Comparative outcomes for children with and without access to cochlear implantation

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ABSTRACT

Comparative outcomes for children with and without access to cochlear implantation

There is uncertainty regarding whether hearing aids or cochlear implants should be recommended for children with severe hearing loss. Comparing the clinical effectiveness of these devices can inform the appropriate management of severe hearing loss in children. UK National Health Service guidance on cochlear implant candidacy was updated in 2019 and did not directly align with any published evidence. This thesis therefore aimed to compare the outcomes of children with hearing loss close to or within the revised NICE candidacy criteria who used hearing aids to the outcomes of children using cochlear implants, while controlling for potentially confounding variables.

A scoping review was conducted of comparative studies of children with severe hearing loss using hearing aids to children using cochlear implants, cataloguing the sparse evidence regarding speech perception and language outcomes. Gaps in the literature were found for comparative studies of outcomes such as tinnitus and listening fatigue. A cross-sectional cohort of children using hearing aids was then characterised. This revealed that the 2019 NICE guidance both widened access to cochlear implantation and increased the number of children for whom eligibility could not be determined based on existing clinical records. Caregiver-reported aural/oral, tinnitus, and listening fatigue outcomes for children with severe or profound hearing loss using hearing aids were compared to those for children using cochlear implants. Children using hearing aids had poorer aural/oral outcomes than children with cochlear implants, apart from those who could be confirmed to fall outside the 2019 NICE guidance. Eligibility screening data were linked to data from the Born in Bradford cohort study, identifying poorer early educational outcomes for children with hearing loss compared to matched controls.

This thesis adds to the evidence base on cochlear implant candidacy via novel findings on children's access to cochlear implantation and the comparative clinical effectiveness of hearing aids and cochlear implants. Incomplete audiometric data, younger age, and additional needs were associated with unknown eligibility for cochlear implantation among children using hearing aids. Children using hearing aids who had previously been audiometrically-eligible for cochlear implants had the highest proportion of families who needed interpreters. Both children newly-eligible for cochlear implantation, and at least some of those of unknown eligibility, might gain better aural/oral outcomes from cochlear implants than from hearing aids. These findings tentatively support the revised NICE guidance, but further research is needed across a wider range of outcomes including direct child reports and behavioural assessments. Linkage to the Born in Bradford cohort data found that standard care was not sufficient for children with hearing loss to achieve the same educational attainment as their typically-hearing peers.

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DECLARATION

I certify that this is my own work, except where indicated by referencing. No part of this thesis has been submitted elsewhere for any other degree or qualification.

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ABBREVIATIONS

ABR	Auditory Brainstem Response
ACIA	American Cochlear Implant Alliance
ANSI	American National Standards Institute
ANSD	Auditory neuropathy spectrum disorder
APP	Auditory Perceptive Performance
ASHA	American Speech Language Hearing Association
ATT	Automated Toy Discrimination Test
AVT	Auditory verbal therapy
BAA	British Academy of Audiology
BCIG	British Cochlear Implant Group
BiB	Born in Bradford
BiCI	Bilateral cochlear implant
Bim	Bimodal
BM	Basilar membrane
BRC	Biomedical Research Centre
BSA	British Society of Audiology
BSL	British Sign Language
CAP	Categories of Auditory Performance
CAPT	Chear Auditory Perception Test
CASL	Comprehensive Assessment of Spoken Language Test
CCT	Consonant Confusion Task
CELF	Clinical Evaluations of Language Fundamentals Test
ChIP	Children's Implant Profile
CI	Cochlear implant
CINAHL	Cumulative Index to Nursing and Allied Health Literature
CNC	Consonant-Nucleus-Consonant Test
CRIDE	Consortium for Research in Deaf Education
CTOPP	Comprehensive Test of Phonological Processing
CUSP	Comparing oUtcomes for Severely and Profoundly deaf children
dB	Decibel
EAS	Electric-acoustic stimulation
EHCP	Education, and health care plan
EYFSP	Early Years Foundation Stage Profile
FAPCI	Functioning After Pediatric Cochlear Implantation Questionnaire
FDA	Food and Drug Administration
GCSE	General Certificate of Secondary Education
GLD	Good level of development (on the EYFSP)
HA	Hearing aid
HeSPAC	Hebrew Speech Pattern Contrasts

HINT-C	Hearing in Noise Test for Children
HL	Hearing level
HTL	Unaided, air-conduction hearing threshold level
Hz	Hertz
ID code	Identification code
IHR	Institute of Hearing Research
IMD	Index of multiple deprivation
IT-MAIS	Infant and Toddler version of the Meaningful Auditory Integration Scale
IQR	Interquartile range
LOCHI	Longitudinal Outcomes of Children with Hearing Impairment
MAIS	Meaningful Auditory Integration Scale
MPT	Minimal Pairs Test
MSAE	Modern Standard Arabic Expressions Assessment
MTLD	Major Stages of Language Development Test
NDCS	National Deaf Children's Society
NHS	National Health Service
NICE	National Institute for Health and Care Excellence
PBK	Phonetically Balanced Kindergarten Test
PEACH	Parents Evaluation of Aural/Oral Performance of Children
PESP	Perceptual Evaluation of Speech Production
PICOS	Population, Intervention, Comparison, Outcome, and Study type
PLS-4	Preschool Language Scale Fourth Edition
PPVT	Peabody Picture Vocabulary Test
PRISMA	Preferred reporting items for systematic reviews and meta-analyses
PTA	Pure tone audiometry
REELS	Receptive-Expressive Emerging Language Scale
SEN	Special educational needs
SD	Standard deviation
SII	Speech intelligibility index
SNHL	Sensorineural hearing loss
SNR	Signal to noise ratio
TAG	Technology appraisal guidance
U.K.	United Kingdom
UniCI	Unilateral cochlear implant
UniEAS	Unilateral electric-acoustic stimulation
U.S.A.	United States of America
WHO	World Health Organisation
WPPT	Word Pattern Perception Test
YAIS	Yorkshire Auditory Implant Service

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CHAPTER 1: INTRODUCTION

In this thesis I seek to add to the evidence base to guide clinicians when choosing whether to offer acoustic hearing aids (HAs) or cochlear implants (CIs) to children with hearing loss within or close to current National Institute for Health and Care Excellence (NICE) guidance on cochlear implantation (TA566). This chapter introduces background information on hearing loss in children, HAs, and CIs. It summarises the evidence available to support candidacy decisions and explains how children are assessed for CI candidacy in the United Kingdom's National Health Service (NHS). Chapter 2 catalogues previously published studies comparing the outcomes of children with severe hearing loss using HAs to children using CIs. The results identify gaps in the evidence base regarding the comparative clinical effectiveness of HAs versus CIs for children with severe hearing loss. Chapter 3 provides a high-level overview of the experimental study design initially planned to fill some of these knowledge gaps, describes the impact of the COVID-19 pandemic and summarises the final study design. Chapter 4 assesses the impact of the latest NICE guidance, by characterising children using HAs without access to CIs in the UK. The results identify trends that might be influencing children's access to CIs and serve as a reference population against which the sample of participants in the survey described in Chapter 5 can be compared. The caregiver survey created new evidence to support clinical decision-making around the provision of HAs or CIs to children within, and close to but outside, current NHS candidacy guidance. Chapter 6 linked data collected for Chapter 4 with data held by the Born in Bradford cohort study, to measure the impact of

hearing loss of any degree on children's early educational outcomes. Chapter 7 discusses the implications of each of the previous chapters for clinical management and future research.

The World Health Organisation (WHO) defines someone with hearing loss as a person who cannot hear as well as a person with normal hearing, whether in one or both ears, leading to "difficulty in hearing conversation or even loud sounds" (World Health Organization, 2021). There are estimated to be over 45,000 children in England with unilateral or bilateral "permanent deafness" of any degree (CRIDE, 2021). Hearing loss can influence every aspect of life, including children's ability to thrive within their homes and schools (Fiorillo et al., 2017; Harris et al., 2017; Wake, Hughes, Poulakis, et al., 2004), and ultimately to fulfil their potential in the workplace and society (Winn, 2007). HAs and CIs can enable most children with hearing loss to communicate via spoken language. However, there is no consensus regarding which children should be offered HAs or CIs (Vickers, de Raeve, et al., 2016) because current evidence regarding paediatric CI candidacy is insufficient to establish criteria without reference to data from adults and is also inconsistent across studies (Bond, Elston, et al., 2009; J. Leigh et al., 2011; Lovett et al., 2015).

In the NHS, clinicians are expected to follow technology appraisal guidance (TAG) published by NICE when deciding whether to offer cochlear implantation to children with severe to profound deafness (NICE, 2019). The first such guidance was published in 2009 (NICE, 2009). It advised that children could be considered candidates for CIs if they had 'severe to profound deafness', defined at that time by NICE as audiometric thresholds >90 dB HL at 2 & 4 kHz in both ears. Over time, this guidance became out of line with

international guidance and practice, with the effect of excluding children in the UK from accessing CIs where children with similar degrees of hearing loss in other countries could access them (Vickers, de Raeve, et al., 2016). Despite again finding limited evidence upon which to base new criteria for CI candidacy, in 2019 NICE revised their definition of 'severe to profound deafness' to include audiometric thresholds up to and including 80 dB HL, so that children with better hearing could now be considered CI candidates. The new guidance adopted was not taken directly from any published evidence, nor was it the same as used by any other country. It was not known how many children would be affected by the change in guidance, nor what their characteristics would be.

The funded period of this PhD began in April 2018, prior to NICE's publication of the revised guidance, and 1 year and 2 months before the revised guidance was put into practice. This change in guidance for the NHS meant that there was a time-limited opportunity to document the outcomes of children with severe hearing loss using HAs who would shortly become eligible for CIs. Describing the outcomes of these children would provide a baseline against which the future outcomes of similar children with CIs could be compared, and allow comparison with the outcomes of children already using CIs. The primary aim of this thesis was to compare the outcomes of children with severe hearing loss using HAs to children using CIs, to inform future decisions about CI candidature.

This chapter introduces some fundamental aspects of audiology including a description of the properties of sound, an overview of the anatomy and physiology of the ear and a summary of the effects of hearing loss on children.

HA and CI technologies are described including their benefits, risks, and cost-effectiveness, followed by an overview of factors influencing the outcomes of children with hearing loss. Studies giving estimates of the hearing loss associated with CI candidacy are appraised. Examples of international candidacy practice are given. Past and current NICE candidacy guidance for CI are then summarised, along with a description of their implementation to give the clinical context for this PhD. This chapter concludes by clarifying the scope of the PhD and stating its aims and objectives.

1.1. What is sound?

Sounds are travelling pressure waves caused by the vibration of a sound-source. Air molecules around the sound-source experience alternating compression and rarefaction, vibrating back and forth around their mean position. The frequency of the vibrations is measured in cycles per second (Hertz; Hz). Its subjective correlate is pitch, with higher-frequency signals perceived as higher-pitched sounds. The intensity of the pressure variations is subjectively correlated to loudness (Pickles, 1988), with higher intensity vibrations perceived as louder sounds.

1.2. Anatomy and physiology of the ear

The peripheral auditory system comprises a pathway, from the arrival of sound at the outer ear to its transduction into neural signals within the inner ear. Its principal components are illustrated in Figure 1.1.

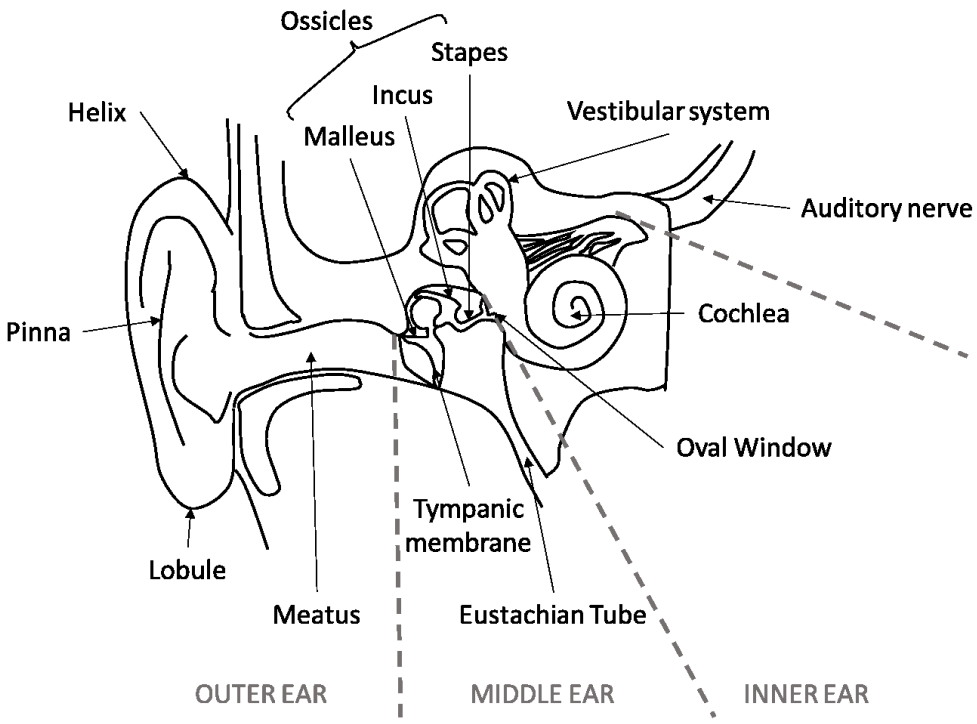


Figure 1.1: Schematic diagram of the ear.

