowledged the relational nature of autonomy in medical decision making (Epstein and Street, 2011, Carnevale et al., 2017). It has been recognised that decision making in healthcare can be challenging, as individuals may respond emotionally to decisions, and complex technical information is presented to individuals who may not fully understand this information, consequently resulting in cognitive overload (Epstein and Street, 2011). Autonomous decision making is therefore emotionally and cognitively challenging, and not commensurate with the relational and socially embedded nature of human beings. Decision making therefore occurs through reference to others and in a shared process of deliberation, as was observed in the present study. Epstein and Street (2011) propose a 'shared mind' approach to decision making in which processes of cognition and attunement (in which individuals try to understand the others' perspectives) are undertaken collaboratively to

enable shared sense making and arrival at a decision. Likewise Carnevale et al. (2017) recognise the relational nature of decision making, paying particular attention to individuals' moral experience, something which was identified in the present study as a significant feature of decision making. Carnevale et al's. (2017) relational ethics framework recognises the simplistic nature of current understandings of ethics in the care of children with complex healthcare needs. They argue that the concept of best interests as understood in law and healthcare currently, aims to detach the best interests of the child from those with whom they are integrally connected. Parents as surrogate decision makers are therefore expected to supress other ethically relevant concerns, such as the overall best interests of their family in making decisions for the child (Carnevale et al., 2017) and this was seen in the present research. The relational ethics framework promotes an open discussion where all stakeholders can share the concerns which are ethically relevant to the decision and come to a better understanding of these in relation to others' concerns. This approach aims to maximise the benefits for all stakeholders and demonstrate that their views have been considered, resulting in a bridging of perspectives and agreement on a course of action (Carnevale et al., 2017). Such approaches may help to further prevent sustained and intractable disagreements which can inhibit SDM from occurring and assist in promoting trusting relationships, collaboration and agreement.

8.8. Context: The law and shared decision making

Although many contextual issues were identified, data analysis revealed the significance of the legal context on the process of decision making and therefore will be the main focus of this section. The legal system in the UK determines that it is the ultimate responsibility of the court to decide what is in a child's best interests (Children Act, 1989). Culture and time were also related to legal issues in decision making. Professional culture was determined by codes of conduct, which were developed with reference to the law, and time was relevant as professionals used their judgement to decide how long they could allow disagreement to continue, before they discussed possible escalation to the courts.

The role of parents as proxy decision makers in paediatrics makes this context of SDM unique (Wyatt et al., 2015, Park and Cho, 2017). However, the challenges that this poses have not yet been acknowledged. The literature

presents the concept of SDM in paediatrics as largely unproblematic (Park and Cho, 2017) and desirable. Although some research suggests that SDM does not always work in practice, such reports are usually followed by suggestions for how to improve the implementation of SDM, rather than a critique of its relevance to paediatrics in the UK legal context. Birchley (2014) provides a comprehensive analysis of the application of SDM within paediatrics, arguing that it has been adopted without sufficient analysis of how it can function within this specific context, as has been identified within this study.

Medical consensus was not always present in the current study for decisions involving disagreement between professionals and families. This, and the lack of research evidence supporting the effectiveness of certain treatments, such as gastrostomy insertion in children with neuro-disability (Mahant et al., 2011, Sleigh and Brocklehurst, 2004, Novak et al., 2013), may have resulted in a recognition that application for a court order for this decision may not be successful. Additionally professionals in the study by Birchley et al. (2017) had little experience escalating cases to the courts, citing the antagonistic nature of doing so and the feeling that a judge arbitrating on medical decisions is inappropriate, as reasons why they do not pursue that action. Given the possibility of a court order being rejected, utilising an antagonistic strategy such as application to the court, which would impact on ongoing relationships, is likely to be a last resort and this explains the lack of use of this strategy in the current study. In one case in the current study where professionals felt certain that the choice made by the parent was not in the child's best interests, they employed manipulative practices aimed at achieving agreement with their preferred treatment option. Establishing trusting relationships in order to persuade parents to agree to the treatment appeared to be a way in which escalating to the courts could be avoided, although in this case, the real threat of court action was eventually utilised. Additionally, in the two other cases in which professionals referred to the limits of parental decisional authority, they cited their professional duty to prevent the child from experiencing harm (General Medical Council, 2007) and referred to the role of the courts in making such decisions (Children Act, 1989). All decisions, which were 'concluded' by the end of this study, had an outcome which was acceptable to all stakeholders,

despite many having previously been contested. This suggests that agreement is predominantly reached through parental re-evaluation of the evidence, prior to the professional perceiving that the child was at risk of harm, and therefore meaning that actual application to the courts was unnecessary.

In situations where the professional was concerned that the child was likely to suffer harm if disagreement continued, they referred to the child's best interests and the legal limits to parental decisional responsibility. Such situations highlight the constraints which the legal context places on the implementation of SDM, as feelings of professional moral duty and the role of the courts as the final arbitrator of the child's best interests become the primary considerations when there is the possibility of harm. Professional reference to the limits to parental responsibility highlighted the pressure put on families in decision making in these circumstances, such that SDM may not be able to occur. If *"SDM involves reciprocal and unbiased information exchange, rather than merely transferring information from the professional"* (Park and Cho, 2017, p.6), agreement to a treatment based on the threat of court action would not constitute SDM. However, in such situations where risk of harm was cited, SDM would no longer be considered appropriate, as in that situation there would be only one viable option and therefore professionals would consider persuasion and/or escalation to the courts a necessary part of their role. However, in the present study professionals did not actually escalate to the courts, which may indicate that they were not sufficiently concerned about harm to the child to remove the option of SDM, yet may have conveyed this perception to the family and therefore exerted pressure on them to agree.

Birchley (2014) argues that the evidence to suggest that SDM has any benefit for children is lacking, but acknowledges that given the ambiguity over who holds the moral authority in paediatric decision making, SDM *"represents the 'least bad' method of making decisions about children"* (Birchley, 2014, p.219). The present study demonstrated that SDM characterised most decisions, but involved a significant amount of moral work by all stakeholders to determine the extent to which they should assert their role in decision making. It suggested that rather than being the 'least bad' method, in situations where SDM is possible, it is a good method of decision making in paediatrics. Although no such incidents

occurred in the present study, reference to the possibility of escalating to the courts highlighted that in some situations of protracted disagreements, a process of SDM with the family may no longer be possible. This study has shown that SDM in relation to children with life-limiting conditions requires professional and legal acquiescence. As such, it may not always be possible and therefore appears to be an ideological pursuit, albeit one which is largely successful in practice.

8.9. Reflexivity

Although a constructionist underpinning was chosen for the study as a result of my understandings of the world and beliefs about the formation and discovery of knowledge, the reality of the social construction of knowledge and its meaning in practice came as a surprise to me. Numerous occasions throughout the study prompted me to reflect on the socially constructed nature of reality. I experienced many encounters where participants seemed to have revealed something new to themselves during our discussions and these prompted me to reflect on the nature of the research, and whether my presence was in fact more than just as a tool for data collection, but rather had some interventional component. My prior reading provided me with the awareness that *"meaning is made through social interaction and language"* (Holloway and Freshwater, 2007, p.20), however the practical observation of meaning making in real time as participants told me stories, was unexpected. It served as a basis for reflection that I had been an instrumental part of the accounts that were provided and of the narrative which has been presented within this thesis. My role as more than an instrument of data collection was further highlighted by two of the professionals in the study who remarked that participating had been useful for them as a form of clinical supervision, enabling them to critically reflect on their own practice.

Due to my extended presence in the lives of these families, acting as another person to keep them company whilst waiting for appointments, trusting relationships had begun to develop and my role as a nurse, which had been fully disclosed to participants, led to them asking what decisions I thought they should make. I informed them that it was important that I did not provide advice as I was there to understand their experiences, rather than influence them and how they portrayed their experiences to me. Most participants were happy with this

answer but after one interview a parent pushed me and said that she was providing me with information and it was not fair that I withheld my opinion. This situation required a delicate balance, between trying to be an impartial observer and not influencing the data (with the awareness that this is never truly achievable), and maintaining a good relationship with the participant. I provided a vague response to her question, which was sufficient to demonstrate that I was also willing to share my opinion and she then divulged more information to me about her feelings about the decision, which were enlightening in relation to her decision making process. Although it felt freeing to be able to express an opinion, I felt somewhat deceitful for not sharing fully my perspective on the matter. This situation explicitly demonstrated to me further that I was not just an instrument of data collection, but an active collaborator in the production of that data, as it appeared that the participant's disclosure about her decision making following my openness, may not have been otherwise shared.

The observational component of this research enabled the parental account of ultimate decisional responsibility to be compared with the occasional observed reliance on professional recommendation. This highlighted the constructed nature of their accounts in relation to their need to demonstrate agency in their child's care and may also reflect the research approach. Families were recruited and asked to nominate people that helped them make decisions, thus portraying my perception that they were central to, and possibly, ultimately responsible for their child's decisions, with other people merely 'helping' them to do so.

Alongside the development of my practical understanding about the relevance of philosophy to research, this study also challenged many of my personal and professional beliefs. As a researcher with an additional identity as a nurse, I entered the field with a professional perspective, having had no previous experience as a parent, or patient (in such a setting). As a nurse entering the field my view was that it is a professional's role to persuade families to accept care that we deem necessary. However, conducting this study has provided me with space to enter the 'shared mind'(Epstein and Street, 2011) of all the stakeholders involved and appreciate the relational ethics (Carnevale et al., 2017) involved in decision making. This has enabled me to recognise that as professionals we may know the science best, but that it is families that know themselves and their

children best. It has alerted me to the idea that the concept of 'best interests' should not be a solely medical one and that other factors, including the child's psychosocial wellbeing and overall family wellbeing (which is likely to impact heavily on the child's overall wellbeing) may actually be more important in the consideration of what is in the child's best interests. It has also alerted me to the indeterminacy of and value pluralism which the concept of best interests is plagued by.

At the start of the study I had some concerns that my role as a professional may predispose me to prioritise or privilege the professional account. However, there was a huge disparity in terms of time spent with professionals and the time spent with families. Often hours were spent with families whilst waiting for numerous appointments over the study period and when visiting their houses for interviews, some time to make small talk before and after the interview occurred. For professionals however, interviews took place around their busy schedules and therefore much less time was spent with them. This meant that I was able to engage with the families' lives, their accounts and their realities more fully than with the professionals, which was valuable in order to try to create an equality of understanding between the different perspectives. My supervisors were also instrumental in highlighting any instances where I began to privilege a particular viewpoint, thereby ensuring that the data were viewed with equal weighting.

Prior to and during this research, I felt as though I was seeking an answer to who ought to be the ultimate decision maker for the child. However, I have emerged from this investigation with an understanding of the need for a collaborative and shared process of decision making. This study has demonstrated that collaborative decisions are made most of the time and that a significant amount of moral work is undertaken to maintain relationships and ensure SDM. This enables families to maintain a sense of responsibility for decisions which they wish to feel responsible for, and conversely distance themselves from feelings of guilt for decisions with less positive outcomes. Awareness of the relational ethics framework has further consolidated my view that the concept of an ultimate decision maker within the field of paediatric health care may be inappropriate. This transition in opinion has resulted from a greater appreciation of the nuanced complexity of the process of decision making for children with

life-limiting conditions. For example, in relation to gastrostomy insertion, prior to this research my professional perspective would have almost certainly been that professionals ought to persuade the family to agree. However, following this research, my opinion is more ambiguous, as I now have a greater understanding of the unique expertise that parents have regarding their child and the child's own perspective. Additionally, the reality is that regardless of the professional perspective, it will be the family who are left to manage the consequences of any decision at home and for the rest of the child's life.