The outer ear comprises the external ear, or pinna, comprising the helix and lobule. The shape of the pinna guides incident sounds to the ear canal, or meatus, the end of which is sealed by the eardrum, or tympanic membrane. The tympanic membrane forms the lateral side of the middle ear, and is thin, cone-shaped, and semi-transparent. Sound waves cause it to vibrate, and the vibrations are conducted through the air-filled middle ear cavity by the ossicles. These small bones couple the tympanic membrane to the oval window of the cochlea (Pickles, 1988).

The inner ear consists of both hearing (cochlear) and balance (vestibular) systems. The cochlea is fluid-filled and comprises three parallel sections, divided by Reissner's membrane and the basilar membrane, coiled in a spiral of two and three-quarter turns. The main function of the cochlea is to transduce mechanical sound vibrations into neural impulses. This is achieved by

movement-induced deflection of the 20 to 30 thousand sensory hair cells in the organ of Corti, which runs along the length of the basilar membrane. The outer hair cells amplify basilar membrane displacement at their place, creating acute sensitivity to soft sounds with a high degree of frequency-specificity (Glasberg & Moore, 2000). The inner hair cells fire in response to the basilar membrane displacement, generating neural signals that accurately represent the pitch, timing and intensity of the incoming sound. The BM responds to different sound frequencies along its length, known as tonotopicity (Greenwood, 1990). Inner hair cells synapse with afferent fibres of the auditory nerve, which connects the cochlea to the brainstem via the internal auditory canal. (Pickles, 1988).

1.3. Hearing loss

As previously stated, hearing loss refers to a person's inability to hear as well as someone with normal hearing (World Health Organization, 2021). Some children have congenital hearing loss. Others may have normal hearing followed by a sudden onset of hearing loss, for example due to infection. This is known as acquired hearing loss. Yet others may have hearing loss that becomes gradually worse over time; this will be referred to as progressive hearing loss. Last, some children may have hearing loss that fluctuates over time; this will be referred to as fluctuating hearing loss. It is possible for more than one type of loss to co-occur in any child.

Hearing losses vary in degree and time-course. Degrees of hearing loss are classified as mild, moderate, severe, or profound by the British Society of Audiology (BSA). These are defined by the average of a person's hearing

threshold levels (HTLs); the softest sound pressure levels at which pure tones (single frequencies) are just detectable compared to normally-hearing adults (BSA, 2018). HTLs are measured in decibels of Hearing Level (dB HL), via tests that will be explained in section 1.4.2. Unless stated otherwise, the BSA classification system is used throughout this thesis (BSA, 2018). Both the onset and degree of hearing loss can differ between right and left ears.

The terms ‘deafness’, or ‘Deaf’, are commonly reserved for people with profound hearing loss who might gain limited benefit from acoustic HAs (World Health Organization, 2021). Those within the Deaf Community may use British Sign Language (BSL) as their main form of communication rather than spoken language. The term ‘hard-of-hearing’ can be used to refer to people with any degree of hearing loss aside from profound.

1.3.1. Sensorineural hearing loss

Most permanent childhood deafness is caused by loss of function within the cochlea and/or hearing nerve. This is described as sensorineural hearing loss (SNHL). SNHL influences sound perception in terms of detection, spectral and temporal resolution, and loudness. Since these influence children’s perception of spoken language, they are described below.

Abnormal outer hair cell function reduces BM motion in response to soft sounds, which become inaudible for the pitches of sound corresponding to the damaged portion of the BM. With increasing inner hair cell dysfunction progressively louder sounds become inaudible, as vibrations fail to be transduced into neural signals (Pickles, 1988).

At the raised intensities needed to stimulate a neural response, inner hair cells will be activated across a wider range of the BM, creating spectrally smeared sound perception. Temporal resolution is also compromised. Sound perception therefore becomes distorted.

SNHL is associated with increased sensitivity to loud sounds. At any frequency, the difference in decibels between the softest detectable sound and the loudest sound that can be tolerated (dynamic range) is reduced for people with SNHL (Pickles, 1988). The effects of SNHL have far-reaching consequences for children. SNHL can impact speech perception (Blamey et al., 2001), language acquisition (Eisenberg, 1982), educational outcomes and employment prospects (Arlinger, 2009), sound localisation (Grieco-Calub & Litovsky, 2012), and quality of life (Wake, Hughes, Collins, et al., 2004).

Up to 15% of children with hearing loss are believed to have auditory neuropathy spectrum disorder (ANSD) (Madden et al., 2002). It is hypothesised that cochlear outer hair cell function is normal in ANSD, but that there are dysfunctions of the cochlear inner hair cells and spiral ganglion neurons (de Siati et al., 2020). Children with ANSD can experience speech perception difficulties disproportionate to their ability to detect sounds (Rance, 2005).

1.3.2. Conductive and mixed hearing loss

Hearing loss can also arise due to impaired conduction of sound to the cochlear through the outer and/or middle ear. Conductive hearing loss in children may be permanent, for example following ear canal atresia (the absence of an opening to the ear canal) or temporary, for example due to otitis media with

effusion (glue ear). Otitis media with effusion is the most commonly occurring cause of conductive hearing loss in childhood, estimated to affect 80% of children on at least one occasion during the first 6 years of life with long-persisting symptoms in around five per-cent of children (Haggard, 1993). Conductive hearing loss results in decreased loudness of sounds and distortion. When sensorineural and conductive losses co-exist, the hearing loss is described as mixed.

1.4. Assessing hearing loss

CIs are intended for people with sensorineural hearing loss of a degree that meets the candidacy guidance applicable in their geographic location. Therefore, to understand the identification of children who might benefit from CIs, it is necessary to understand the methods used to establish both type and degree of hearing loss in children.

1.4.1. Otoscopy and tympanometry

The likelihood of a conductive loss being present can be estimated via visual inspection of the outer ear and eardrum using an otoscope (otoscopy) and by acoustic tests of the middle-ear's admittance of sound (tympanometry). The middle ear most readily admits sound when the air pressure is equal on both sides of the eardrum. During tympanometry the entrance to the ear canal is acoustically sealed with a soft probe tip. A tone is played into the ear canal and its admittance to the middle ear is measured while the air pressure in the ear canal is gradually varied. Test results can be interpreted to reveal the presence

of middle ear effusion, a poorly aerated middle ear, a perforated eardrum or patent grommet, eardrum hypermobility, or normal middle-ear admittance of sound (Brooks, 1993). The latter outcome means that any hearing loss identified is likely to be sensorineural. However, tympanometry used alone may yield false-positive results (Krueger & Ferguson, 2002). Even when the presence of a middle ear disorder is confirmed, neither otoscopy nor tympanometry can reveal the impact of the disorder on hearing. This is the role of audiometry.

1.4.2. Audiometry

During audiometry, the intensity of pure tones (discrete frequencies of sound) are systematically varied to determine the lowest intensity that is audible. These are known as hearing threshold levels (HTLs). Air-conduction thresholds are measured using acoustic stimuli. It is also possible to place a bone-vibrator on the mastoid bone behind the ear, which can bypass outer- and middle-ear disorders to directly stimulate the inner ear. Significant differences between air- and bone-conduction HTLs indicate a conductive component to the hearing loss (BSA, 2018).

For most clinical applications, sound intensity is measured via the normalised loudness scale dB HL. The average level at which any pure tone frequency is detected by normally-hearing young adults is defined as 0 dB HL. Sounds less intense than 0 dB HL are assigned a negative value. The upper limit of most clinical measurements is 120 dB HL and frequencies over the range 0.25 to 8 kHz are routinely assessed (BSA, 2018).

HTLs are plotted on an audiogram (Figure 1.2). The intensity of the HTLs is plotted on the Y axis, with intensity increasing toward the bottom of the chart. Lower-pitched HTLs plotted on the left, and high pitches on the right along the X axis. Figure 1.2 shows two example audiograms. The first, plotted in blue, illustrates hearing within the normal range. The second example, plotted in red, shows a hearing loss that involves poorer audibility of high-pitched sounds compared to low-pitched sounds.

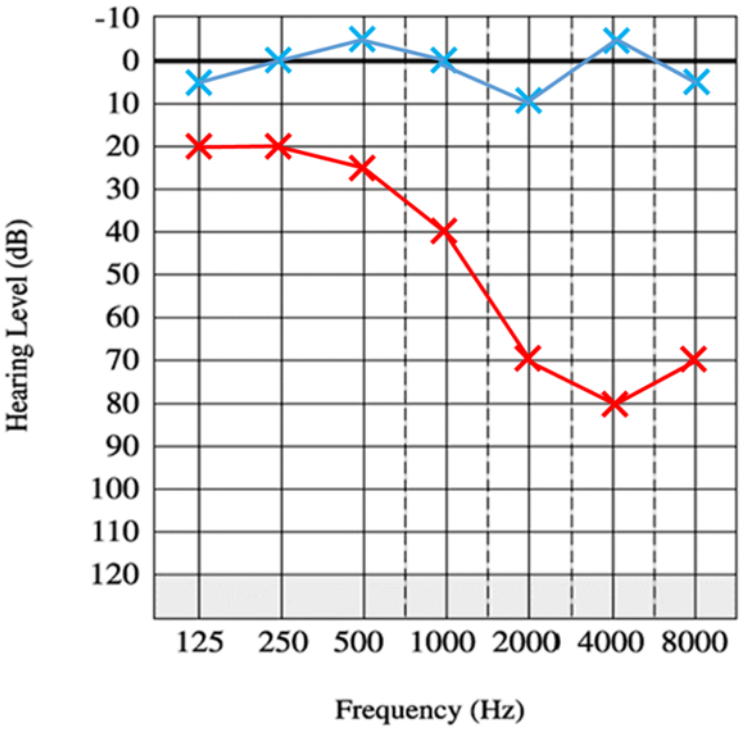


Figure 1.2: An audiogram

An audiogram plotting Hearing Threshold Levels (HTLS) by frequency. HTLs are marked by crosses. Sounds below the blue and red lines are audible to the ear with normal HTLs and a high-frequency hearing loss respectively.

Test-retest repeatability has been reported to be 93% or better on pure tone audiometry for both older children and adults across 0.5 to 4 kHz, (Orchik & Rintelmann, 1978; Schmuziger et al., 2004). However, children's behavioural

responses can be influenced by their motivation, understanding of the instructions, developmental ability, and earphone placement (Wood, 1993), creating a risk that measured thresholds may not be reliable for some children.

1.4.3. Audiometry adaptations for paediatrics

The gold standard of audiometry is for a patient to actively participate, following the recommended procedure issued by the British Society of Audiology (BSA, 2018). For children too young to respond by pressing a button every time they hear a tone, as an adult would, the procedure may be modified, with the child engaging in a game-like procedure in response to detecting a tone. Infants with head control when seated can be conditioned to turn their head toward a visual reward when a sound is heard (BSA, 2018). The optimum way to present sounds is via headphones or insert earphones, as this allows for ear-specific measurements. If a child will not tolerate this, an alternative method is soundfield audiometry, where sounds are played into the room via loudspeakers and the child listens with both ears together. In this case, the measured HTL will represent the threshold of either both ears detecting the sound simultaneously, or the threshold of whichever ear is more sensitive to sounds of that frequency. Children who are unable to be conditioned to respond in any way, due to lack of head control in early infancy or developmental delay for example, may be assessed using electrophysiological methods (BSA, 2021).