Despite this, professionals do have a duty, with their knowledge of the physical impact of treatment decisions or refusals, to ensure that the child does not come to harm. I recognise that when I am in the professional role, my perspective may transition once again towards a more paternalistic one, through the necessity to act in accordance with my professional code of conduct. Such a consideration further demonstrates the role conflict which exists when a clinician concurrently engages in research and practice.

This study suggested that persuasion and pressure, either from professionals or from family members, are not the factors which lead to agreement, rather it is recognition of evidence that the suggested treatment is necessary. Nevertheless, persuasion, by all stakeholders, was observed to be an intrinsic part of the process. It has prompted me to consider whether as professionals, the desire to act in a persuasive capacity over a number of years results from the need to justify their presence in the lives of these families who have become expert in the child's care. Nevertheless, it is important to be able to define where a family refusal to agree to a medically recommended treatment constitutes a case of neglect and safeguarding procedures need to be commenced. Such a situation is less clear and my opinion remains that intervention at this point is likely to be necessary.

On overall reflection, this process has provided me with a detailed insight into the complexity of decision making, the competing moral work of stakeholders and the significant care burden, which the families I have routinely cared for in hospital, carry in their daily lives. It has radically altered my worldview and I

believe that I have emerged from the process as a more enlightened clinician and a more thoughtful researcher.

8.10. Strengths and Limitations

This study aimed to elicit multiple perspectives regarding decision making, through multiple stakeholder interviews. The multiple perspectives elicited and the longitudinal design acted as a strength for this study, as they facilitated an understanding of the inter-relations involved in the process of decision making (Kendall et al., 2009) and were able to capture any changing opinions or events (Murray et al., 2009) respectively. The serial interviews, alongside my presence at appointments, enabled the development of trust and a rapport with the participants, in which intersubjective understandings were developed (Cook and Crang, 1995). Serial interviews also facilitated the discussion of sensitive topics within subsequent interviews and the progressive focusing of the research question (Stake, 1995, Murphy et al., 1999). The case study approach enabled the specifics of individual cases to be analysed, so as to identify how multiple perspectives were integrated in the process of decision making and this provided understanding of the commonalities across cases, which an alternative approach may not have facilitated.

It is not possible to identify the characteristics of the families who were not invited to or chose not to participate in the study. For this reason no comparison can be made between those families who took part and those who did not, which may have shed a different light on the process of decision making. The study recruited a small number of cases, but the volume of data contained within these cases enabled a depth of analysis and insight which would not have been possible with a larger sample, given the study resources, and this therefore acts as both a strength and limitation of the study.

The ethnicity of the recruited families was representative of the local population, which acts as a strength regarding the cultural appropriateness of the findings. However, the exclusion of families who could not speak English, may have meant that other factors pertinent to decision making for non-English speaking families were not identified.

In one case, obtaining the narrated views of significant others was not possible, as they did not wish to nominate anyone else. Although it would have been useful in such cases to interview significant others, observations of decision making interactions provided some indication of the perspectives of professionals and other family members. However, how people interact within appointments may not be fully representative of their feelings on the case, as what is said will be constructed in response to the specific context, purpose of the consultation and interaction with those present.

The recruitment to this study resulted in the dominance of the maternal perspective over both the paternal, child and professional view. Mothers were the ones who primarily attended appointments and were therefore likely to be the ones who received the study information and decided whether to participate. Not all mothers nominated professionals, or fathers, to take part which may have been due to a worry that they were too busy to participate, or due to a perception that they alone were the ultimate decision maker. However, particularly for professionals, observations demonstrated that they were indeed instrumental in decision making, so their lack of nomination by mothers was in itself an interesting insight. The lack of the paternal voice within this study means that the extent to which the mothers' perspectives can be considered representative of the fathers' opinions is unclear and the process of decision making within the parental dyad remains unknown. Further work should aim to recruit parent dyads and specifically focus on fathers' views, including one-to-one interviews with them, in order to address this knowledge deficit.

The child voice in the present study was minimal, but this was a result of the age and cognitive abilities of the children recruited. Thus the child's role in decision making was not able to be fully explored within the current study and therefore additional issues relating to their role in the process may not have been possible to illuminate. This presents another area in need of further research.

8.11. Implications for future research

This study has provided an insight into the complex and nuanced process of decision making for children with life-limiting conditions. However, this research, in consideration with the wider literature, has raised many more

questions which would benefit from further research. As noted, one such question relates to the experiences and perspectives of fathers on their role in decision making. Similarly the role of the child in decision making would also benefit from further investigation.

This research provided an understanding of the strategies which enable the maintenance of good working relationships and collaborative decision making. While there remains room for improvement in the process of decision making, it is striking that conflict and escalation to the court is rare and primarily collaboration between families and professionals is maintained. However, in order to fully understand the process of decision making for children with life-limiting conditions, an investigation of decision making in situations where there is protracted disagreement, conflict and escalation to the courts, is warranted. Such an investigation could be ethically fraught, but nevertheless could help to identify the antecedents of such conflict and the most effective solutions at overcoming such disagreements.

Further research should also seek to better understand inter-professional communication practices of professionals who care for children with many other specialty colleagues. Interventional research aimed at improving these communication practices is also of immediate importance. Given the technologically advanced age within which we live, it is relevant to consider how current systems can be developed to promote better communication and information sharing between teams and organisations. Research and clinical consideration of strategies to minimise the need for the family to adopt the additional burden of care coordinator on top of their other caring responsibilities is warranted.

In the present study professionals referred to the limits of parental decisional responsibility in relation to concerns they had about harm to the child. This was not perceived to be a big issue for families, but weighed heavily on professionals who appeared to be undertaking a significant amount of moral work to determine whether the harm was sufficient to warrant escalation to the courts. Further guidance from professional bodies on what constitutes sufficient harm to warrant such escalation, would be useful in assisting professionals to make such

decisions. Such detailed guidance could also help to prevent the 'roster lottery' as described by Wilkinson and Truog (2013), so that physician variability in ethically challenging decisions is minimised.

This research also highlighted the necessity for professionals to engage in critical reflection in relation to their decision making practice. Two of the doctors who participated in the study remarked that their participation had been useful for them as a form of clinical supervision in which they were facilitated to critically reflect on their own practice. This suggests that active professional reflection on their approaches to decision making, may be useful for their professional development and facilitate a greater awareness of the complexities of decision making and the most effective approaches.

Finally, this research has powerfully demonstrated the work undertaken by all stakeholders in decision making for children with life-limiting conditions. It has highlighted the importance that families place on making decisions for their children, but that the relational context within which decisions are made, means that their autonomy is effectively relational, rather than individual. Although conflict was not a key feature of this research, its possibility was alluded to, suggesting that in some situations, the strategies which were found to be so effective at promoting collaboration in this study, may be insufficient. Additionally the protracted nature of some decisions in the present study highlighted the need for better awareness between stakeholders of the underlying assumptions, ethical factors and wider considerations which may be present for some decisions. Professionals should be aware of the relational ethics approach (Carnevale et al., 2017) to decision making and adopt this in their practice. This approach may assist individuals to identify the moral work that the other stakeholders are undertaking in their decision making and therefore better avoid disagreements and promote decisions which are made in partnership.

This approach could also assist in identifying perceived evidence thresholds and therefore help to facilitate discussion of when the treatment is likely to transition from the professionals' perception of being the moral ideal, to their professional duty, or legal requirement. Professional awareness of relational ethics may further facilitate the maintenance of good relationships and possibly ensure that

contentious decisions occur in the context of understanding and support, rather than sustained pressure. Research is needed to further examine the effectiveness of this approach within paediatrics.

8.12. Conclusion

The conceptual framework which has been developed from the findings of the current study has been presented and has facilitated the understanding of decision making as a relational process, situated within a specific cultural, professional and legal context. Understanding decision making as a relational endeavour, within which individuals with specific ethical concerns, perceived moral duties and emotional influences, suggests the need for an approach in which these are discussed openly and appreciated by the other relevant stakeholders. In order to develop and maintain trusting relationships, professionals ought to adopt a relational ethics approach to their decision making interactions and thereby more effectively facilitate collaboration and SDM.

Chapter 9. Conclusion

This study has explored the process of decision making for children with life-limiting conditions, including the experiences and preferences of those involved, and how decisions are communicated and negotiated within the child's network of care. The study adopted a longitudinal qualitative case study approach, utilising methods of observation, serial interviews and documentary analysis of the child's medical notes. Eleven cases were recruited. In one case the parents consented only to participate in a single interview, one case withdrew after two months and the other cases were followed for between six and 12 months. This research demonstrated the importance of good quality relationships and effective communication for decision making and highlighted the different types of decisions which were perceived to exist. Future planning was identified to be an integral aspect of decision making. The data uncovered numerous strategies that are employed in the quest for agreement between the various stakeholders involved. For example, persuasion, appeal to the evidence and invoking the concept of best interests, which included reference to the possibility of escalation to the courts, although this occurred rarely. Justification was also used by individuals as a way of demonstrating, to themselves, to me as a researcher or to other stakeholders, that they were acting in a morally responsible manner.

This study has enabled the temporal, processual and relational nature of decision making to be more fully illuminated. It facilitated a detailed understanding of individuals' opinions regarding decisions and how these were negotiated and combined in the endeavour to reach a decision agreeable to all. It displayed the largely cooperative nature of decision making, albeit demonstrating the distribution of power for different decisions, which influenced the extent to which decisional responsibility was distributed between the family and professionals.

Parents articulated moral responsibility for decision making which stemmed from their belief that, given their intricate knowledge of their child, they knew what was best for them. This moral imperative enabled them to assert their opinion within the process of decision making, request assistance in decision

making or refer to professional guidance when they felt unable to make the decision themselves. Professionals perceived a moral duty to engage families in decision making and guide them towards a decision which would be, in their opinion, in the child's best interests. Such feelings of moral duty promoted a largely collaborative process of decision making, which was a frequent occurrence for children with life-limiting conditions. However, such feelings of moral duty could also result in conflict when perceptions of the child's best interests diverged. Although such conflict was not observed within this research, professionals in several cases alluded to the possibility of escalating sustained disagreement to the courts in order to obtain a decision which they considered to be in the child's best interests. Even though exceptional, this option highlighted that there may be limits to collaboration in decision making. Such instances were rare and it appeared that re-evaluation of the existing evidence and assessment of new evidence led to agreement, prior to the point at which SDM would become impossible. Additionally, collaborative relationships were maintained through a process of containment. Containment was achieved largely through parental recognition of the moral work that professionals were undertaking when they challenged parental decisional responsibility. The recognition of these strategies highlighted the importance of developing and maintaining strong and effective relationships, which promote collaboration, weather disagreement and eventually result in mutual agreement.

The rarity of conflict and unresolved disagreements demonstrates the effectiveness of the strategies, which this study has outlined, that are routinely employed in the process of decision making. Greater professional insight and awareness of the strategies and processes of decision making could enable more effective communication and support for families confronting the challenges of treatment decision making for children with life-limiting conditions.

Appendices

Appendix 1: Thematic synthesis exemplary quotes

Theme	Sub-theme	Exemplary quotes
Decisional Factors	Timing of decisions	"Thinking too much, I think that gets you into a lot of trouble. That is what I kind of adopted the philosophy of. You kind of deal with it one day at a time and you deal with whatever needs to be dealt with at the time. Because I think if you constantly ponder on the future, I don't think you would ever be able to function on a daily basis. I wouldn't be able to function on a daily basis." (Erby et al, 2006 - Parent) Not all were first-time decisions. For half, the decision had been previously considered and rejected or postponed, leading it to become an on-going choice to be revisited when (medical/health) circumstances demanded or the young person initiated revisiting the decision. (Mitchell 2012 – Results Section) There a distinction between the overall decision process, which can last many years as a child's symptoms worsen and parents continually assess their child's needs, and the actual process of deciding, the latter of which is defined as the time during which families weigh up the appropriateness of technological support, and acquire the information they need to make an informed decision. For some families, the duration for these different aspects of the decision process is the same, and this was certainly the case for families who had not expected a recommendation for technological support to be made. (Nicholson, 2012 – Results Section) Several participants expressed the opinion that the resource was probably not appropriate for distribution in acute situations, in ICU for instance, where a child's death was imminent and/or unexpected, and parents had very little time to make a decision and process rapidly developing devastating events. However, they thought that where a child's health was slowly deteriorating and where there was a possibility of planning ahead, it would confer benefits. (Delany et al., 2017 – Results section) All of the parents in this study had previously experienced serious illnesses and hospital admissions for their child. These

		experiences made parents think about what their wishes would be when their child became critically ill again, even before physicians started to speak about this issue. For this reason, almost half of the parents mentioned that they felt it was a missed opportunity that physicians did not take the initiative to talk about EoLDs when the child was still in a stable condition. (Zaal-Schuller et al, 2016)
	Consequences	"When he used to have that tube in his nose and he was always pulling it out, and it was always getting sore and we were always having to put it back in his stomach. It was horrible. It was just horrible you know, having to go through that every time it came out or dislodged itself. So you know, in that sense, it made having a decision, a PEG or a button, it made it easier really in that sense. Because you didn't want to see that all the time." (Nicholson, 2012 - Parent) A minority of parents 'framed' the decision in terms of finding something local to occupy the young person during the day. However most drew on notions of 'normality', talking about their son/daughter's right as a young adult to move on in life, to make their own choices and achieve maximum independence within the challenges set by their physical impairments and (where relevant) learning difficulties (Mitchell et al, 2011 – Results section)
	Decisional considerations	The young people engaged in a complex process of trade-offs, considering both the costs/risks and benefits to their health of accepting a medical intervention, alongside broader social and emotional quality of life factors such as the importance of 'normality' and preserving nonmedicalised aspects of their life. Choosing when to accept an intervention often involved balancing these needs or aspirations with medical necessity, sometimes even life-preservation. (Mitchell et al, 2011 – Results section) "[H]e's never going to see, hear, walk . . . you feel you're as bottom as you can go . . . how do you as an individual ultimately interpret that? Is that what you really want? Or is that good for, for [child]? How does that play out in your life plan and what does it mean for everybody else, including [daughter], you know? Our other daughter and ourselves as well, still as a family, how's all this gonna work if you go down 'route A' as opposed to 'route B'?" (Rapoport et al, 2013 - Parent)
	Difficulty in decision making	Multiple barriers to successfully managing the process were identified, including time constraints, conflicting clinical demands and lack of formal training in communication skills. In the PICU setting, a lack of established rapport with the family before having to raise the issue of end-of-life care for the first time during acute situations, including resuscitation, was identified as causing particular difficulty (Mitchel and Dale, 2015 – Results section)

		"And we're exhausted at that point, absolutely at the end of our tether, you know, and then making these decisions when you're in that state. In this totally, mentally, physically exhausted state, and that's not taken into consideration all the time." (Nicholson, 2012 - Parent) The decision about mechanical ventilation was hard for me. I wish I could decide at the moment that mechanical ventilation has to be installed. If she gets a heart attack or something is wrong with her heart, I would say, "I don't want that." But if something else is wrong, something that has never been a problem before, in that case, it's a pity not to try mechanical ventilation. (Zaal-Schuller et al, 2016 – Parent)
	Future planning	"If they make an ACP they may still change their mind right at the very end, um … But at least they will have had the opportunity to sit down and seriously think about what they want for their child." (Mitchell and Dale, 2015 - Nurse) There was some evidence in the data that acknowledgement is more likely for children and young people with conditions where it is expected there will be feeding or respiratory problems, especially when compared to children with undiagnosed or very complex conditions. For these families, a child's swallowing and respiratory function is sometimes monitored for years, and the decision was described by some families as a planned one, gradually introduced to families at different time points before being initiated at the appropriate time. In fact some young people recall always knowing they would need ventilation and therefore accepting it as inevitable, and do not always identify having made a decision about ventilation per se, but rather about the timing, type, and location of initiation (Nicholson , 2012 – Results Section) It just made sense. In the years before it was very clear to me. . . only it had never been written down before. . . but for me, it was very clear that these parents. . . that these parents did not want to push it to the limits and did not want their child to end up at the PICU with invasive ventilation, failing to wean off the ventilator. (Zaal-Schuller et al, 2016 – Physician)
Relational Factors	Gathering opinions	"Our end decision would have still be mine. But it helped having somebody else's input, other people's decisions coming in as well…..You know their points of view came in as well and I thought yes. You know sometimes when you're in that situation you can't think, you can't always think straight really can you, sort of think what's best, and so having other people's decisions made it slightly easier for me ." (Nicholson, 2012 - Parent)

		I mean I do listen to mum a fair bit, I mean it's my decision in the end but I do listen to mum a lot .. . cos she's very clever, I don't know it just, well she's my mum and she knows best ... yeah, I trust my mum's opinion. (Mitchell, 2012 – Young Person)
	Involvement in decisions	Within the discussion of young people's developing capacity, it is clear that parents play a central role in facilitating their child's participation in decisions that affect them. However, like children, parents embark on their own journey as decision makers, and this in some ways resembles the journey children make in that parents also develop capacity for decision-making over time, and the parents in this study provide remarkably similar accounts of moving from being dependent on the healthcare professionals involved in their child's life to assuming control of the decisions that need to be made for their child. (Nicholson, 2012 – Results section) "Well now as I'm getting a lot older they'll [family] listen to me but like when I were younger, five or six like, me mum had to make decisions for me and so did the hospital but now, like when I got to, like 10, that's when me mum and our [brother] started saying like 'no, do you want to do this?" (Mitchell, 2012 – Young person)
	Trust and confidence in staff	"The only one time we didn't stay, she just, she lost a day of her life because somebody messed up with drugs…..So she's never been left after that. I mean cock ups have happened since, with the drugs, I mean they're strong drugs….that's why whenever she goes in now, we always say "we'll medicate Emily"." (Nicholson, 2012 - Parent) "[S]he had that casual and confident way about her that just makes you feel that she knows what she's talking about. You have faith in her, and, it just makes it all work." (Rapoport et al, 2013 - Parent) "I'd definitely feel there'd be situations where the nurse would be appropriate to give it or to talk to the doctors about the doctors giving it. You know, some families respond best to the bedside nurse because the bedside nurse is there for 8 to 12 hours at the time whereas some of them only really trust their medical team and if that's the case, then the medical team would be the most appropriate because some of them find the social worker the most consistent to being throughout their admission so the social worker would be the most appropriate". (Delany et al., 2017 - PICU nurse) Parents had many experiences in the past that physicians had not taken into account. Two parents blamed their physicians

		for making medical errors that caused severe brain damage in their child. These negative healthcare encounters had contributed to a critical attitude towards physicians. (Zaal-Schuller et al, 2016 – Results Section)
	Agreement in decisions	"At some point, once you realise that you have a better idea of what's wrong with your child, and how serious it is, you can say "well actually, we respect your opinion but on this occasion…."…..we grew to trust ourselves to make the right decisions for Robert." (Nicholson, 2012 - Parent) "Before you can convince any parents, you have to convince the other specialties. You have to bring them on board. If they're not on board, you have no chance, or your chances with the family are much much less." (Mitchell and Dale, 2015 - Doctor)
	Communication and Information exchange	"It was quite hard hitting, it was quite honest questions, that actually you know it might be that, [now referring to the consultant] "I've got to be honest with you Lee. I might actually put you under anaesthetic and actually, we might not be able to do it because, can you see from these x-rays, you know, some of your bits aren't in all the right places"." (Nicholson, 2012 - Parent) Health care professionals who were helpful to parents acknowledged the child's perceived poor QoL and feeding difficulties and explained why FANH (Foregoing Artificial Nutrition and Hydration) was a reasonable option. Parents appreciated that the option was discussed in a sensitive, nonjudgmental, and supportive way. They also appreciated being given enough time to reach their decision; within hours to a few days of being presented with the option all parents concluded that FANH was best for their child. (Rapoport et al, 2013 – Results section) "It doesn't particularly say anything new or anything that we wouldn't tell parents anyway but I think the beauty of it is that it gives them something to take away and, you know, with any of the sessions that we have with our families, the information is so complex and there's so much of it that I don't really expect them to take more than about 10 to 20% of it away in their minds, which is why we have these meetings repeatedly but I see enormous value in them being able to take something away and read quietly later because I think a lot of what's written in the book would trigger memories about what was said in meetings that would allow them to feel more comfortable with the decision at a time when they're drowning in complex information overload". (Delany et al., 2017 – Neonatologist)

		"So I tell them it's a very serious condition and that it's a progressive condition. And if the parents ask me, "How will it go?" In those cases, I answer, "How was your child's condition one month ago? And how is it now?" I say, "You can draw a line though that." (Zaal-Schuller et al, 2016 – Physician)
	Information and Support	"The person who's on the ventilator understands because they're on it. They know how they feel." (Nicholson, 2012 - Parent) Written information about the procedure, or supplementary information found on the internet or provided by third party organisations such as condition specific charities is useful but it does not make up for the invaluable information and opinion provided during discussions with healthcare professionals, and with other families themselves (Nicholson, 2012 – Results section) Young people without LCIs felt that peers with similar conditions could be a source of 'inside information' and/or emotional support. 'Inside information' was regarded as 'lived' or experiential information from a person who had previously faced a similar choice. This was valued as it was a type of information which the young people felt professionals or parents could not provide. (Mitchell, 2012 – Results section) "It would help those who are struggling to make a decision – seeing others have gone through it would help". (Delany et al., 2017 - PICU nurse)
Family factors	Coping	"Sometimes I think I talk about things and I start to cry, and I don't know why….I get so upset. [Child's name] ain't going to get better. I don't know what to do, I'm doing the best I can, but I start to talk about it and my chest gets so tight. I don't know what I can do to help him, and the next thing I know I'll be crying." (Wissow et al, 2001 – Child's Guardian) When hope was not accessible to parents, some expressed a tendency to get from day to day by avoiding the reality of muscular dystrophy to some extent. (Erby et al, 2006 – Results section)
	Awareness	"Dad: "We didn't realize. We thought, oh it's just a bit of food. It's got in with her snot basically. We were thinking things like that. And it were getting worse wasn't it?