1.5. Classifying degree of hearing loss

Audiometry is used to classify the degree of hearing loss; however, classification systems vary internationally. In the U.K., the standard system is outlined in the BSA Recommended Procedure for Pure Tone Audiometry (BSA, 2018). Alternative systems are used by the American Speech Language Hearing Association (ASHA) and the WHO. Table 1.1 summarises the differences between these frameworks. It is not unusual for CIs to be described as suitable for people with 'severe to profound' hearing loss, both in the scientific literature and in information for the public online, without reference to any classification system. Table 1.1 shows that the interpretation of this term depends on which framework the reader assumes is being used, for both the level and frequency range of the HTLs.

Table 1.1: Different hearing loss classification systems

	BSA	ASHA	WHO
Frequencies over which thresholds averaged (kHz)	0.25, 0.5, 1, 2 and 4	Unspecified[¶]	0.5, 1, 2 and 4
Classifications			
Normal	≤ 20	≤ 10 to 15	≤ 25
Slight	—	16 to 25	26 to 40
Mild	21 to 40	26 to 40	—
Moderate	41 to 70	41 to 55	41 to 60
Moderately severe	—	56 to 70	—
Severe	71 to 95	71 to 90	61 to 80
Severe to profound	—	—	—
Profound	≥ 95	≥ 91	≥ 81

Key to Table: BSA = British Society of Audiology; ASHA = American Speech Language Hearing Association; WHO = World Health Organisation

[¶]ASHA do not issue formal guidance in the same way as the BSA or WHO. However, their website references a paper contrasting different classification systems, within which the author advocates for averaging the three most elevated thresholds out of 0.5, 1, 2 and 4 kHz to derive a value that will fall into one of the ASHA hearing loss categories above (Clark, 1981).

The differences between these classification systems means that CI candidacy guidance will be interpreted differently across different countries if specified only using hearing loss descriptor terms. For clarity, guidance must be given that refers to specific HTLs at specific audiometric frequencies.

1.6. Impact of hearing loss

Hearing loss impacts a child's ability to understand and to be understood via spoken communication. Children with hearing loss have poorer ability to perceive speech (Blamey et al., 2001) which in turn impacts their ability to produce speech and develop spoken language (Moeller et al., 2007; Wake, Hughes, Poulakis, et al., 2004). Permanent childhood hearing loss of any degree can cause poorer educational outcomes, with attainment gaps consistently found between children with hearing loss and their typically-hearing peers upon leaving compulsory education (Hendar & O'Neill, 2016; NDCS, 2021). Without normal hearing in both ears, children do not develop typical spatial listening skills such as sound localisation (Grieco-Calub & Litovsky, 2010; Zheng et al., 2015) or speech perception in noisy environments (Ching et al., 2018). It is therefore not surprising that hearing loss can impact children's quality of life (Looi et al., 2016; Roland et al., 2016) and mental health (Fellinger et al., 2012). In addition to the impact of hearing loss on communication, severe and profound hearing loss is associated with other auditory symptoms such as tinnitus and aversion to loud sounds (Ramos Macías et al., 2015).

Tinnitus is the perception of sound from within the head or ears without any external source. Prevalence estimates vary between 23.5% and 62.2% of

children with hearing loss (Rosing et al., 2016). It can have significant detrimental impact on children's sleep, ability to concentrate, ability to hear, and anxiety levels (Smith et al., 2019). As described in Section 1.3.1., reduction of a person's dynamic range in cochlear hearing losses can lead to decreased tolerance of loud sounds as well as poorer audibility of soft sounds. Increased sensitivity to or intolerance of environmental sounds of a range of intensities is known as hyperacusis and has been associated with conditions including tinnitus (Sheldrake et al., 2015) and autism (Khalfa et al., 2004).

1.7. Hearing devices

To minimise the potential negative impacts of hearing loss, the NHS offers a range of technology that aims to improve the audibility and clarity of sounds. Acoustic hearing aids and cochlear implants are the focus of this thesis and will be described in more detail in sections 1.7.1 and 1.7.2. Hearing aids are the standard care for hearing loss of mild or moderate degree, and when hearing aids can no longer provide adequate benefit the alternative is to provide cochlear implants for children with severe to profound sensorineural hearing loss as defined by TAG566 (NICE, 2019). An alternative technology is bone conduction hearing aids. While not the focus of this thesis, bone conduction aids will be briefly described in section 1.7.3 since they are relevant to Chapter 6.

Another alternative device is active middle-ear implants. Some models of middle-ear implant have a small overlap in audiometric indications with the 2019 NICE guidance for CI (Banakis Hartl & Jenkins, 2020); however, their

use in children is rare in the UK. Auditory brainstem implants are intended for patients with absent auditory nerves, and vibrotactile aids are body-worn devices that vibrate the skin in response to sound, rather than providing a percept of hearing. Both of these are highly specialist devices and outside the scope of this thesis. The following sections therefore describe the function of HA and CIs, the technologies representing the frontline and most common management options for children with severe or profound sensorineural hearing loss, with an overview of their benefits, risks, and limitations.

1.7.1. Hearing aids

Standard care, within the NHS and internationally, for children with mild, moderate, or severe sensorineural hearing impairment is to provide digital HAs (Feirn et al., 2014).

1.7.1.1 Hearing aid function

Sound that arrives at a HA microphone is converted into an electric signal. A digital algorithm is applied, based upon the child's HTLs. Digital signals are converted back into sound at the receiver (loudspeaker) and presented to the meatus via tubing that, for more severe losses, is embedded within an earmould that occludes the ear canal. Children's earmoulds are commonly made from soft silicon, based on an impression of the child's ear, to ensure a snug fit. The processed, amplified sound then travels through the outer and middle ear to cause BM motion in the cochlea and stimulate the cochlear sensory hair cells (Katz, 2002). Figure 1.3 shows a hearing aid in situ.

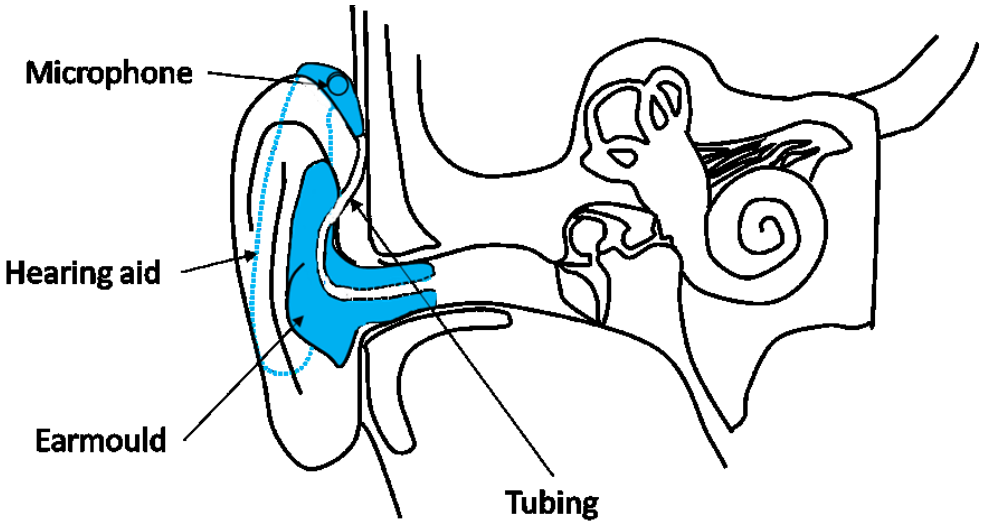


Figure 1.3: Schematic diagram of a hearing aid

1.7.1.2. Hearing aid benefits

HAs significantly improve the speech and language development of children with mild to severe hearing loss, via increased audibility of conversational speech (Tomblin et al., 2014). NHS commissioning guidance (Fleming, 2016) confirms their cost-effectiveness, albeit based on data from adults not children. Few papers assess the efficacy of HAs versus no intervention. This may be because their routine use for so many decades in wealthy countries means there has been little opportunity to document the outcomes of a no-intervention group. However, evidence in adults with mild to moderate hearing loss shows that HAs improve both quality of life and listening outcomes (Ferguson et al., 2017). The amount of audibility provided by HAs is associated with children's rate of language growth (Tomblin et al., 2014). It is also known that congenitally deaf children who do not receive HAs before 6 months of age experience a lasting detriment to their language development, regardless of the

degree of hearing loss (Yoshinaga-Itano et al., 1998). It is therefore assumed that continued lack of intervention would result in increasingly poor spoken language development in comparison to normally-hearing children. Children's outcomes with HAs worsen with increasing degree of hearing loss (Lovett et al., 2015), leading to a need to establish at what point a child would benefit more via CIs.

1.7.1.3. Hearing aid risks

One risk of HA use is that occluding earmoulds prevent ear canal aeration, risking the uncomfortable build-up of wax or other debris, or lead to ear canal infections (Orji et al., 2014; Seidel et al., 2019). There is a risk that over-amplification of sounds might worsen children's hearing loss (Macrae, 1991) and the risk of battery ingestion if disposable rather than rechargeable batteries are used without the appropriate tamper-proof battery drawers, however these risks can be avoided by following best practice (British Academy of Audiology Service Quality Committee, 2022).

1.7.1.4. Hearing aid limitations

HA-amplified sound must travel through any conductive barrier, meaning it is impeded by the presence of middle ear disorders which are prevalent in children (Haggard, 1993). Sound is then transduced within the cochlea. At this stage, any loss of outer or inner hair-cells, or hearing nerve function, will introduce distortion before the sound signal ascends to the central auditory system. There is a limit to the amount of amplification HAs can provide. The

combination of this technical limitation along with the physiological limitations of a damaged cochlea means that HA use yields less benefit in speech perception with increasing degree of hearing loss (Leigh et al., 2016; Lovett et al., 2015).

1.7.2. Cochlear implants

The need for an intervention to bypass cochlear hair cells prompted the development of CIs. Figure 1.4 illustrates a CI system in situ. Sound arrives at the microphone and is converted into a digital signal within the speech processor. A wire connects the speech processor to a transmitting coil, which contains a magnet. This magnet is attracted to a magnet within the internally implanted receiver/stimulator. The coil transmits frequency modulated signals to the implant, which has an array of stimulating electrodes. The electrodes emit electrical pulses that stimulate the spiral ganglion. The resultant neural signals are conducted along the auditory pathway and perceived as sound (Clarke, 2003).

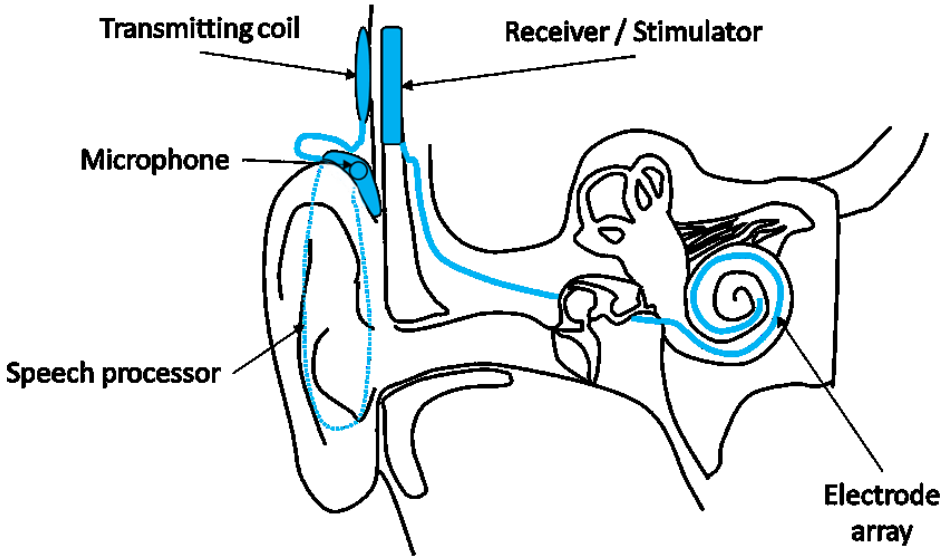


Figure 1.4.: Schematic diagram of a cochlear implant.

A standard CI will have an electrode array of between around 20 to 30 mm. However, some patients with stable HTLs and good low-frequency hearing can be given a shorter-length electrode array. These provide electrical stimulation to represent only high-pitched sound and simultaneously provide acoustic amplification for low-pitched sounds to the same ear. This is known as electric-acoustic stimulation (EAS) (Gantz et al., 2016).

1.7.2.1. Cochlear implant benefits

CIs provide a sensation of hearing that is sufficient for even profoundly deaf children to develop spoken language (Ruben, 2018), with research showing that early implantation allows some children to develop at a rate similar to that of normal-hearing children (Niparko et al., 2010). Unlike many children's HA fittings, there is generally no need for the ear canal to be occluded, decreasing the risk of outer ear infection. For profoundly deaf children, CIs are acceptably safe and cost-effective for the NHS, improving quality of life

(Bond, Mealing, et al., 2009), speech understanding in quiet and background noise, sound-source localisation, language acquisition, and access to mainstream education compared to outcomes from HAs (Barton et al., 2006a, 2006b). Profoundly deaf children with no intervention are unlikely to develop normal spoken language and instead are likely to be dependent upon communicating via sign language (Stokoe, 2005).

1.7.2.2. Cochlear implant risks

There are risks associated with any surgical procedure, such as allergic reactions to anaesthetic medication. CI surgery also carries specific risks including cutaneous or soft tissue infections, meningitis, facial nerve damage, altered taste, cholesteatoma, cerebrospinal fluid leak, inducement of tinnitus or dizziness and misplacement or movement of the CI magnet or electrode array, and cessation of internal device function, requiring CI replacement (Loundon et al., 2010). In addition, the trauma of electrode array insertion can result in the loss of any remaining cochlear function (Scheperle et al., 2017). Hence, the decision to insert a CI can remove the option of ever reverting to HA use in that ear. The recipient will need to remain under the care of a CI centre for as long as the implant is used, for their safety.

1.7.2.3. Cochlear implant limitations

The effectiveness of CI systems can be limited by both technical and anatomical factors. CIs cannot provide normal perception of sound's temporal, spectral, or intensity qualities as the stream of electrical pulses they deliver

primarily encode only the slow changes in amplitude over time and there are limitations on how fast they can be delivered and produce distinct changes in neural responses within the cochlea (Wilson & Dorman, 2008). Stimulation from any electrode also results in neural firing across a range of the basilar membrane (referred to as 'current spread'), which is dependent on the location of the electrode and the kind of stimulation paradigm that is used, resulting in sub-optimal spectral resolution (Clarke, 2003). The dynamic range of electrical stimulation is also smaller than that of normal acoustic listening, resulting in poorer speech discrimination for people using CIs compared to people with normal hearing, particularly in the presence of background noise (Nelson et al., 2003). The CI technology acts at the periphery of the auditory system, but to be heard must stimulate neural signals that are conveyed along the auditory nerve to the brain. Therefore, their effectiveness can be limited by conditions such as cochlear ossification following meningitis, where electrically-insulating calcification of the cochlea prevents the electrical pulses from optimally reaching the auditory nerve (Yan et al., 2019), or absent or dysfunctional auditory nerves that cannot convey the signals to the brain (Peng et al., 2017). In the absence of cochlear or neural abnormalities, long-duration auditory deprivation can leave children unable to benefit from bilateral hearing due to irreversible cortical reorganisation (Sharma et al., 2002).

1.7.3 Bone conduction aids

Bone conduction aids vibrate sound signals onto the mastoid bone, bypassing the outer and middle ears to stimulate the cochlea. Unlike CIs, bone

conduction aids are not indicated for people with bilateral severe to profound sensorineural losses but for those with bone-conduction HTLs of less than 60 dB HL (Rahne & Plontke, 2022). They are therefore not a viable option for the children within scope for the primary aims of this thesis. However they are mentioned here because they were used by some of the children in the Born in Bradford collaboration presented in Chapter 6.

1.8. Factors influencing outcomes of children with hearing loss

Deaf children's development is influenced by many factors aside from the type of hearing device they receive. Both listening and language skills are significantly impacted by the degree and duration of auditory deprivation a child experiences. There are multiple critical periods for auditory development (Friedmann & Rusou, 2015). A lack of auditory stimulation in early life alters the development of the central auditory system (Sharma et al., 2005) and can lead to cross-modal re-organisation (Buckley & Tobey, 2011). Greater auditory deprivation is associated with poorer oral / aural outcomes, whether due to a child's age at intervention relative to the onset of hearing loss (Niparko et al., 2010; Yoshinaga-Itano et al., 1998), the duration of daily HA or CI use following first fitting (Walker, Holte, et al., 2015), or the unilateral auditory deprivation associated with asymmetric deafness (Gordon et al., 2015).

It is estimated that around one third of children who end up receiving CIs have additional disabilities and that outcomes among this group are more variable than children who do not have these additional difficulties (Birman et al., 2012; Mesallam et al., 2019). These can include developmental disorders that affect social communication such as attention deficit hyperactivity disorder or

autism and can create difficulties in accurate hearing assessment (Beers et al., 2014). Poorer listening outcomes are seen in children with global developmental delay (Berrettini et al., 2008). Tinnitus can affect children's ability to listen (Chadha et al., 2009). Last, even for children with normal hearing, language outcomes are influenced by the amount and complexity of spoken language they are exposed to (Gilkerson et al., 2018). This includes the language environment at home and school, as well as access to specialist support services (Yoshinaga-Itano et al., 2020) and may explain the association between children's language development and maternal education (Noblitt et al., 2018). The factors above must be controlled for in comparisons of children's outcomes with different hearing devices, to avoid reaching erroneous conclusions about their relative efficacy.

The following section will present the findings of the comparative studies that have been used to support CI candidacy guidance.

1.9. Evidence available to support candidacy guidance

Audiometric CI candidacy recommendations proposed by research studies include average unaided HTLs of between 88 to 96 dB HL across 0.5, 1 and 2 kHz (Davidson, 2006), 80 dB HL across 0.5, 1, 2 and 4 kHz (Lovett et al., 2015), and 60 to 65 dB HL across 0.5, 1 and 2 kHz (Leigh et al., 2016). The lack of agreement between studies was influenced in part by different choices the authors made regarding how much certainty of benefit was required to recommend CIs over HAs. Lovett et al. chose odds of 4:1 of better outcomes with CIs than HAs as this ratio had resulted in acceptable cost effectiveness for CIs in adults (U.K. Cochlear Implant Study Group, 2004) and was used in

prior children's CI candidacy recommendations (Boothroyd, 1993). Lovett et al. acknowledged that policy makers may wish to base guidance on different odds, and so presented a range of options for both four- and three-frequency averaged HTL criteria with odds from 1:1 to 8:1. The impact of changing the odds of improvement across this range depended on which of the three outcome measures were used, changing the audiometric guidance by between 9 and 36 dB (Lovett et al., 2015). Leigh et al. (2016) reported audiometric guidance ranging from 60 to 82 dB HL averaged across 0.5, 1 and 2 kHz as the odds of improvement increased from 75% to 95%. Another source of variability in suggested audiometric criteria was the level and type of speech stimulus used to derive the recommendations. Davidson (2006) found significantly better performance for children with CIs than HAs for HA users with 3-frequency averaged HTLs of 97 to 113 dB HL when words were presented at 70 dB SPL with no background noise, compared to the non-overlapping range of 88 to 96 dB HL when the same stimuli were presented at the softer level of 50 dB SPL, i.e., basing guidance on the perception of softer speech led to the recommendation of CIs for children with more residual hearing. Leigh et al. found different cusps depending on whether phoneme or sentence measures were used, deriving from the same dataset audiometric criteria of 75 dB HL based on sentence perception in quiet (Leigh et al., 2011) and 65 dB HL based on phoneme perception in quiet (Leigh et al., 2016). Lovett et al. (2015) found a 10 dB difference in the cusp depending on the type of background noise used during the same word perception test. Holding the odds of improved performance with CIs steady at 4:1, results indicated a 4-frequency HTL criteria of 86 dB HL based on the McCormick Toy

Discrimination test (McCormick, 1977; Ousey et al., 1989) in pink noise, compared to 76 dB HL for the same test with Dutch babble as a background noise.

Rather than comparing outcomes from groups of children, Leal et al. (2016) used an alternative approach based on 16 hypothetical audiograms where unaided HTLs were fixed at either 90 or 80 dB HL for both 2 and 4 kHz and thresholds at 0.5 and 1 kHz were the same as each other and varied from 20 to 90 dB HL in 5 dB steps. The authors derived aided speech intelligibility index (SII) values (American National Standards Institute, 1997) for each audiogram. The SII varies from 0 to 1 and indicates what proportion of speech is audible to a listener via a combination of their hearing loss and HA amplification, where 0 indicates no speech is audible via a HA and 1 indicates that all speech is audible. In each audiogram modelled, the SII value was 0.65 or below. Based on previous research (Stiles et al., 2012; Tomblin et al., 2015) this finding suggested that any childhood hearing loss where HTLs were equal to or greater than 80 dB HL at 2 and 4 kHz would result in inadequate benefit from hearing aids.

The studies described above form the evidence base that has been cited to justify recent audiometric candidacy guidance by both the British Cochlear Implant Group (BCIG) and the American Cochlear Implant Alliance (ACIA). However, as described, there was no one consistent recommendation from these various studies. It is then not surprising that there has not been a consistent approach to translating the evidence into clinical recommendations. The following paragraphs set an international context for this thesis, by illustrating examples of clinical guidelines and practice.

1.10. International audiometric CI candidacy guidance

Simultaneous or sequential bilateral CIs are permitted in France, where children with moderate or worse hearing loss can be considered candidates based on their worse-hearing ear but having a mild loss or typical hearing in the better-hearing ear precludes implantation of the worse-hearing ear (Simon et al., 2019). In Belgium, the audiometric criterion for the ear to be implanted also depends on the symmetry of the hearing loss. In the ear(s) to be implanted, three or more thresholds including 0.5, 1, 2, and 4 kHz must equal or exceed 70 dB HL for bilateral losses, or ≥ 85 dB HL in asymmetric losses (ASGB, 2019). Canadian practice varies by province and individual clinics are free to determine their own inter-implant intervals and audiological criteria. In 2011, examples included: 'bilateral severe to profound hearing loss' in Nova Scotia; 'severe hearing loss' in Quebec; 'manufacturer indications approved by both Health Canada and the US Food and Drug Administration' in Alberta; to no fixed audiometric criteria but the application of 'best available evidence' at one centre in Ontario (Hanrahan, 2011). In the United States, Food and Drug Administration (FDA) unaided threshold criteria vary between CI manufacturer, including a four-frequency average across 0.5, 1, 2 and 4 kHz of ≥ 90 dB HL for Med-El (Gfoeller, 2019) and > 80 dB HL for Cochlear Corp. (Cochlear Corporation, 2021). However, a survey of American Neurotology Society members found that 78% of respondents reported carrying out 'off label' CI surgeries within the previous 2 years, with 31% having provided CI(s) to children with more residual hearing than indicated by the FDA. Both simultaneous and sequential bilateral implantation is permitted. These examples are not exhaustive but serve to illustrate the range of international

practice. This sets the context for audiometric candidacy guidance in the UK, which will be described in the following section.

1.11. NICE cochlear implant candidacy guidance

The provision of CIs in the UK follows guidance from NICE. Their first guidance, TAG166, was issued in 2009 (NICE, 2009). To be eligible for CIs on the NHS, children needed to have 'severe-to-profound deafness'. This was not defined in line with any of the recognised hearing loss classification systems described in section 1.5, but rather as having HTLs ≥ 90 dB HL at 2 kHz and 4 kHz in both ears (i.e., within the shaded area of Figure 1.5. below). In addition, children had to be shown not to receive 'adequate benefit' from HAs, defined by NICE as having "speech, language, and listening skills appropriate to age, developmental stage and cognitive ability". This guidance was established following a consultation process and was based on the best available evidence at the time. However, over subsequent years CI technology and candidacy assessment processes improved. Increased awareness of the benefits of early intervention led to earlier implantation and both factors led to better outcomes for children with CIs. Globally, this led to a gradual widening of CI indications until the 2009 NICE guidance was among the most conservative in the world, as children who would have received CIs elsewhere remained ineligible for CIs in England and Wales (Vickers, de Raeve, et al., 2016).