		Mum: “Yeah but because she were eating it didn’t matter.”” (Nicholson, 2012 - Parent) Over time, however, Sebastian’s parents noticed a change in Sebastian’s breathing overnight, and they were unsure of the reasons for this. This development triggered Sebastian’s parents to seek medical advice, and this point can be seen as the start of the decision process that led to overnight ventilation. (Nicholson, 2012 – Results section) Considerations about the most appropriate time to provide parents with the resource as well as the best approach to communicating information to parents was also linked to diagnostic and prognostic uncertainty. Interviewees spoke of wanting to assist parents to move from a denial of the facts to a greater understanding of the complexity of the prognosis and felt that the resource may assist in this process. (Delany et al., 2017 – Results section)
	Educating and fighting	“So I took them all kinds of stuff. All kinds of literature and stuff, and I kind of just threw it on the table at this meeting and said, ‘It’s kind of like your job to just read it.’ ” (Erby et al, 2006 - Parent) “You have to have all the information to make the right choices, especially if it’s a child, for your child. So you have to fight. Don’t just sit back and take their word for it. You have to fight.” (Nicholson, 2012 - Parent) The parents mentioned that disturbing disagreements arose especially after an acute deterioration of their child’s condition because decisions had to be made under time pressure and often without their regular physician. In those cases, the parents felt that they were not heard and that the physicians regarded the life of their child as less valuable than the life of a typically developed child. This strengthened their feeling that they had to advocate for their child. (Zaal-Schuller et al, 2016 – Results Section)
	Family life	“A lot goes on behind the scenes, it’s like a fabulous film but nobody knows what goes on to make it … and it’s a lot of work, a lot of stress and, in-between all that, people are trying to live normal lives aren’t they, you know, do the supermarket shopping and take the kids to the dentist … all the mundane things you do as a parent … I do a lot of juggling, I keep a lot of balls in the air.” (Mitchell et al, 2011 - Parent) “You look at trees differently. You embrace life in a different way. We have done more traveling because we feel now that

		[our son] has to experience everything before he runs out of time to experience stuff, you know? We'll have dessert for dinner. We kiss the monkeys. You're not in pain. Everyone else is so serious about ... we just let our children be children and we live a wonderful life because of [our son]." (Erby et al, 2006 - Parent)
System Factors	Problems with the system	As well as the loss of key resources for decision-making, families talk about the separation of services due to the different infrastructure making up adult health care. Some young people will have several new consultants, none of whom take overall responsibility for their care, and none of whom specialise in their condition, instead each having an organ or function which they accept responsibility for. In some cases, these professionals can be based at different hospitals therefore further fragmenting the care young adults receive. For families who are able to maintain a relationship with one of the professionals they have come to trust over many years, this resource is highly valued, and can become the primary contact for on-going problems their adult child may now experience. (Nicholson, 2012 – Results section) "Now it may have been August time but it required several people to liaise in order to make a decision about providing oxygen but they were on holiday at different times....and every night Sebastian had this problem so it's incredibly cruel and reflects this indifferent system of rule following.....Anyway, eventually they all came back, had a consultation. And it was agreed that yes Sebastian certainly did need oxygen." (Nicholson, 2012 –Parent) "These organisations are like super tankers…..They've got a momentum of their own and they don't, they're inflexible. They can't cater for special circumstance. They're sort of like sausage machines. So children like Sebastian, they can't cope with." (Nicholson, 2012 - Parent)
	Professionals	"When I was a registrar it was easier for me because I had to just sit and have a debrief with my … consultant, as I love to cry. But now, I have to be this brave person and it's very … very difficult." (Mitchell and Dale, 2015 - Doctor) "Anybody who says the NHS works hasn't experienced it properly….I reckon 80% of the staff who work in the NHS aren't that interested….They turn up, do their job, go home, forget about it. The other 20% actually care. But you've got to find them." (Nicholson, 2012 - Parent) "By the time they've [nurses on the ward] done 12 hour shifts. If it's full on, by time you've got to 10 hours they're absolutely

		zonked. It's too long…..Three days….Three 12 hour shifts. 36 hours that's it. Get it all done in three days…..but then if you got a good nurse, where you were confident with, and you thought, bloody hell she's good." (Nicholson, 2012 - Parent)
	Medicalisation	"So I think there are many examples in my head and in my career where we have over-medicalised the end-of-life and families have clung on to the medical management rather than the holistic palliative care management of children, such that they feel that they're failing their child if they don't ask for everything." (Mitchell and Dale, 2015 - Doctor)
	Access to support	"It may well have been that I phoned the consultant, I was told by the secretary of one consultant, "consultants don't talk to the patients. You know, you can't ring up and have an informal conversation". So when you're meeting barriers like that it's difficult. We had a heck of a struggle to rectify, to overcome this problem." (Nicholson, 2012 - Parent) "At the end of the meeting he pushed his card across and he said, and he turned it over and on the back was written his home telephone number. And he said "you've a difficult time, any problems ring me at home." Now that obviously is the sort of person you want. Not some-one who thinks, I am, you know, mini god." (Nicholson, 2012 - Parent)

Appendix 2: Ethics and Governance Approvals

NHS
Health Research Authority
East Midlands - Nottingham 1 Research Ethics Committee
Royal Standard Place
Nottingham
NG1 6FS

Telephone: 0115 8839273

30 September 2015

Professor Bridget Johnston
Professor in Palliative and Supportive Care
University of Nottingham,
Sue Ryder Care Centre,
University of Nottingham
Room B61C, Queens Medical Centre, Nottingham
NG7 2UH

Dear Professor Johnston,

Study title:	**Decision making and future planning for children with complex illnesses: a qualitative multi-stakeholder, longitudinal study**
REC reference:	**15/EM/0377**
Protocol number:	**15068**
IRAS project ID:	**165505**

Thank you for your letter of 24th September 2015, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair.

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details. Publication will be no earlier than three months from the date of this opinion letter. Should you wish to provide a substitute contact point, require further information, or wish to make a request to postpone publication, please contact the REC Manager, Ms Rachel Nelson, NRESCommittee.EastMidlands-Nottingham1@nhs.net.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Conditions of the favourable opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission ("R&D approval") should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements.

Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at http://www.rdforum.nhs.uk.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of approvals from host organisations

Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publically accessible database within 6 weeks of recruitment of the first participant (for medical device studies, within the timeline determined by the current registration and publication trees).

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non clinical trials this is not currently mandatory.

If a sponsor wishes to contest the need for registration they should contact Catherine Blewett (catherineblewett@nhs.net), the HRA does not, however, expect exceptions to be made. Guidance on where to register is provided within IRAS.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Ethical review of research sites

NHS sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Non-NHS sites

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

Document	*Version*	*Date*
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [2015 To Whom it May Concern Liability Insurance]	V1	21 July 2015
Interview schedules or topic guides for participants [TOPIC GUIDE Decision making and future planning for children with complex illness V1 030815]	Final Version 1.0	03 August 2015
Letter from sponsor [2015_08_04_13_03 REC Sponsorship Letter]	V1	04 August 2015
Other [REPLY SLIP Decision making and future planning for children with complex illness V1 030815]	Final Version 1.0	03 August 2015
Other [DISTRESS PROTOCOL Decision making and future planning for children with complex illness V1 030815.docx]	V1	03 August 2015
Participant consent form [CONSENT FORM CHILD 16-18 Decision making and future planning for children with complex illness v1.1 220915]	V1.1 220915	22 September 2015
Participant consent form [CONSENTFORM PARENT Decision making and future planning for children with complex illness V1.1 220915]	V1.1 220915	22 September 2015
Participant consent form [CONSENTFORM PARENT FOR CHILD Decision making and future planning for children with complex illness V1.1 220915]	V1.1 220915	22 September 2015
Participant consent form [CONSENTFORM PROFESSIONAL Decision making and future planning for children with complex illness V1.1 220915]	V1.1 220915	22 September 2015
Participant consent form [CONSENTFORM SO Decision making and future planning for children with complex illness V1.1 220915]	V1.1 220915	22 September 2015
Participant information sheet (PIS) [PIS CHILD 7-10 Decision making and future planning for children with complex illness V1.1 220915]	V1.1 220915	22 September 2015
Participant information sheet (PIS) [PIS CHILD 11-15 Decision making and future planning for children with complex illness V1.1 220915]	V1.1 220915	22 September 2015
Participant information sheet (PIS) [PIS CHILD 16-18 Decision Making and future planning for children with complex illness V1.1 220915]	V1.1 220915	22 September 2015
Participant information sheet (PIS) [PIS PARENT Decision making and future planning for children with complex illness v1.1 220915]	V1.1 220915	22 September 2015
Participant information sheet (PIS) [PIS PARENT FOR CHILD Decision making and future planning for children with complex illnesses v1.1 220915]	V1.1 220915	22 September 2015
Participant information sheet (PIS) [PIS PROFESSIONAL Decision making and future planning for children with complex illness v1.1 220915]	V1.1 220915	22 September 2015
Participant information sheet (PIS) [PIS SIGNIFICANT OTHER Decision making and future planning for children with complex illness v1.1 220915]	V1.1 220915	22 September 2015
REC Application Form [REC_Form_04082015]		04 August 2015
Research protocol or project proposal [PROTOCOL Decision making and future planning for children with complex illness V1.1 220915]	V1.1 220915	22 September 2015

Summary CV for Chief Investigator (CI) [CV Bridget Johnston 090715]	V1	09 July 2015
Summary CV for student [EP Research CV 230615]	V1	23 June 2015
Summary CV for supervisor (student research) [Kristian Pollock 2 p 9.7.15]	v1	09 July 2015

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Reporting requirements

The attached document *"After ethical review – guidance for researchers"* gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The HRA website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website:
http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/

HRA Training

We are pleased to welcome researchers and R&D staff at our training days – see details at http://www.hra.nhs.uk/hra-training/

15/EM/0377	**Please quote this number on all correspondence**

With the Committee's best wishes for the success of this project.

Yours sincerely,

pp

Dr Carl Edwards
Chair

Email: NRESCommittee.EastMidlands-Nottingham1@nhs.net

Enclosures: "After ethical review – guidance for researchers"

Copy to: *Mrs Angela Shone*

Maria Koufali

NHS
Health Research Authority

Professor Bridget Johnston
Professor in Palliative and Supportive Care
University of Nottingham
Sue Ryder Care Centre
University of Nottingham
Room B61C
Queens Medical Centre
Nottingham
NG7 2UH
Email: hra.approval@nhs.net

05/07/16

Dear Professor Bridget Johnston,

Letter of HRA Approval for a study processed through pre-HRA Approval systems

Study title:	**Decision making and future planning for children with complex illnesses: a qualitative multi-stakeholder, longitudinal study**
IRAS project ID:	**165505**
Sponsor:	**University of Nottingham**

Thank you for your request for HRA Approval to be issued for the above referenced study.

I am pleased to confirm that the study has been given **HRA Approval.** This has been issued on the basis of an existing assessment of regulatory compliance, which has confirmed that the study is compliant with the UK wide standards for research in the NHS.

The extension of HRA Approval to this study on this basis allows the sponsor and participating NHS organisations in England to set-up the study in accordance with HRA Approval processes, with decisions on study set-up being taken on the basis of capacity and capability alone.

If you have submitted an amendment to the HRA between 23 March 2016 and the date of this letter, this letter incorporates the HRA Approval for that amendment, which may be implemented in accordance with the amendment categorisation email (e.g. not prior to REC Favourable Opinion, MHRA Clinical Trial Authorisation etc., as applicable). If the submitted amendment included the addition of a new NHS organisation in England, the addition of the new NHS organisation is also approved and should be set up in accordance with HRA Approval processes (e.g. the organisation should be invited to assess and arrange its capacity and capability to deliver the study and confirm once it is ready to do so).

IRAS project ID	165505

Participation of NHS Organisations in England

Please note that full information to enable set up of participating NHS organisations in England is not provided in this letter, on the basis that activities to set up these NHS organisations is likely to be underway already.

The sponsor should provide a copy of this letter, together with the local document package and a list of the documents provided, to participating NHS organisations in England that are being set up in accordance with HRA Approval Processes. It is for the sponsor to ensure that any documents provided to participating organisations are the current, approved documents.