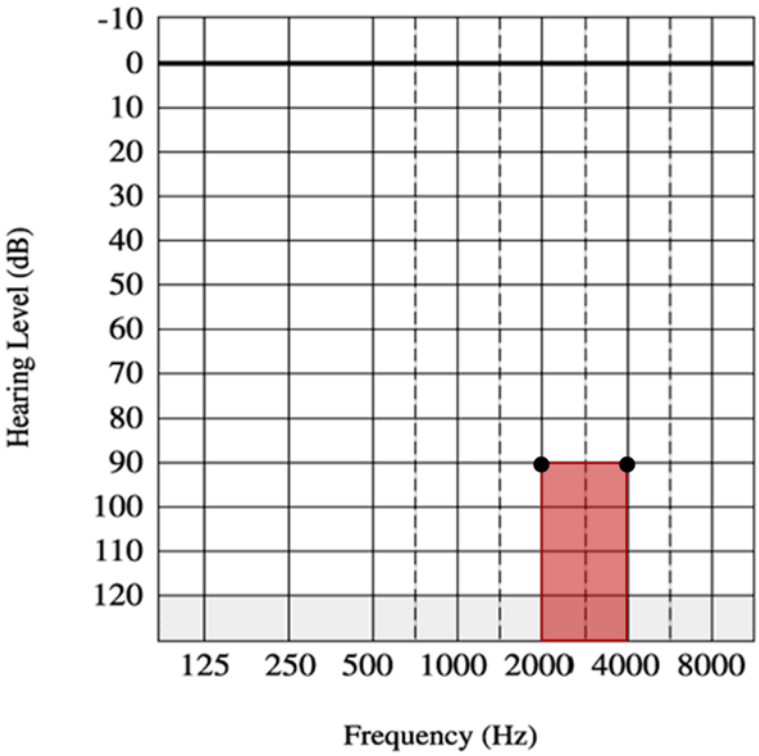


Figure 1.5: NICE TAG166 (2009) audiometric candidacy guidance

Figure 1.5. Legend: Hearing threshold levels at both 2 and 4 kHz needed to lie within the shaded area for both ears.

Concerns were raised by the British Cochlear Implant Group (BCIG) Working Group on Candidacy that the audiometric criterion should be widened to capture those likely to benefit more from CIs than from HAs (Vickers, Kitterick, et al., 2016). NICE initiated a consultation on a part-review of its 2009 CI guidance (NICE, 2018). In January 2019, a final appraisal document was received by consultees, proposing that the definition of 'severe to profound deafness' be changed to HTLs of 80 dB HL or greater at any two or more frequencies including 0.5, 1, 2, 3, and 4 kHz. 80 dB HL falls within the range of cusps suggested by research studies described above. However, there was no evidence for the use of any two frequencies rather than an average across two or more frequencies, nor for the inclusion of 3 kHz. NICE

confirmed that the proposed amendment was to be adopted and it was enacted from June 2019 in TAG566 (NICE, 2019). This revised audiometric guidance is illustrated in Figure 1.6. Reference to Table 1.1 confirms that this is different to any internationally recognised definition of 'severe to profound' hearing loss. Neither of the definitions of 'severe to profound' hearing loss used by NICE aligned with the ASHA, WHO or BSA classification systems.

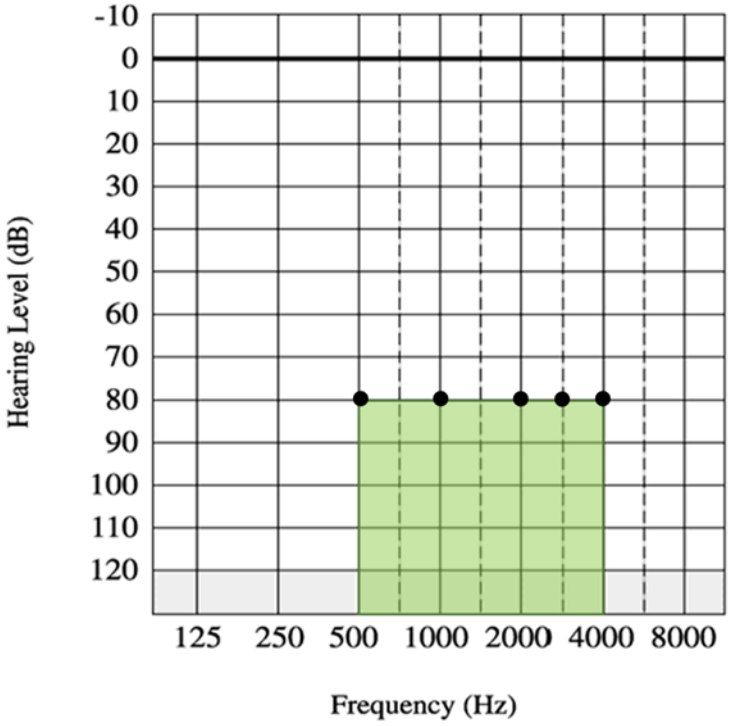


Figure 1.6: NICE TAG566 (2019) audiometric candidacy guidance
Hearing threshold levels at any two or more of the frequencies highlighted with a black circle must fall within the shaded area in both ears.

In contrast to other countries, NICE did not revisit their requirement for both of a child's ears to meet the audiometric criterion for either ear to be implanted. Nor did it review its previous decision to only fund bilateral CIs if both implants were given in one surgical procedure, rather than sequentially on separate occasions (NICE, 2018). Hence in England and Wales it is not

permitted for a CI to be implanted into a child's worse-hearing ear unless or until their better-hearing ear also meets the audiometric criteria (NICE, 2019).

1.12. Other factors influencing access to cochlear implantation

Once a child is referred for assessment at a CI service, further criteria and clinical discretion are applied locally before a child is permitted access to CIs (Lazaridis et al., 2010). These criteria stem from a framework to discern which children were likely to succeed with CIs, first proposed by Hellman et al., (1991). Clinicians evaluated children and their families across 11 domains thought to influence outcomes, selecting "no concern", "mild to moderate concern" or "great concern" for: chronological age; duration of deafness; medical/radiological findings; multiple handicapping conditions; functional hearing; speech and language abilities; family support and structure; the parent(s)' and child's expectations of the child's outcomes with CI; educational environment; availability of support services; and cognitive learning style. The framework was named the Children's Implant Profile (ChIP).

The ChIP has been modified by different groups over time. Different versions are used at different CI centres in place of the original version, e.g., the Nottingham ChIP (Nikolopoulos et al., 2004) and Great Ormond Street Hospital GOSHChIP (Edwards et al., 2009). ChIP ratings are not always formally quantified, or even applied consistently within each CI centre (Lazaridis et al., 2010). However, it is standard practice in the UK for the multi-disciplinary CI team to evaluate a child and their family across a range of domains such as those within the ChIP before offering CIs, to avoid surgery

for children unlikely to benefit. This includes assessing whether the child meets the NICE guidance requirement of not receiving ‘adequate benefit’ from HAs. This is a complex judgement to make at the very young ages when cochlear implantation is now routinely considered, as children can be too young to complete behavioural assessments of HA benefit. The NHS pathway from HA use under local audiology services to cochlear implantation, including audiometric and ChIP assessments, is summarised in Figure 1.7.

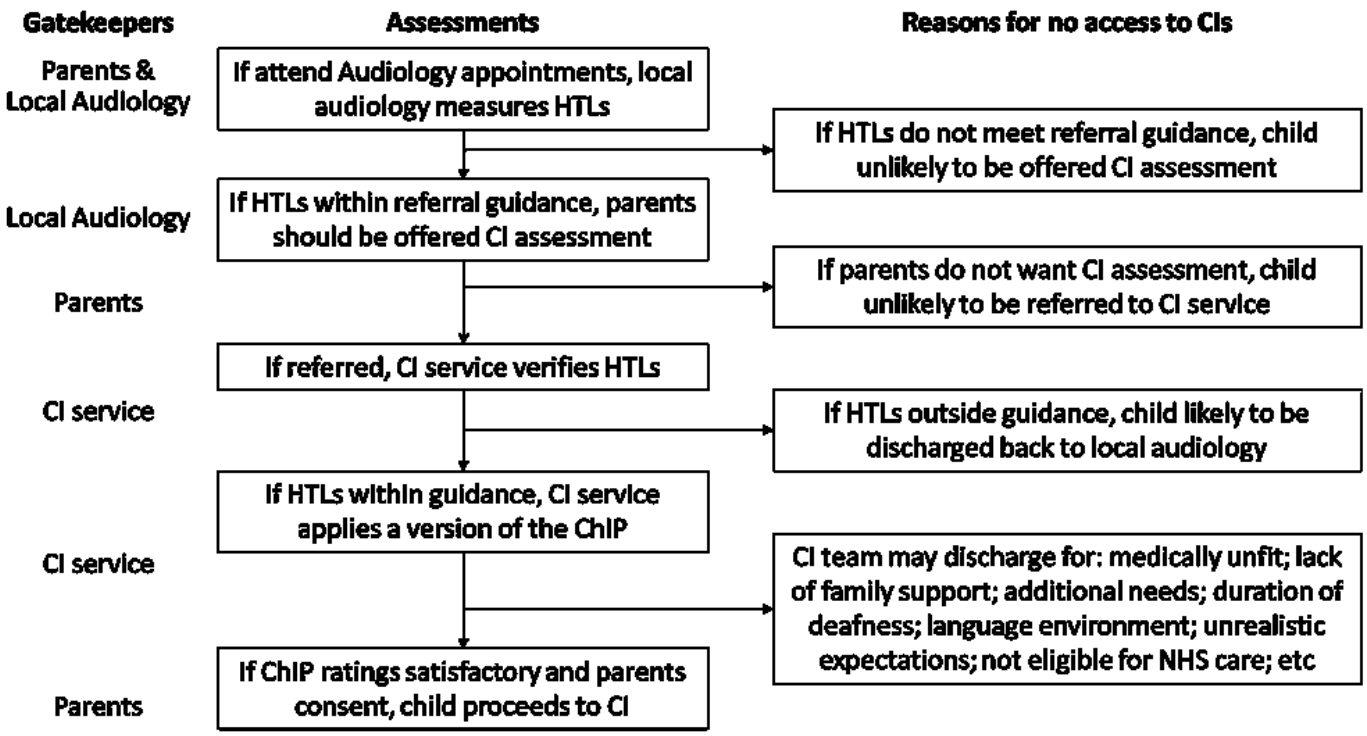


Figure 1.7: Child's pathway from Local Audiology to cochlear implantation

Figure 1.7. Legend: CI = cochlear implant; ChIP = Children's Implant Profile; HTLs = hearing threshold levels; NHS = National Health Service.

Figure 1.7 illustrates how a child's access to CIs is controlled by different gatekeepers throughout the care pathway. It also highlights the role of HTL criteria within the wider assessment process. Without regular and complete audiometry at the HA service, appropriate referral to a CI centre might not occur. Further, without meeting the NICE audiometric criterion, there is a much-reduced likelihood of a child receiving CIs, as surgery would be dependent on a successful application to the commissioners for special funding. Children with congenital deafness undergo this CI eligibility assessment as infants, making it difficult to diagnose any other additional difficulties or to assess their cognitive ability as a context for the listening and language skills they have developed via HAs. This places greater weight on meeting the HTL guidance.

It is important to note that Figure 1.7 shows a simplification of the care pathway and exceptions are sometimes made to meeting the NICE audiometric guidance. One example is that of ANSD. If neural dys-synchrony is diagnosed, a child might be offered CIs even if their HTLs fall outside the guidance, as it is recognised that their HTLs may fluctuate and are less strongly associated with speech perception (Rance, 2005). However, these cases are the exception rather than the rule. For most children, meeting NICE criteria is a requirement for access to CIs because otherwise commissioners may refuse to fund the procedure.

1.13. The Born in Bradford cohort study

The Born in Bradford (BiB) cohort study recruited families of babies born at Bradford Royal Infirmary between 2007 and 2011 to explore how medical,

environmental, behavioural, and social factors affect children's health and development (Raynor, 2008; Wright et al., 2013). The BiB dataset has enabled the creation of novel healthcare evidence across many varied conditions including, but not restricted to, visual impairment, the impact of air pollution on health, and musculoskeletal function (Bruce et al., 2016; Craig et al., 2022; Garcia et al., 2021). For reasons that will be explained in Chapter 3, this study presented an opportunity to link BiB data with audiological data for the first time to investigate the impact of hearing loss on children's educational outcomes. This secondary study will be presented in Chapter 6.

1.14. Scope of the thesis

The following sections give a rationale for the scope of this thesis, in the context of UK healthcare commissioning. It was beyond the scope of this thesis to address comparative outcomes with HAs and CIs for every clinical scenario.