For non-commercial studies the local document package should include an appropriate Statement of Activities and HRA Schedule of Events. The sponsor should also provide the template agreement to be used in the study, where the sponsor is using an agreement in addition to the Statement of Activities. Participating NHS organisations in England should be aware that the Statement of Activities and HRA Schedule of Events for this study have not been assessed and validated by the HRA. Any changes that are appropriate to the content of the Statement of Activities and HRA Schedule of Events should be agreed in a pragmatic fashion as part of the process of assessing, arranging and confirming capacity and capability to deliver the study. If subsequent NHS organisations in England are added, an amendment should be submitted to the HRA..

For commercial studies the local document package should include a validated industry costing template and the template agreement to be used with participating NHS organisations in England.

It is critical that you involve both the research management function (e.g. R&D office and, if the study is on the NIHR portfolio, the LCRN) supporting each organisation and the local research team (where there is one) in setting up your study. Contact details and further information about working with the research management function for each organisation can be accessed from www.hra.nhs.uk/hra-approval.

After HRA Approval

In addition to the document, *"After Ethical Review – guidance for sponsors and investigators"*, issued with your REC Favourable Opinion, please note the following:

- HRA Approval applies for the duration of your REC favourable opinion, unless otherwise notified in writing by the HRA.
- Substantial amendments should be submitted directly to the Research Ethics Committee, as detailed in the *After Ethical Review* document. Non-substantial amendments should be submitted for review by the HRA using the form provided on the HRA website, and emailed to hra.amendments@nhs.net.
- The HRA will categorise amendments (substantial and non-substantial) and issue confirmation of continued HRA Approval. Further details can be found on the HRA website.

The HRA website also provides guidance on these topics and is updated in the light of changes in reporting expectations or procedures.

IRAS project ID	165505

Scope

HRA Approval provides an approval for research involving patients or staff in NHS organisations in England.

If your study involves NHS organisations in other countries in the UK, please contact the relevant national coordinating functions for support and advice. Further information can be found at http://www.hra.nhs.uk/resources/applying-for-reviews/nhs-hsc-rd-review/.

If there are participating non-NHS organisations, local agreement should be obtained in accordance with the procedures of the local participating non-NHS organisation.

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please email the HRA at hra.approval@nhs.net. Additionally, one of our staff would be happy to call and discuss your experience of HRA Approval.

HRA Training

We are pleased to welcome researchers and research management staff at our training days – see details at http://www.hra.nhs.uk/hra-training/.

If you have any queries about the issue of this letter please, in the first instance, see the further information provided in the question and answer document on the HRA website.

Your IRAS project ID is **165505.** Please quote this on all correspondence.

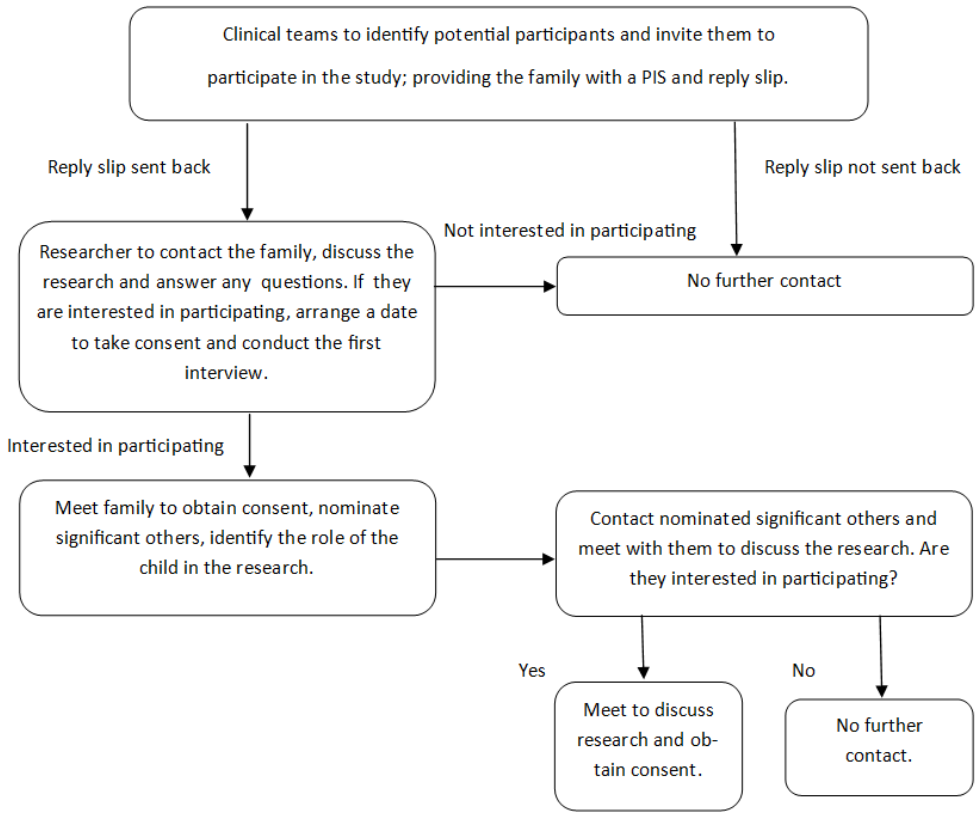
Yours sincerely

Kate Radford
Applications Administrator

Email: hra.approval@nhs.net

Copy to: Angela Shone, University of Nottingham
Dr Maria Koufali, Nottingham University Hospital NHS Trust

Appendix 3: Recruitment flowchart



Appendix 4: Interview topic guides

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Sherwood Forest Hospitals NHS

Nottingham University Hospitals NHS

Topic Guide
Final Version 1.0

The MAPS Study



Decision making and future planning for children with complex illness.

Interviews will be semi-structured, with a broad opening question asking the participant about the child at the centre of the case; both in terms of their illness and their context (family, personality, values and goals). Although the topics of decision making and future planning will be covered within the interview, the way in which these are initiated are likely to be different for each participant and asked in relation to the responses given to the opening question.

Professionals:

Professionals will be asked to provide an account of the child's illness, their family context, goals of care and their prognosis. The interviews will cover the important decisions that the family have had to consider and the likely decisions that they may consider in the future. They will also seek to identify the professional's perception of the family's receptivity to discussions about decision making and future planning and the way in which such discussions are usually initiated with this family.

Parents:

Parents will be asked about their family's story, their child's illness and their understanding of it. The interviews will seek to identify their previous experience of having made decisions or plans for the future and the results of these, who was involved in decision making and their thoughts about what they would do if similar event occurred in future. The interviews will also seek to discover how they feel about planning for their child's future care.

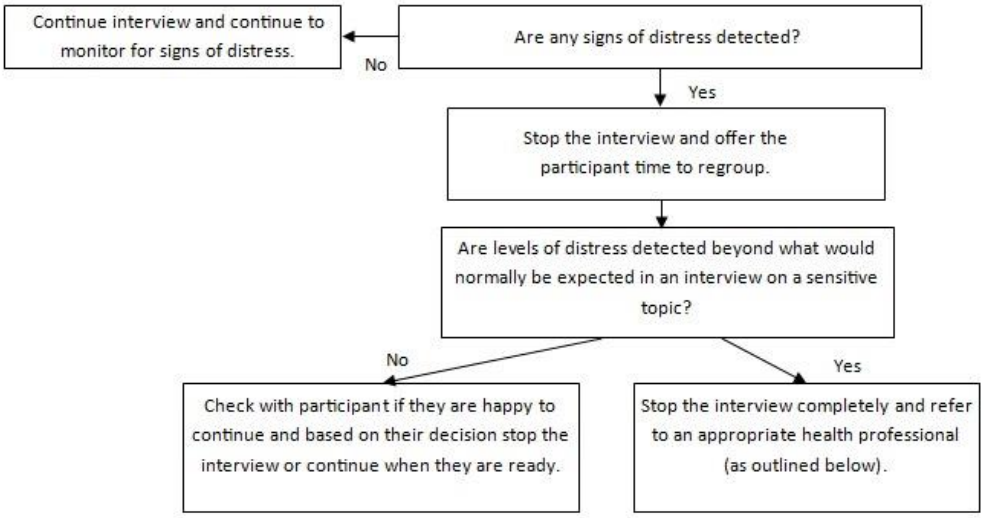
Children:

The child will be asked to tell the researcher about themselves and their family. They will be asked what they understand about their illness so that the researcher is aware of their level of knowledge about their condition. They will be asked how they feel about their treatment and how these treatments are chosen. The interviews will also focus on the child's goals, wishes and ambitions for the future.

Significant others:

The interviewees will be asked about their relationship to the family and their awareness of the child's illness. The interviews will focus on their role in decision making, their previous experience of having assisted in making decisions for the child, the goals of care for the child and their anticipation of the child's future.

Appendix 5: Distress protocol



Appendix 6: Participant Information Sheets

Participant Information Sheet – Child 7-10 years old

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Participant information sheet (Child 7-10yrs)
Final Version1.1

The MAPS Study

Decision making and future planning for children with complex illness.

Hello. My name is Emma – this is me ↑ and I am a children's nurse. My supervisors are called Bridget and Kristian! We would like you to help us with a project we are doing.

We want to find out how families of children who get ill a lot decide what to do when they become unwell.

Why is this project being done?

We want to find out your feelings about being ill and how you help your parents and the doctors decide what treatments you should have when you are ill.

Why me?

You have been chosen because you get ill sometimes. With your help we can find out how children help their parents and doctors decide what treatments to have when they are ill.

What will happen to me if I take part?

- We will come to your hospital appointments to see what happens. The appointments may be audio recorded if you agree.
- We will speak to you a few times for about 20 minutes. Your parents can be in the room if you would like them to be. Our talk may take place in your home or at the hospital or clinic. We will ask you questions about your feelings and experiences. If you agree, this will be recorded on a tape recorder.
- You can do drawings or show us pictures to tell us how you feel.

If you would like to help with this project you will be asked to say that you have agreed to take part and that you are happy to tell us what it is like being ill and how you help your parents decide on treatments. We will also talk to your parents and other people who help make decisions about your care.

Do I have to take part?

No. Not if you don't want to. Nobody will mind.

You can stop talking at any point and you do not have to tell us why. You do not have to answer any questions you do not want to.

We will not tell anyone what you say unless it would help to keep you safe.

We will let the doctors and nurses know what we find out about what children think about their care, but we won't use your name.

We hope the information you give us will help families of other children who are ill.

What if I am unhappy about something to do with the project?

If you feel unhappy about anything to do with the project, please discuss it with your parents and they will be able to find out if the researchers can improve things next time.

What do I do now?

You need to think about whether you would like to take part or not. We will need your agreement for you and your family to take part in this project. Please read this leaflet, talk to your parents and let them know if you have any questions or if you would like to talk to us.

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Participant information sheet (11-15yrs)
Final Version1.1

The MAPS Study



Decision making and future planning for children with complex illness.

Name of Researcher(s): Emma Popejoy, Professor Bridget Johnston, Dr Kristian Pollock

We would like to invite you to take part in a research study. Before you decide, we would like you to understand why the research is being done and what it would involve for you. We will go through the information sheet with you and answer any questions you have. Talk to others about the study if you wish. Please do ask us if anything is not clear.



What is the purpose of the study?

The research aims to find out how children and young people feel about being ill and how, together with their parents and doctors, they help make decisions about their care. Knowing more about this will help doctors and nurses provide better support to families and children in the future.

Why have I been chosen?

You have been invited to take part in this study because you have a complex illness. We are interested in what you feel about your illness and how it is treated.

Do I have to take part?

No. It is up to you to decide if you want to take part in the study. You can stop at any time and without giving a reason. You do not have to answer questions or speak to the

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researcher unless you want to. Leaving the study will not affect your care and no-one will be upset with you. Just tell your parents and the researcher if you want to do this. If you do wish to take part, we will need your agreement for you and/or your family to take part in this study.

What will I have to do?

If you decide you would like to take part, a researcher (Emma Popejoy - a Registered Children's nurse) will be present at your hospital appointments for a year and during ward rounds if you go into hospital, always with your agreement: the researcher will not attend these appointments if you do not want her to. Clinic appointments may be audio recorded if everyone present agrees. If you, or anyone else present, does not agree to this, the appointment will not be recorded. The researcher will still be present to observe the appointment though, if everyone agrees. You will also be invited to speak to the researcher three times in the year. These meetings will usually take place in your home, or can take place in the hospital or clinic and last for between 30-60 minutes. The interviews can take place on your own, or with someone else if you wish. They will be about your experience of helping to make decisions about your care, the people involved in making these decisions and whether the decisions have changed.

If you do decide to take part you will keep this information sheet and be asked to sign a form or to say that you have agreed to take part and understand what it involves. Your parents will also have to agree.

Who will know that I took part in the study?

Your parents and the doctors and nurses who look after you when you are ill will know that you are taking part in the study. We will not use your name in any reports of the study, and no one else would be able to know who you are.

What you tell us will be kept private, and no one else will know what you said. The only exception would be if you told us something that indicates that you, or someone else, was

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at risk of harm. In that case we may need to tell your parents or doctors so that they can keep you safe.

What if I am unhappy about something related to the study?

If you feel unhappy about anything related to the study, please discuss it with your parents and they will be able to find out if the researchers can improve things next time.

What will happen to the information we collect?

We will write a report to let people know the results of the study. Your words may be used in this but your name will not, so no one will know that you took part. All participants will be sent a summary of the findings and recommendations at the end of the study if they would like to receive this.

Further information and contact details

Emma Popejoy, Researcher
Research Student
School of Health Sciences
University of Nottingham
Room B33, Queen's Medical Centre
Nottingham NG7 2HA
Email: ntxep3@nottingham.ac.uk



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Participant information sheet (16-18yrs)
Final Version1.1

The MAPS Study



Decision making and future planning for children with complex illnesses: a multi-stakeholder, longitudinal study.

Name of Researchers: Emma Popejoy, Professor Bridget Johnston, Dr Kristian Pollock

We would like to invite you to take part in a research study. Before you decide, we would like you to understand why the research is being done and what it would involve for you. We will go through the information sheet with you and answer any questions you have. Talk to others about the study if you wish. Please do ask if there is anything that is not clear.

What is the purpose of the study?

The research aims to find out how children and young people experience being ill and their role in making decisions about their treatment, so that services can be improved. It also aims to identify how young people, their families, 'significant others' and professionals make decisions about the care they are to receive and how they plan for their future care. We are interested in finding out how these decisions are made initially, whether the decisions change over time and whether there are any benefits or problems associated with planning for the future. We hope that getting a better understanding of how people experience making decisions and future planning will help improve care for young people and their families, in the future.

Why have I been chosen?

You have been invited to take part in this study because you have a complex illness. We are interested in your experience and are hoping to involve up to 15 children and young people in this study in total.

Do I have to take part?

No. 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 form to say that you have agreed to take part in the study, and understand what this involves. You can stop taking part

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in the study at any time and without giving a reason. This would not affect your care or legal rights.

What will I have to do?

If you decide you would like to take part, a researcher (Emma Popejoy - a Registered Children's nurse) will be present at your hospital appointments over the course of 12 months and also during ward rounds if you go into hospital. Clinic appointments may be audio recorded if you and others present permit this. If you, or anyone else present does not agree to this, the appointment will not be recorded. The researcher will still be present to observe the appointment though, always providing everyone agrees. You will also be invited to be interviewed around three times during those 12 months. The interviews can take place on your own, or with someone else if you wish. They may take place in your home, or another convenient place of your choice and will last no longer than an hour. With your permission, the interviews will be recorded on a tape recorder. The interviews will be about your experience of making or participating in health care decisions and planning for your future. They will also cover topics such as who else is involved in making decisions, whether these decisions have changed at all, the information needed to make decisions and thoughts about planning for the future. We will also ask for your permission to access your medical records to see how decisions about your care are documented.

So we can get to know more about you we would like to talk to people who are important to you (like your parents, doctors or friends). We would like to know how they are involved in supporting you and making decisions about your care. We will ask your permission to talk to these people and we will not talk to anyone without your agreement.

Will my taking part in the study be kept confidential?

Yes. What you tell us will be kept confidential and no one except the researchers will know what you say in the interviews. The only exception would be if you tell us something that indicates that you, or anyone else, are at risk of harm. In that case, we may need to tell somebody else to keep you safe, but we would talk to you about this first.

What are the possible disadvantages and risks of taking part?

We do not think that there are any risks involved in taking part in this study. We hope that you will find taking part to be an interesting experience. It is possible that discussing some topics might make you feel sad or upset. If you become upset at any time during a visit we will ask you whether you want to stop the conversation or have a break.

What are the benefits of taking part?

We cannot promise the project will help you but we hope this study will improve our understanding of how young people and their families make decisions about their care and their future.

What if there is a problem?

If you have a concern about any aspect of this project, please speak to the researcher (Emma Popejoy) or ask to speak to the Chief Investigator (Bridget Johnston). We will do our best to answer your questions (contact details are given at the end of this information sheet). If you remain unhappy and wish to complain formally, you can contact the Patient Advice and Liaison Service at your local hospital:

What will happen to the information we collect?

We will write a report of the findings which will be available for anyone to read. Your words may be used in this, but your name will not, so no one will know that you took part. All participants will be sent a summary of the findings and recommendations at the end of the study if they would like to receive this.

Further information and contact details

If you wish to take part or would like to find out more about the study, please contact the researchers directly (using the contact details below) or use the reply form and prepaid envelope included with this information sheet, within 30 days of receiving this letter.

Emma Popejoy, Researcher
School of Health Sciences
University of Nottingham
Room B33, Queen's Medical Centre
Nottingham NG7 2HA
Email: ntxep3@nottingham.ac.uk



Professor Bridget Johnston
School of Health Sciences
University of Nottingham
Room B61a/b, Queen's Medical Centre
Nottingham NG7 2HA
Tel: 0115 8231201
Email bridget.johnston@nottingham.ac.uk

Dr Kristian Pollock
School of Health Sciences
University of Nottingham
Room B61a/b, Queen's Medical Centre
Nottingham NG7 2HA
Tel: 0115 8231201/0115 8230810
Email: Kristian.pollock@nottingham.ac.uk

Participant information sheet (Parent)
Final Version 1.1

The MAPS Study



Decision making and future planning for children with complex illnesses: a multi-stakeholder, longitudinal study.

Name of Researcher(s): Emma Popejoy, Professor Bridget Johnston, Dr Kristian Pollock

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. Please do ask us if there is anything that is not clear.

What is the purpose of the study?

The purpose of the study is to find out how children with complex illnesses, their families, 'significant others' and professionals make decisions about the care they are to receive and how they plan for their future care. We are interested in finding out how these decisions are made initially, whether the decisions change over time and the perceived benefits or problems associated with planning for the future. We hope that getting a better understanding of the experiences of these individuals in decision making and future planning will help improve care for this population in the future.

Why have I been invited?

You have been invited to take part in this study because you have a child who has been diagnosed with a complex illness. A health professional that cares for your child has identified you as someone who might be interested in taking part in the study.

Do I have to take part?

No. 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 to say that you have agreed to take part in the study, and understand what this involves. 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 or your child's medical care or legal rights.

What will happen to me if I take part?

If you decide you would like to take part, you will be invited to remain in the study for follow up over the course of a year. If you agree, a researcher (Emma Popejoy - a Registered Children's nurse) will be present at your child's clinic appointments during this period and also during ward rounds if your child is admitted to hospital during the study period. The researcher will only be present at such occasions with your agreement, and you can decide at any point that you or your child would prefer the researcher to leave. Clinic appointments may be audio recorded if permitted by those present.

Following periods of observation, you will be invited to take part in an interview, where you will be invited to talk about your involvement in discussions about your child's care. The interviews will be about your previous experience of having made health care decisions for your child or plans for their future and the outcomes of these decisions. The interviews will also focus on who was involved in making these decisions, how you feel about the decisions made and how you feel about planning for your child's future care. During the study period, you will be invited to participate in approximately three interviews. The interviews can take place on your own, or with someone else if you wish. They can take place in your own home, or in another location of your choice, such as at the Hospital or the University of Nottingham. With your permission, the interviews will be recorded using a digital audio recorder. If you decide you would like to take part, or would like further information about the study please return the reply slip in the envelope provided or contact the researcher, Emma Popejoy, directly using the contact details below.

You will also be invited to identify a health professional and any additional 'significant others' (for example, extended family members, religious leader, friends etc.) who are important in your decision making, who we could ask to take part in interviews to discuss their involvement in your child's care. We will also ask for your permission to access your child's medical records at the end of the study to see how decisions about their care have been documented and to periodically access their electronic medical records to identify dates of clinic appointments and any episodes of hospitalisation. You can still take part in the study even if you don't want to agree to either or both of these requests.

Will my child be asked to participate?

If your child is aged between 7 and 15 years and is able to understand what the study is about, we will ask your permission to invite your child to participate. If your child is willing and you also agree to them taking part, the researcher will invite them to talk about their experience of illness and how they are involved in making decisions about treatment. They will also be interviewed approximately three times during the study period. If you do not wish for your child to be involved, or your child does not agree to take part, you may still participate in the research if you would like to do so. If your child is aged 16 or over, they will be asked to decide for themselves if they wish to take part in the study and if they are happy for you and others to be involved in discussion of their care.

What are the possible disadvantages and risks of taking part?

We realise that the topic we are studying can be difficult to discuss. We ask you to consider very carefully how you would feel about sharing your experience of your child's illness and care with the researcher. It is possible that you may feel upset at some time during the interview. However, you will never be under any pressure to answer questions or talk about topics that you prefer not to discuss. The interviews will focus on your experience and what was important to you. You can stop or take a break at any time. If you feel this would be helpful, the researcher can give you information about how to access local sources of support.

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What are the possible benefits to taking part in the study?

We cannot promise the study will help you or your child directly but the information we get from this study will help health professionals to understand the experiences of families and other professionals in making difficult health care decisions for their child. This should help to improve the care and support provided to families in future. Some people find it helpful to have the opportunity to reflect on, and share, difficult experiences with someone who has not been directly involved in providing care.

What if there is a problem?

If you have a concern about any aspect of this project, please speak to the researcher (Emma Popejoy) or ask to speak to the Chief Investigator (Bridget Johnston). We will do our best to answer your questions (contact details are given at the end of this information sheet). If you remain unhappy and wish to complain formally, you can contact the Patient Advice and Liaison Service at your local hospital:

Will my taking part in the study be kept confidential?

Yes. We will follow ethical and legal practice and all information about you will be handled in confidence, in accordance with the Data Protection Act, 1998.

All interview data (audio and video recordings and interview transcripts) will be anonymised and kept **strictly confidential**, stored in a secure and locked office, and on a password protected database. Only members of the research team will have access to the information you provide for the study. Any information about you which leaves the university will have

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your name and address removed (anonymised) and a unique code will be used so that you cannot be recognised from it.