1.14.1. Focus on aural / oral outcomes

Some families of children with hearing loss may want their child to communicate via British Sign Language (BSL) instead of spoken communication. However, many families would like their child to have the opportunity to learn to listen and talk so that they can be part of 'the hearing world', with or without also learning BSL and integrating into the Deaf community. This thesis focuses on outcomes related to spoken language rather

than BSL fluency, because the cost-effectiveness and provision of CIs on the NHS is based on spoken language development.

1.14.2. Classification of hearing loss based on the better-hearing ear

In some countries, clinicians can implement a treatment plan for each patient's right and left ears individually. For instance, a CI can be provided to the worse-hearing ear, while the other ear might be optimally fitted with a HA or may have normal hearing and require no device at all. In most wealthy nations, subsequent implantation of the better-hearing ear would be permitted should the hearing loss in that ear worsen. This approach is intended to optimise children's binaural listening, especially in noisy environments and for locating the sources of sounds (Polonenko et al., 2018). Provision of optimum hearing devices to both ears at each stage of childhood limits the impact of aural preference syndrome, where asymmetric, long-term auditory deprivation leads to central reorganisation that reduces the ability of a child to ever listen optimally via the more deprived ear (Gordon et al., 2015). In contrast, the 2019 NICE criteria define CI candidacy with reference to the better-hearing ear. Hence in England and Wales, both of a child's ears must have HTLs within audiometric criteria for that child to be offered a CI for either ear. Furthermore, sequential bilateral implantation is not funded on the NHS. Children must either receive two CIs in one procedure or remain using only one CI for the rest of their lives, regardless of any future deterioration in the hearing of their non-implanted ear (NICE, 2019). To ensure relevancy to NHS practice, this thesis classifies children according to their better-hearing ear.

1.15. Aims and objectives

There is a need for more research into the comparative outcomes of a large group of children with severe hearing loss using HAs compared to a large group of implanted children who are either very similar in terms of the factors known to influence outcomes with hearing devices or can be characterised accurately enough to control for any differences between the groups. This PhD aimed to compare the clinical effectiveness of CIs to the clinical effectiveness of HAs for children with hearing losses close to NHS audiometric CI eligibility, to support children's CI candidacy decision-making in the context of the NHS.

A secondary aim was to measure the impact of hearing loss on children's early educational outcomes by linking data from the present study with data from the BiB study.

These aims were met by achieving the following objectives:

Chapter 2: Conduct a comprehensive scoping review of the literature comparing the clinical effectiveness of HAs for children with severe hearing loss with the clinical effectiveness of CIs, to inform stakeholders and identify gaps in the knowledge-base.

Chapter 4: Assess the impact of TAG566 (NICE 2019) on children's eligibility for CIs by studying children without access to cochlear implantation.

Chapter 5: Compare the clinical effectiveness of HAs for children with hearing within or close to eligibility for CIs to the clinical effectiveness of CIs.

Chapter 6: Measure the impact of hearing loss on the early years educational outcomes of children screened for the present study and enrolled in the Born in Bradford cohort study.

Chapter 7: Synthesise and discuss the findings of the thesis in terms of the implications for children's hearing care and suggestions for future research.

CHAPTER 2: A SCOPING REVIEW OF STUDIES COMPARING OUTCOMES FOR CHILDREN WITH AND WITHOUT ACCESS TO COCHLEAR IMPLANTATION

An alternative version of this scoping review was published as:

A scoping review of studies comparing outcomes for children with severe hearing loss using hearing aids to children with cochlear implants.

Killan, C.F., Hoare, D.J., Katiri, R., Pierzycki, R.H., Adams, B., Hartley, D.E.H., Ropar, D., Kitterick P.T.

Ear and Hearing 43(2): 290-304, March/April (2022)

Student contribution:

I was involved in the development of the search strategy, with input from the supervision team. I carried out all of the searches, downloaded results into record management software, transferred into excel and removed duplicate records. I performed title and abstract screening for all records. Co-screening was carried out by one of several other members of the team. I also carried out full text screening for every record passed to this stage, with one of the other team members co-screening each record. I extracted data from all included records and this was checked by one other member of the team in each case. I took the lead in data presentation and manuscript writing, with other team members contributing critically to the final manuscript.

2.1. INTRODUCTION

This chapter presents a comprehensive scoping review of the literature comparing the clinical effectiveness of HAs for children with severe hearing loss with the clinical effectiveness of CIs, to inform stakeholders and identify gaps in the knowledge-base. By cataloguing previously published comparative studies, and identifying gaps in the evidence base, this scoping review provided a foundation for the design of the experimental work described in subsequent chapters, ensuring that the PhD research would be novel and clinically relevant. This introduction describes previously published reviews of literature around children's CI candidacy.

One seminal systematic review of literature published up to July 2007, was published by Bond, Elston, et al., (2009). The authors concluded that unilateral CIs were clinically effective and cost-effective for children with bilateral profound hearing loss (>95 dB HL). No recommendations were made for CI for children whose better-ear unaided thresholds averaged 70 to 95 dB HL, because of an absence of evidence at that time for what outcomes were likely to improve. They found an absence of data on quality of life or educational attainment and recommended that these should be measured in future studies to improve the evidence upon which CI candidature guidance is based. They recommended that studies were needed to establish the benefits of CIs for children with additional needs, and to determine the location of the audiometric cusp beyond which CIs would be unlikely to provide clinically meaningful benefits and/or cease to be cost-effective compared to HAs.

Since the latest publication date for studies reviewed by Bond, Elston et al. (2009) of July 2007, many studies of children with hearing loss have been

published, and both clinical practice and HA and CI technologies have evolved. Increasingly, CIs are fitted bilaterally rather than unilaterally, closer in line with HA practice, and age at implantation has tended to decrease. (Ramsden et al., 2012; Teagle et al., 2019; Yoshinaga-Itano et al., 2018). Manufacturers have introduced improved features such as new sound processing algorithms and microphone directionality options (Lorens et al., 2010; Spriet et al., 2007). Therefore, comparing the outcomes of children using HAs and CIs must be reviewed regularly, because changes in practice and technology might influence the cusp at which implantation should be considered.

The research studies described in Chapter 1 indicated that the audiometric cusp for candidacy may now lie somewhere within the range of 65 – 95 dB HL, i.e., severe hearing loss (Davidson, 2006; Leigh et al., 2016; Lovett et al., 2015). De Kleijn et al. (2018) presented a review of comparative studies of children with severe hearing loss using HAs to children using CIs, including 10 records. This review provided a valuable summary of studies that could be used to define audiometric criteria, however the literature search was restricted to studies of speech production, speech perception, receptive language, and auditory performance only. There remained a need to catalogue how other outcomes varied between these groups, including quality of life and educational attainment. Further, provision of HAs or CIs to children with more residual hearing to lose than traditional CI candidates might also affect outcomes that may not be routinely measured in the clinic such as spatial hearing, listening effort and fatigue, psychosocial outcomes (such as emotional disorders, behavioural problems and social difficulties), vestibular function,

tinnitus, and music perception (Dorman et al., 2016; Fiorillo et al., 2017; Ganek et al., 2020; Killan et al., 2018; Looi, 2014; Winn, 2007; Wong et al., 2017). Optimising these outcomes might not be the primary goal when choosing a listening device. However, they are important as they can impact children's quality of life, mental health, social and recreational participation, sleep, and educational attainment (Camarata et al., 2018; Fellinger et al., 2015; Inoue et al., 2013; Smith et al., 2019; Vecchiato et al., 2013).

In summary, there was no review of recent studies comparing the outcomes of children with severe hearing loss using HAs to children using CIs that included a wider range of outcomes than those directly related to speech reception and speech and language development. A scoping review can achieve this goal (Arksey & O'Malley, 2005), because it is designed to clarify what is known and what is not known and identify areas for future research.

This chapter describes the findings of a scoping review that addresses the question 'What research has been conducted comparing outcomes in children using CIs to outcomes in children using HAs with severe hearing-loss in the better-hearing ear?' For this review, a definition of 'severe' inclusive of the WHO, ASHA, and BSA classification systems was used (British Society of Audiology, 2018; Clark, 1981; World Health Organisation, 1991), that is, average unaided HTLs in the better-hearing ear between 61 to 95 dB HL. Bond, Elston, et al., (2009) found no studies comparing children using CIs to children using HAs with thresholds in this range. Applying this definition of 'severe' was intended to remove bias in study selection for or against countries using different classification systems and to capture all potentially relevant studies published since those reviewed by Bond, Elston, et al., (2009). It also

included the range of audiometric criteria proposed by Davidson (2006), Leigh (2011 & 2016) and Lovett (2015).

Two objectives were defined toward answering the scoping review question:

i) To catalogue the characteristics of studies pertinent to candidature of children with severe hearing loss for cochlear implantation.

ii) To identify gaps in the evidence base regarding comparative outcomes for children with severe hearing-loss using HAs and children using CIs, to inform future research projects and identify opportunities for evidence synthesis.

2.2. METHODS

This scoping review was designed, conducted, and presented in line with guidance from the Joanna Briggs Institute and the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) extension for scoping reviews (Tricco et al., 2018).

2.2.1. Eligibility criteria

To be included in the review, records needed to contain data from either a group of children with severe hearing-loss who were HA users compared to a group of children using CIs, or data from a group of children with severe hearing-loss assessed before and after they received CIs. Outcomes of interest included all those that could feasibly be influenced by the provision of a CI or HA. Qualitative, quantitative, and mixed methods studies were all included.

We aimed to ensure that our review complemented rather than duplicated Bond, Elston, et al., (2009). We therefore searched for studies published from July 2007 to the present, immediately following the search by Bond, Elston, et al., (2009) but overlapping the period considered by de Kleijn et al., (2018) due to the more restricted range of outcomes they considered. Only peer-reviewed records were included. We included both open-access and non-open-access articles. Because of resource limitations, only records with full texts written in English were included.

2.2.1.1 Participant inclusion and exclusion

All participants needed to be aged less than 18 years and meet our definition of 'severe' hearing loss. Records were therefore excluded if they had only normally-hearing participants, participants who were all profoundly deaf or traditional CI candidates (e.g., described using terms such as "profoundly deaf", "total deafness", "severe-to-profound"), and those where all participants had normal or near-normal hearing in one ear; i.e., "single-sided deafness". For the purposes of this review, severe hearing loss was defined as pure tone thresholds in the better-hearing ear, averaged across 0.5 to 4 kHz, of 61 to 95 dB HL. During full-text screening, studies were excluded if it was not possible to confirm that all HA users had unaided thresholds within our definition of severe hearing-loss. When it was not possible to determine this from the text, the corresponding authors were contacted for clarification. In the absence of confirmation that any individual participants in the HA group met this criterion (HTLs were not reported or could not be obtained directly from authors), studies were included if where the reported participant characteristics for the

average HTLs of the better-hearing ear fell within the range described above. If only group characteristics were reported, studies were included where the group had a mean HTL average within the defined range. If only qualitative descriptions of the degree of hearing loss were given with no supporting audiometric data, studies were included that reported children's hearing fell within the "severe" range. No audiometric inclusion criteria were applied to children in the CI groups of between group comparison studies.

2.2.1.2. Intervention inclusion and exclusion

Intervention inclusions for the HA group were that children wore at least one acoustic HA and no CI. Intervention inclusions for the CI group were that children used at least one multi-electrode, intra-cochlear hearing implant. The CI group could include children with unilateral CI alone, unilateral CI with a contralateral acoustic HA (bimodal aiding), unilateral or bilateral short arrays for electric-acoustic stimulation (EAS) or bilateral CI. Intervention exclusions for both HA and CI groups included any use of auditory brainstem implants, bone-conduction devices, and vibrotactile aids.

2.2.1.3. Outcome inclusion and exclusion

Outcomes that might plausibly be influenced by the provision of either CIs or HAs, such as listening, language, speech production, reading, music perception, balance, dizziness, tinnitus, educational attainment, psychosocial outcomes, mental health and quality of life were included. Studies that did not measure such outcomes were excluded. Illustrative examples of outcomes not

within scope included, but were not restricted to, surgical techniques, development of new tools, or audits of patient pathways.