Your personal data (name, address, telephone number) will be kept for 3 – 6 months after the end of the study so that we are able to contact you about the findings of the study if you have told us that you would like to receive these. All other data (research data) will be kept securely for 7 years before being disposed of securely. During this time all precautions will be taken by all those involved to maintain your confidentiality.

The only circumstances under which confidentiality would be broken would arise if the researchers were made aware of actions or situations resulting in serious risk of harm to yourself or others. The researchers would discuss this with you and consider the need to raise the matter with an appropriate person.

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 or your child's medical care being affected. If you withdraw we will ask your permission for the data already collected about your child to be retained for analysis. However, if you wish, this data will be excluded from the study.

What will happen to the results of the research study?

We will not know the results of this project until after the last participant has received their final visit, which will be around August 2016.

This project will form part of a thesis for a higher degree and will be reported in publications in medical journals and presented at various conferences. None of the participants will be identified by name in any reporting of the findings.

The results of the study will also be fed back to the service to ensure professionals are aware of families' experiences. All participants will be sent a summary of the findings and recommendations at the end of the study if they would like to receive this.

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Who has reviewed the study?

All research in the NHS is looked at by independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed and given favourable opinion by the Nottingham 1 Research Ethics Committee.

What do I have to do?

If you wish to take part or would like to find out more about the study, please contact the researchers directly or use the reply form and prepaid envelope included with this information sheet, within 30 days of receiving this letter. They will contact you directly to discuss this. You do not have to respond if you do not want to take part. However, it would be very helpful if you could indicate this by signing and returning the reply slip: you do not need to include any additional contact details. You may also contact the researchers directly by phone or email using the contact details given below.

Further information and contact details are given overleaf.

If you decide you would like to take part or want further information please return the reply slip in the envelope provided. Please contact us using the details below if you have any questions.

Emma Popejoy, Researcher
School of Health Sciences
University of Nottingham
Room B33, Queen's Medical Centre
Nottingham NG7 2HA
Email: ntxep3@nottingham.ac.uk

Dr Kristian Pollock
School of Health Sciences
University of Nottingham
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Tel: 0115 8231201/0115 8230810
Email: Kristian.pollock@nottingham.ac.uk

Professor Bridget Johnston
School of Health Sciences
University of Nottingham
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Tel: 0115 8231201
Email bridget.johnston@nottingham.ac.uk

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Participant information sheet (Parent – Child Participation)
Final Version1.1

The MAPS Study



Decision making and future planning for children with complex illnesses: a multi-stakeholder, longitudinal study.

Name of Researcher(s): Emma Popejoy, Professor Bridget Johnston, Dr Kristian Pollock

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. Please do ask us if there is anything that is not clear.

What is the purpose of the study?

The purpose of the study is to find out how children with complex illnesses, their families, 'significant others' and professionals make decisions about the care they are to receive and how they plan for their future care. We are interested in finding out how these decisions are made initially, whether the decisions change over time and the perceived benefits or problems associated with planning for the future. We hope that getting a better understanding of the experiences of these individuals in decision making and future planning will help improve care for this population in the future.

Why has my chid been invited?

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You child has been invited to take part in this study because they have been diagnosed with a complex illness. A health professional that cares for your child has identified that they may be interested in taking part in the study.

If your child is aged between 7 and 15 years and is able to understand what the study is about, we will ask your permission to invite your child to participate. If your child is willing and you also agree to them taking part, the researcher will invite them to talk about their experience of illness and how they are involved in making decisions about treatment. They will also be interviewed approximately three times during the study period. If you do not wish for your child to be involved, or your child does not agree to take part, you may still participate in the research if you would like to do so. If your child is aged 16 or over, they will be asked to decide for themselves if they wish to take part in the study and if they are happy for you and others to be involved in discussion of their care.

Does my child have to take part?

No. You and your child can decide whether or not they take part. If they do decide to take part you will be given this information sheet to keep and be asked to sign a consent form to say that you have given permission for your child to take part in the study, and understand what this involves. They will also be asked to sign or say that agree to participate. If your child decides to take part, they are still free to withdraw at any time and without giving a reason. This would not affect your or your child's medical care or legal rights.

What will happen to my child if they take part?

If your child decides to take part, you will be invited to remain in the study for follow up over the course of a year. A researcher (Emma Popejoy - a Registered Children's nurse) will be present at your child's clinic appointments during this period and also during ward rounds if your child is admitted to hospital during the study period. The researcher will only be present at such occasions with your agreement, and you can decide at any point that you or your child would prefer the researcher to leave. Clinic appointments may be audio recorded if permitted by those present.

Following periods of observation, your child will be invited to take part in an interview, where they will be invited to talk about their illness, how they help make decisions about their care and their goals for the future. The interviews will only cover topics that your child is comfortable with and discuss their illness only in terms of what they understand. During the study period, your child will be invited to participate in approximately three interviews. The interviews can take place on their own, or with you if they wish. They can take place in your own home, or in another location of your choice, such as at the Hospital or the University of Nottingham. With your and your child's permission, the interviews will be recorded using a digital audio recorder.

We will also ask for your permission to access your child's medical records at the end of the study to see how decisions about their care have been documented and to periodically access their electronic medical records to identify dates of clinic appointments and any episodes of hospitalisation. Your child can still take part in the study even if you don't want to agree to either or both of these requests.

What are the possible disadvantages and risks of taking part?

We realise that the topic we are studying can be difficult to discuss. We ask you to consider very carefully how your child would feel about sharing their experience of their illness and care with the researcher. It is possible that they may feel upset at some time during the interview. However, they will never be under any pressure to answer questions or talk about topics that they prefer not to discuss. They will be allowed to stop or take a break at any time.

What are the possible benefits to taking part in the study?

We cannot promise the study will help you or your child directly but the information we get from this study will help health professionals to understand the experiences of families and other professionals in making difficult health care decisions for their child. This should help to improve the care and support provided to families in future.

What if there is a problem?

If you have a concern about any aspect of this project, please speak to the researcher (Emma Popejoy) or ask to speak to the Chief Investigator (Bridget Johnston). We will do our best to answer your questions (contact details are given at the end of this information sheet). If you remain unhappy and wish to complain formally, you can contact the Patient Advice and Liaison Service at your local hospital:

Will my child's taking part in the study be kept confidential?

Yes. We will follow ethical and legal practice and all information about you will be handled in confidence, in accordance with the Data Protection Act, 1998.

All interview data (audio and video recordings and interview transcripts) will be anonymised and kept **strictly confidential**, stored in a secure and locked office, and on a password protected database. Only members of the research team will have access to the information your child provides for the study. Any information about your child which leaves the university will have your name and address removed (anonymised) and a unique code will be used so that they cannot be recognised from it.

Your child's personal data (name, address, telephone number) will be kept for 3 – 6 months after the end of the study so that we are able to contact you about the findings of the study if you have told us that you would like to receive these. All other data (research data) will be kept securely for 7 years before being disposed of securely. During this time all precautions will be taken by all those involved to maintain your confidentiality.

The only circumstances under which confidentiality would be broken would arise if the researchers were made aware of actions or situations resulting in serious risk of harm to

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your child or others. The researchers would discuss this with you and consider the need to raise the matter with an appropriate person.

What will happen if my child doesn't want to carry on with the study?

Your child's participation is voluntary and they are free to withdraw at any time, without giving any reason, and without their legal rights or medical care being affected. If they withdraw we will ask your permission for the data already collected to be retained for analysis. However, if you wish, this data will be excluded from the study.

What will happen to the results of the research study?

We will not know the results of this project until after the last participant has received their final visit, which will be around August 2016.

This project will form part of a thesis for a higher degree and will be reported in publications in medical journals and presented at various conferences. None of the participants will be identified by name in any reporting of the findings.

The results of the study will also be fed back to the service to ensure professionals are aware of families' experiences. All participants will be sent a summary of the findings and recommendations at the end of the study if they would like to receive this.

Who has reviewed the study?

All research in the NHS is looked at by independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed and given favourable opinion by the Nottingham 1 Research Ethics Committee.

What do I have to do?

If you wish to take part or would like to find out more about the study, please contact the researchers directly or use the reply form and prepaid envelope included with this information sheet, within 30 days of receiving this letter. Please inform the researcher if you are happy for them to invite your child to participate in the interview component of the

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study. The researcher will then discuss the study with your child and ask them if they wish to take part.

Further information and contact details are given overleaf.

Please contact us using the details below if you have any questions.

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Participant information sheet (Professional)
Final Version1.1

The MAPS Study



Decision making and future planning for children with complex illnesses: a multi-stakeholder, longitudinal study.

Name of Researcher(s): Emma Popejoy, Professor Bridget Johnston, Dr Kristian Pollock

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. Please do ask us if there is anything that is not clear.

What is the purpose of the study?

The purpose of the study is to find out how children with complex illnesses, their families, 'significant others' and health care professionals make decisions about the care the child is to receive and plan for the future. We are interested in finding out how these decisions are made initially, whether the decisions change over time and the perceived benefits or problems associated with future planning. We hope that getting a better understanding of the experiences of these individuals in making decisions and future planning will help improve care for this population in the future.

Why have I been invited?

You have been invited to take part in this study because you are a professional caring for a child who has been diagnosed with a life-limiting illness. The parent or child has identified you as a professional who is important in their decision making and future planning and as someone who might be interested in taking part in the study.

Do I have to take part?

No. 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 to say that you have agreed to take part in the study, and understand what this involves. If you decide

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to take part you are still free to withdraw at any time and without giving a reason. This would not affect your legal rights or career development.

What will happen to me if I take part?

If you decide you would like to take part, you will be invited to participate in several ways over the course of a year. A researcher (Emma Popejoy - a Registered Children's nurse) will be present at the child's clinic appointments during this period and also during ward rounds if the child is admitted to hospital. Clinic appointments may be audio recorded if permitted by those present. During the study period, you will be invited to participate in approximately three interviews. These can take place in your office, your own home, or in another private location of your choice. With your permission, the interviews will be recorded using a digital audio recorder.

The interviews will be about the child and their illness, their family context, goals of care and their prognosis. They will also cover the important decisions that the family have had to consider and the likely decisions that they may need to consider in the future. They will also seek to identify how discussions about decision making and future planning are initiated and undertaken with this family.

What are the possible disadvantages and risks of taking part?

We realise that the topic we are studying is emotive. We ask you to consider very carefully how you would feel about sharing your experience with the researcher. It is possible that you may feel upset at some time during the interview. However, you will never be under any pressure to answer questions or talk about topics that you prefer not to discuss. The interviews will focus on your experience and what was important to you. You can stop or take a break at any time. Counselling support after the interview can be accessed through your Trust's Occupational Health Service.

What are the possible benefits to taking part in the study?

We cannot promise the study will benefit you directly but the information we gather may help provide a better understanding of how people experience health care decision making for children with life-limiting illnesses. It may also help to identify the support needs of professionals and families in having these discussions and making decisions. This information should help to improve the care and support provided to families in future.

What if there is a problem?

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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 researchers' contact details are given at the end of this information sheet. If you remain unhappy and wish to complain formally, you can contact the Ethics Committee Secretary, Mrs Louise Sabir, UoN Faculty of Medicine & Health Sciences Research Ethics Committee, c/o School of Medicine Education Centre (Medical Courses Office), B Floor, Medical School, QMC Campus, Nottingham University Hospitals, NG7 2UH. E-mail: louise.sabir@nottingham.ac.uk.

Will my taking part in the study be kept confidential?

Yes. We will follow ethical and legal practice and all information about you will be handled in confidence, in accordance with the Data Protection Act, 1998.

All interview and observation data (audio recordings and interview transcripts) will be anonymised and kept **strictly confidential**, stored in a secure and locked office, and on a password protected database. Any information about you which leaves the university 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 3 – 6 months after the end of the study so that we are able to contact you about the findings of the study if you have told us that you would like to receive these. 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.

The only circumstances under which confidentiality would be broken would arise if the researchers were made aware of actions or situations resulting in serious risk of harm to yourself or others. The researchers would discuss this with you and consider the need to raise the matter with an appropriate person.

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.

What will happen to the results of the research study?

We will not know the results of this project until after the last participant has received their final visit, which will be around August 2016.

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This project will form part of a thesis for a higher degree and will be reported in publications in nursing/medical journals/presented at various conferences. None of the participants will be identified by name in any reporting of the findings.

The results of the study will also be fed back to the service to ensure professionals are aware of families' experiences and the issues involved in discussions about decision making and future planning. All participants will be sent a summary of the findings and recommendations at the end of the study if they would like to receive this.

Who has reviewed the study?

All research in the NHS is looked at by independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed and given favourable opinion by the Nottingham 1 Research Ethics Committee.

What do I have to do?

If you wish to take part or would like to find out more about the study, please contact the researchers directly or use the reply form and prepaid envelope included with this information sheet, within 30 days of receiving this letter. They will contact you directly to discuss this. You do not have to respond if you do not want to take part. However, it would be very helpful if you could indicate this by signing and returning the reply slip: you do not need to include any additional contact details. You may also contact the researchers directly by phone or email using the contact details given below.

Further information and contact details: If you decide you would like to take part or want further information please return the reply slip in the envelope provided. Please contact us using the details below if you have any questions.

Emma Popejoy, Researcher
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Dr Kristian Pollock
School of Health Sciences
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Participant information sheet (Significant Other)

Final Version 1.1

The MAPS Study



Decision making and future planning for children with complex illnesses: a multi-stakeholder, longitudinal study.

Name of Researcher(s): Emma Popejoy, Professor Bridget Johnston, Dr Kristian Pollock

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. Please do ask us if there is anything that is not clear.

What is the purpose of the study?

The purpose of the study is to find out how children with complex illnesses, their families, 'significant others' and professionals make decisions about the care the child is to receive and plan for the future. We are interested in finding out how these decisions are made initially, whether the decisions change over time and the perceived benefits or problems associated with future planning. We hope that getting a better understanding of the experiences of these individuals in making decisions and future planning will help improve care for this population in the future.

Why have I been invited?

You have been invited to take part in this study because you are a significant person in the life of a child who has a complex illness. The parent or child has identified you as someone who is important in helping them make decisions and as someone who might be interested in taking part in the study.

Do I have to take part?

No. 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 to say that you have agreed to take part in the study, and understand what this involves. 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.

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What will happen to me if I take part?

If you decide you would like to take part, you will be invited to participate in the study in a variety of ways over the course of a year. You will be invited to take part in approximately three interviews during the yearlong study period. Each interview is likely to take approximately an hour and can take place on your own or with a member of the child's family if you wish. These can take place in your own home, or in another location of your choice. With your permission, the interviews will be recorded using a digital audio recorder. A researcher (Emma Popejoy - a Registered Children's nurse) will also be present to observe medical care and decision making at the child's clinic appointments during this period and also during ward rounds if the child is admitted to hospital. You may be present on some of these occasions. Clinic appointments may be audio recorded if permitted by those present.

The interviews will be about your relationship to the family, your experience of assisting them in making health care decisions for the child and helping them to plan for the future. The interviews will focus on your role in decision making, your previous experience of having assisted in making decisions for the child, the goals of care for the child and your anticipation of the child's future care. If you decide you would like to take part or want further information please return the reply slip in the envelope provided.

What are the possible disadvantages and risks of taking part?

We realise that the topic we are studying is emotive. We ask you to consider very carefully how you would feel about sharing your experience with the researcher. It is possible that you may feel upset at some time during the interview. However, you will never be under any pressure to answer questions or talk about topics that you prefer not to discuss. The interviews will focus on your experience and what was important to you. You can stop or take a break at any time. If you feel this would be helpful, the researcher can give you information about how to access local sources of support.

What are the possible benefits to taking part in the study?

We cannot promise the study will help you but the information we gather may help health professionals to have a better understanding of how people experience health care decision making for children with complex illnesses and how they can provide better support for this. This should help to improve the care and support provided to families in future. Some people find it helpful to have the opportunity to reflect on, and share, difficult experiences with someone who has not been directly involved in providing care.

What if there is a problem?

If you have a concern about any aspect of this project, please speak to the researcher or ask to speak to the Chief Investigator. We will do our best to answer your questions (contact details are given at the end of this information sheet).

If you remain unhappy and wish to complain formally, you can contact the Ethics Committee Secretary, Mrs Louise Sabir, UoN Faculty of Medicine & Health Sciences Research Ethics Committee, c/o School of Medicine Education Centre (Medical Courses Office), B Floor, Medical School, QMC Campus, Nottingham University Hospitals, NG7 2UH.
E-mail: louise.sabir@nottingham.ac.uk.

Will my taking part in the study be kept confidential?

Yes. We will follow ethical and legal practice and all information about you will be handled in confidence, in accordance with the Data Protection Act, 1998.

All interview and observation data (audio recordings and interview transcripts) will be anonymised and kept **strictly confidential**, stored in a secure and locked office, and on a password protected database. Any information about you which leaves the university 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 3 – 6 months after the end of the study so that we are able to contact you about the findings of the study if you have told us that you would like to receive these. All other data (research data) will be kept securely for 7 years. After this time your data will be disposed of securely. 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.

The only circumstances under which confidentiality would be broken would arise if the researchers were made aware of actions or situations resulting in serious risk of harm to yourself or others. The researchers would discuss this with you and consider the need to raise the matter with an appropriate person.

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 we will ask your permission for the data already collected to be retained for analysis. However, if you wish, this data will be excluded from the study.

What will happen to the results of the research study?

We will not know the results of this project until after the last participant has received their final visit, which will be around August 2016.

This project will form part of a thesis for a higher degree and will be reported in publications in nursing and medical journals and presented at various conferences. None of the participants will be identified by name in any reporting of the findings.

The results of the study will also be fed back to the health service to ensure professionals are aware of families' experiences. All participants will be sent a summary of the findings and recommendations at the end of the study if they would like to receive this.

Who has reviewed the study?

All research in the NHS is looked at by independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed and given favourable opinion by the Nottingham 1 Research Ethics Committee.

What do I have to do?

If you wish to take part or would like to find out more about the study, please contact the researchers directly or use the reply form and prepaid envelope included with this information sheet, within 30 days of receiving this letter. They will contact you directly to discuss this. You do not have to respond if you do not want to take part. However, it would be very helpful if you could indicate this by signing and returning the reply slip: you do not need to include any additional contact details. You may also contact the researchers directly by phone or email using the contact details given below.

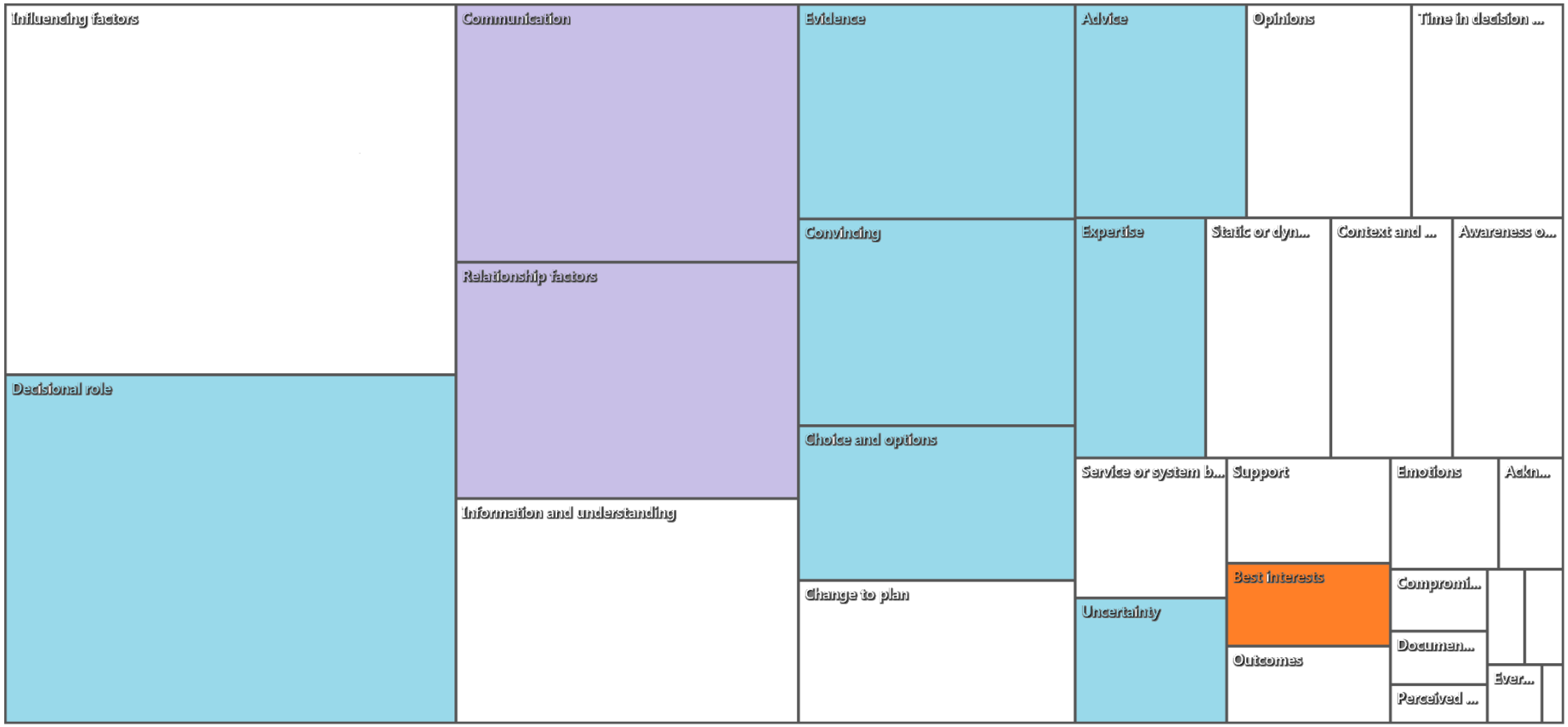
Further information and contact details: If you decide you would like to take part or want further information please return the reply slip in the envelope provided. Please contact us using the details below if you have any questions.

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Appendix 7: Coding hierarchy and explanation



This coding hierarchy exported from the NVivo 11 dataset demonstrates how the themes which are presented in the findings chapters were arrived at. This hierarchy chart has been colour coded to identify the emergence of the themes. Lilac codes resulted in the core theme of 'Communication and Relationships', blue codes resulted in the core theme of 'The process of decision making' and the orange code resulted in the core theme of 'Best interests'. Other major codes, such as 'influencing factors' and 'information and understanding' ran as key strands throughout all of the core themes and have been incorporated into all three chapters, where relevant. Other less frequently identified codes have either been incorporated similarly, for example 'opinions', or were identified as being less pertinent to the process of decision making and therefore have not been featured within this thesis. Although the core theme of 'Best interests' did not feature heavily in this coding hierarchy, it was a significant part of the narrative in a number of the cases and therefore deemed essential to include within this thesis.

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