2.2.1.4. Study design inclusion and exclusion

Study designs within scope included observational or interventional studies documenting the outcomes of HAs versus CIs. This included peer-reviewed studies in scientific or medical journals reporting randomised controlled trials, quasi-randomised controlled trials, before and after studies, non-randomised controlled trials, cross-over studies, cohort studies, and case control studies. Case studies and case series were excluded during title and abstract screening. However, studies that were passed to full text screening were retained if they included data from a sub-group with severe hearing loss or individual data for participants who met our inclusion criteria. Study designs out of scope included reviews of any kind, along with magazine articles, conference presentations, practice guidelines, expert opinions, book chapters, manufacturers' articles, predictive modelling and simulation studies, editorials, letters to the editor, workshop summaries, and online training courses.

2.2.2. Information sources

Medline (using OvidSP), the Cumulative Index to Nursing and Allied Health Literature (CINAHL) (using EBSCOhost) and PubMed were searched.

2.2.3. Search strategy

The search strategy identified records where titles, abstracts or keywords included terms for "child" AND "hearing aid" AND "cochlear implant". Strategies were developed through team discussion and included alternative phrasing for each term. An example search strategy (Medline) is shown in Table 2.1. The search results were exported into EndNote, and duplicates removed. The remaining records were exported into Excel. Initial searches were conducted in April 2019, and further update searches were conducted in October 2019 and September 2020. All records were assigned a study code at this point, to enable tracking them through the study selection process.

Table 2.1: Medline search strategy

Search #	Ovid Medline Search term
1	child*.ab,ti.
2	paediatric.ab,ti
3	pediatric.ab,ti
4	CHILD/
5	ADOLESCENT/
6	amplif*.ab,ti
7	"hearing aid*".ab,ti.
8	HEARING AIDS/
9	"cochlea* implant*".ab,ti.
10	"cochlea* prosth*".ab,ti.
11	COCHLEAR IMPLANTS/
12	COCHLEAR IMPLANTATION/
13	1 OR 2 OR 3 OR 4 OR 5
14	6 OR 7 OR 8
15	9 OR 10 OR 11 OR 12
16	13 AND 14 AND 15

2.2.4. Selection of sources of evidence

Study selection was based on the PICOS framework (Population, Intervention, Comparison, Outcome and Study type). First, all titles and abstracts were screened by the student investigator and, independently, one of two colleagues. Any discrepancies regarding inclusion or exclusion were resolved through discussion. Where no consensus could be reached, the final decision regarding inclusion was made by a third reviewer, with the majority verdict being accepted. If there was insufficient information in the title and abstract to establish whether a study met the eligibility criteria, it was passed for full-text screening. The same PICOS framework was applied at the full-text screening stage. Study types included: a) between groups studies, where a group of children with severe hearing-loss using only acoustic HA(s) were compared to another group of children using at least one CI, or HA users with a wider range of hearing-loss were compared to a group of CI users for the purpose of determining the cusp of candidature; and b) repeated measures longitudinal studies, where children with severe hearing-loss were assessed both before implantation using only acoustic HA(s) and again after implantation using at least one CI.

2.2.5. Data charting process

A data-charting tool was developed and data from all eligible studies charted by the student investigator then verified by one of four co-reviewers. Any disagreements were resolved through discussion.

Children were characterised by where they had been recruited from, cognitive function, communication mode, additional needs, age at assessment, age at

diagnosis of hearing loss, age at first HA fitting, and age at first cochlear implantation.

Intervention data extraction included the number of children in the HA group using unilateral or bilateral HAs, and the number of children in the CI group(s) who used unilateral, bilateral, bimodal, or short-array implants for EAS in either ear.

Outcome measures used in each comparison were categorised into the following broad categories: speech perception, speech and language, quality of life, psychosocial, sound localisation, listening fatigue, balance, educational, tinnitus, and music perception. Within the speech perception category, outcome measures were classified into phoneme, word, sentence and supra-segmental speech perception, and into categories of tests administered in quiet, or in background noise, extracting the type of noise and signal to noise ratio used. Within the speech and language category, outcome measures were classified into phonological skill, receptive language, expressive language, speech production, voice, communication and pragmatics, and generalised auditory performance. To characterise the studies, we extracted participants' country of residence, publication year, title, objectives, and whether one group of children had been compared before and after cochlear implantation, or two or more separate groups of children had been compared. Figure 2.1 summarises the selection of studies for data extraction.

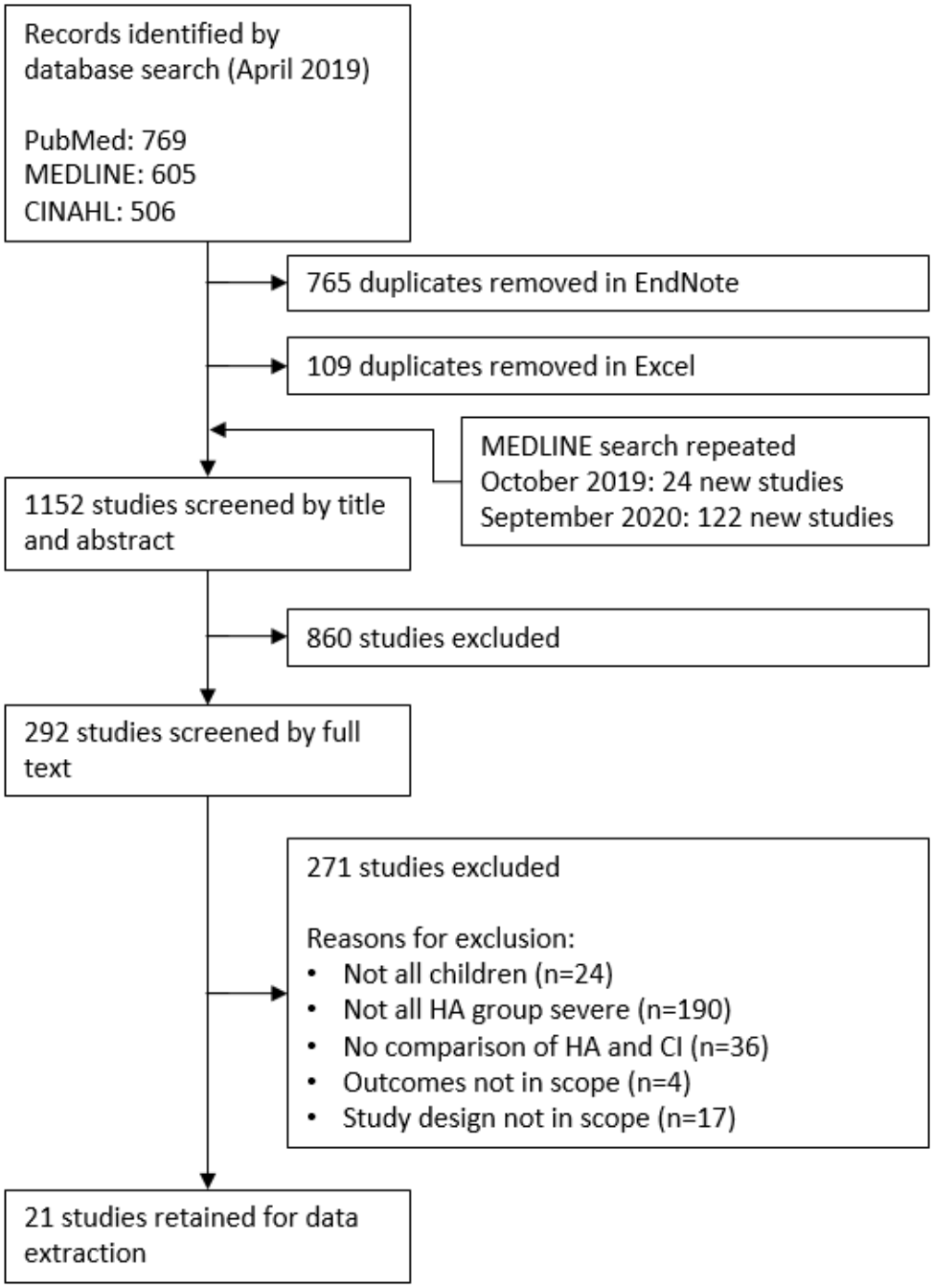


Figure 2.1: Study selection flow diagram
Flow diagram of record identification and selection.

2.2.6. Charting results

Data were extracted into Excel to create summary tables and figures. These included participant characteristics, the interventions studied, outcome

measures reported, and study design. Speech perception and speech and language outcome measures were presented in tables, classified into sub-categories so that comparisons could easily be made between studies, and gaps in the literature visualised. Outcomes measured in fewer studies, such as spatial listening, were described in the text. The availability of group or individual data, and whether statistical comparisons had been made between HA and CI users were charted. Records were categorised by participants' country of residence, publication year, and study type and size. To identify gaps in knowledge, results were synthesised across participant, intervention, outcome measures, and study type groups. It was beyond the remit of a scoping review to conduct evidence synthesis or appraisal (Arksey & O'Malley, 2005), therefore these were not performed.

2.2.7. Stakeholder consultation

Arksey & O'Malley (2005) suggest an optional stage of the scoping review methodology of seeking stakeholder feedback on the results of the review. To this end, nine experts from range of relevant professions were approached to comment on a full manuscript of preliminary findings that included 18 studies published up to October 2019. They were asked to comment on the appropriateness of the interpretation of the data, the real-world relevance of the findings, discussion points they felt should be added, and suggestions for future research that had not been identified. Responses were received from two stakeholders, one with expertise in researching outcomes in children using HAs and CIs who also works for a government agency advising on the

commissioning of healthcare services, and the second a Paediatric Audiologist with expertise in HA fitting and CI referral.

2.3. RESULTS

The following sections catalogue the records in terms of the children who participated, interventions studied, outcomes and outcome measures used, data and analyses of these outcomes, and study designs.

2.3.1. Participants

Table 2.2 catalogues key characteristics of the children studied, including the inclusion or exclusion of children with additional difficulties, onset of hearing loss, where children were recruited from, and descriptions of their communication mode and cognitive function. Most studies excluded children with additional difficulties. Exceptions were Wong et al., (2017) where children with and without additional needs were recruited into a population-based cohort study, and Trevisi et al., (2016) who studied children with CHARGE syndrome, all of whom had additional difficulties. Most studies that reported cognitive function represented children with typical non-verbal intelligence. The children with CHARGE syndrome studied by Trevisi et al., (2016) communicated using a variety of spoken and signed methods. Otherwise, studies were of children who primarily used spoken language. Most children were reported to have had congenital or early-onset hearing loss, although it was usually not possible to distinguish congenital severe or

profound losses from congenital hearing loss of a milder degree that later progressed to severe or profound levels.

Table 2.2: Participant characteristics

Author (Publication year)	Additional Difficulties		Onset of hearing loss	Recruited from	Average better ear unaided thresholds of HA users (dB HL)	Communication mode	Cognitive function
	Yes	No					
(Baudonck et al., 2010)			All congenital	University	All HA users 70 - 90 in the better-hearing ear	All used Dutch oral communication	All had minimal intelligence quotient of 80.
(Baudonck et al., 2011)			All prelingual	University	HA group mean = 83.33. No range stated	All enrolled in oral/aural rehabilitation programs.	Normal non-verbal intelligence.
(Ching & Cupples, 2015)			All under 3 years	Population based cohort (LOCHI study).	Average in the better-hearing ear >60 and described as “severe”	English as primary form of communication, with or without sign.	Cognitive ability at or slightly above age-appropriate levels.
(Eriks-Brophy et al., 2013)			All before 6 months.	AVT programmes	Participants HT28 = 73.3, HT01 = 70, HT17 = 63.3, HT07 = 85, HT29 = 61.7	All used English on a regular basis and all children enrolled in AVT programmes.	NS
(Fitzpatrick et al., 2012)			Known or presumed under 3 years.	Children’s Hospital	Group pure tone average 68.7 (SD=8.5)	More than 90% enrolled in spoken language rehabilitation programmes.	Non-verbal intelligence in the average range.

(Hammer & Coene, 2016)			NS	Schools for deaf children and an Audiology programme	Group mean of 75 for eligible sub-group of 4-year-old HA users	All monolingual speakers of Dutch, using auditory / oral communication.	NS
(Jallu et al., 2019)			NS	Government medical college	Described as "severe"	NS	Children with cognitive delay excluded.
(Kawar et al., 2019)			Prelingual bilateral SNHL	Deaf and hard of hearing centres under Ministry of Education	Described as "severe"	Oral communication in Arabic, some exposed to sign language but none used this regularly.	No learning, behavioural or developmental delay
(Leigh et al., 2011; 2016)			HA group: NS; CI group: all prelingual.	Schools, early intervention centres and a CI centre.	Sub-group in group comparison described as "severe" (2011). Included HA users with wide range of hearing impairment for regression analyses (2011, 2016).	All English as a primary language, minority of participants total communication.	Normal to borderline cognitive status (not > 1SD from the mean).
(Meister et al., 2015)			NS	Audiology Centres.	All in sub-group classed as "severe" >60 and ≤80	NS	NS

(Most & Peled, 2007)			All prelingual.	NS	Sub-group average = 77 (SD=4.96)	All used spoken language as main form of communication.	No reading difficulties reported by teachers.
(Rezaei et al., 2017)			CI: 75% congenital, 25% prelingual. HA: 70.83% congenital, 29.16% prelingual	NS	Group mean = 88.33	NS	Nonverbal IQ within the normal range
(Trevisi et al., 2016)			NS	Audiology Centres.	Participants 20, 24, 26 and 29 met ASHA definition of severe	Some verbal, some sign, some verbal with sign, some very limited communication abilities.	Heterogeneous and often severe disabilities including intellectual delay.
(Wong et al., 2017)			NS	Population based cohort (LOCHI study).	All individuals within range 61 - 80	Majority spoken language, some spoken language with sign, one child sign only, some unknown.	Severe HA users nonverbal IQ = -.027(SD=1.18); CI group: 0.024(1.12) (Mean Z scores).
(Lovett et al., 2015)			NS	Hospitals, educational services and charities.	Included HA users with wide range of hearing impairment for regression analyses	All learning spoken English as a first or bilingual language.	Nonverbal IQ whole HA group = 104.7 (SD=12.1); CI group = 107.5 (SD=10.4).

(Gantz et al., 2016)	NS	NS	N=1 congenital, N=1 diagnosed aged 4 years, N=1 unknown	University based hospital centre.	Participants: 2 = 90.00; 4 = 92.50; 5 = 85.00	All used English as the primary spoken language and enrolled in programmes with an emphasis on spoken language.	NS
(Gratacap et al., 2015)			N=6 congenital, N=1 fluctuating	CI centre.	Participants: BA = 87; CM = 71; PJ = 88; AA = 95; NA = 95; JA = 87; LM = 93	All received speech therapy.	NS
(Meredith et al., 2017)	NS	NS	All post-lingual progressive	CI centre.	Participants: 3 = 75.00; 4 = 88.75; 5 = 82.5; 6 = 87.50; 8 = 87.5	Normal early speech and language development by parent report.	NS
(Park, Teagle, et al., 2019)	NS	NS	NS	CI centre at university hospital.	Arm 1 Participants: 3 = 82.5; 6 = 95; 7 = 82.5; 8 = 83.75; 19 = 91.25; 20 = 95	NS	NS
(Tzifa & Hanvey, 2013)			N=2 congenital, N=2 progressive, N=1 acquired	Hospital CI centre.	Participants: 1 = 76.25; 2 = 66.25; 3 = 68.75; 4 = 66.25; 6 = 77.5	Use of spoken language implied in the text.	NS

Key to Table 2.2: Additional difficulties: Did the study include children with known additional difficulties that might affect listening and or language development; AVT = Auditory Verbal Therapy; HA = hearing aid; CI = cochlear implant; IQ = intelligence quotient; LOCHI = Longitudinal Outcomes of Children with Hearing Impairment Study; N = number of participants; NS = not stated; SD = standard deviation; SNHL = sensorineural hearing loss. Individual participants' unaided thresholds averaged over 0.5 to 4 kHz.

Figure 2.2 illustrates participants' ages at intervention and assessment, where this information was available. For between groups studies, age at first HA fitting for the HA group and age at first implantation for the CI group are plotted side-by-side. For the repeated measures studies, only age at first implantation is plotted. Age at HA fitting ranged from a few months to 7 years. Age at first implantation ranged from under 1 year to 16 years of age, with the children in repeated measures studies typically implanted later than those from between groups studies. Ages at assessment ranged from 2 to 17 years. Less discrepancy in age at assessment was seen between the study design types, reflecting the shorter duration of CI use typically experienced by children in repeated measures studies. Some studies pooled data for the CI and HA groups by age at first hearing aid fitting (Ching & Cupples, 2015; Eriks-Brophy et al., 2013) or age at assessment (Eriks-Brophy et al., 2013; Kawar et al., 2019; Most & Peled, 2007). These are plotted as HA group data, with dashed lines indicating the range, where available. Baudonck et al. (2010) did not report a mean or distribution for age at first HA fitting but reported that their HA group all received their first device before two years of age. Kawar et al. (2019) reported that all children studied had been fitted with some device by the age of 1 year. All other missing plots shows that ages at intervention or assessment were not available from the article (Trevisi et al., 2016).

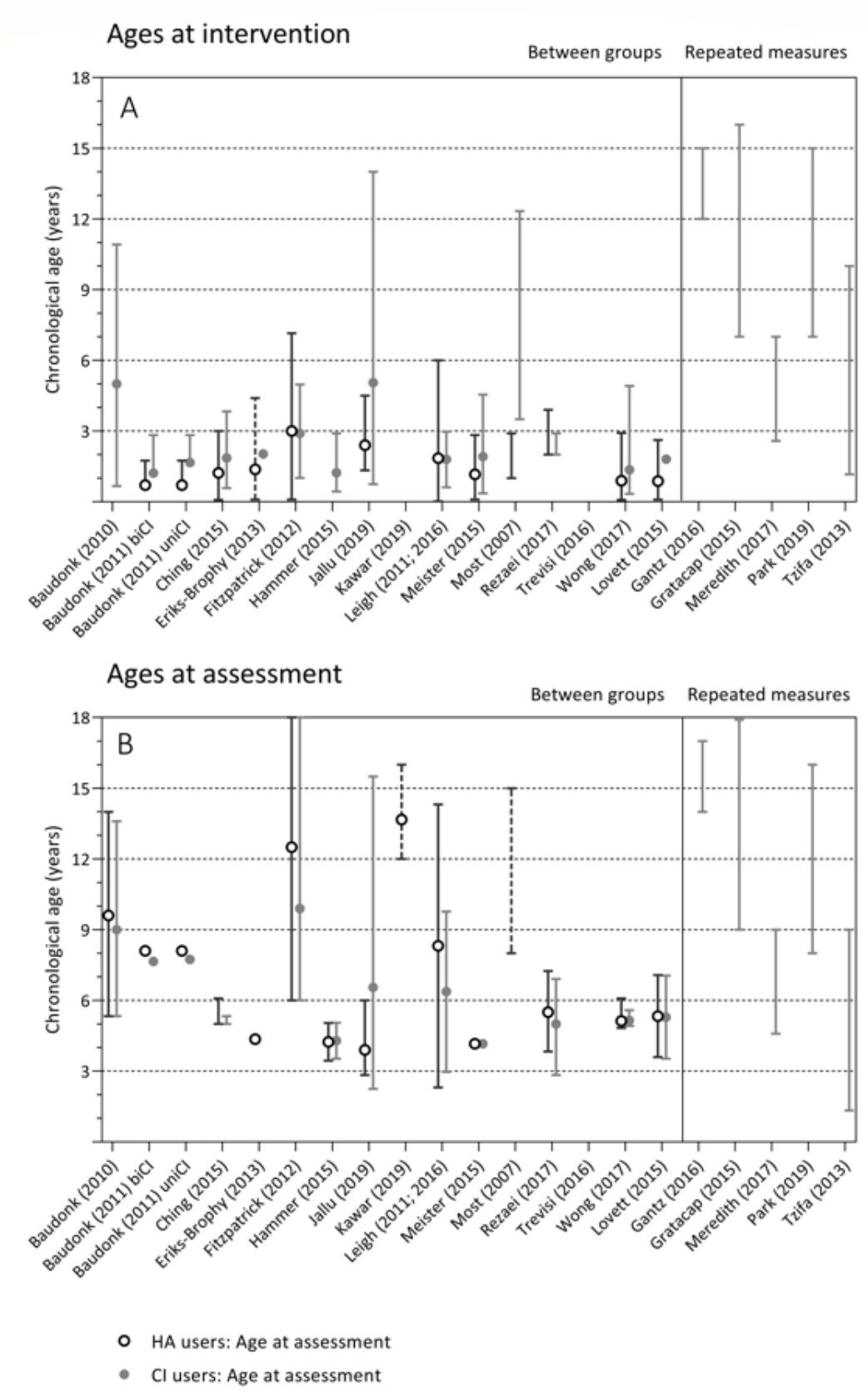


Figure 2.2: Ages at intervention and assessment

Comparison of children's age at intervention and assessment in the included studies. 'Between-groups' and 'repeated measure' studies are shown on the

left- and right-hand side of the vertical line, respectively. Means and ranges are plotted where available in manuscripts or provided via personal communication with the authors. If no range was stated but a mean and standard deviation was available, the distribution is plotted as the mean plus or minus two standard deviations, unless a more accurate upper or lower limit could be inferred from the text. For group comparison studies ages are plotted for HA and CI users separately where available. Where age data for sub-groups of HA users with severe hearing loss were not available, means and distributions are shown for the whole HA group. Where CI and HA data were pooled for ages at first HA fitting (Ching & Cupples, 2015; Eriks-Brophy et al., 2013) or age at assessment (Eriks-Brophy et al., 2013; Kawar et al., 2019; Most & Peled, 2007) these are plotted as HA group data and bars indicating the ranges, where available, are dashed. Studies with missing data did not report intervention and assessment age information.

2.3.2. Interventions

Most children using CIs were implanted unilaterally (Table 2.3), whether with a standard CI or shorter electrode array designed to provide EAS. Five records did not state whether children received one or two CIs. Baudonck et al. (2011) compared their HA group to one group of unilaterally implanted children, and to a separate group of bilaterally implanted children.

2.3.3. Outcomes

Table 2.3 provides an overview of the outcomes that were assessed. Speech perception and speech and language outcome measures were the most frequently studied domains; however data on quality of life, psychosocial outcomes, sound-source localisation and educational outcomes were also available.

Table 2.3: Data map of study design, size, interventions and analyses available for different outcome domains

First author and publication year	Country	Study Type	CI fitting	CI group size	Severe HA group size	Speech perception	Speech & language	Quality of Life	Psychosocial	Sound localization	Listening fatigue	Balance	Educational	Tinnitus	Music perception
Baudonk (2010)	Belgium	BG	NS	N = 29	N = 15		G S								
Baudonk (2011)	Belgium	BG	biCI	N = 13	N = 10		G S								
		BG	uniCI	N = 14	N = 10		G S								
Ching (2015)	Australia	BG	biCI	N = 20	N = 13		G S								
			Bim	N = 10											
Eriks-Brophy (2013)	Canada	BG	uniCI	N = 15	N = 5		I								
Fitzpatrick (2012)	Canada	BG	uniCI	N = 21	N = 20	G S	G S						G S		
Hammer (2015)	Belgium	BG	Bim	N = 15	N = 9		G S								
Jallu (2019)	India	BG	NS	N = 15	N = 13		I								
Kawar (2019)	Israel	BG	NS	N = 19	N = 27		G S								
Leigh (2011)	Australia	BG & RS	uniCI	N = 75	N = 21	G S									
Leigh (2016)			biCI	N = 5		R									
Meister (2015)	Germany	BG	uniCI	N = 38	N = 14		G S								
Most (2007)	Israel	BG	NS	N = 10	N = 10	G S									
Rezaei (2017)	Iran	BG	NS	N = 15	N = 15		G S								
Trevisi (2016)	Italy	BG	NS	N = 7	N = 4		I								
Wong (2017)	Australia	BG	NS	N = 110	N = 54		G S		G S						

Lovett (2015)	U.K.	RS	biCI	N = 28	NS[1]	R	I[2]	I[2]		I[2]		
Gantz (2016)	U.S.A.	RM	uniEAS	N = 3		I S	I S			I		I S
Gratacap (2015)	France	RM	uniCI[3]	N = 7		I						
Meredith (2017)	U.S.A.	RM	uniCI[4]	N = 5		I						
Park (2019)	U.S.A.	RM	UniEAS	N = 6		I						
Tzifa (2013)	U.K.	RM[5]	uniCI biCI	N = 3 N=2			I					

Key to Table 2.3: CI = multi-channel cochlear implant; uniCI = unilateral CI; biCI = bilateral CI; Bim = bimodal; NS = not specified; uniEAS = unilateral electric-acoustic cochlear implant; BG = between groups study; RS = regression study; RM = repeated measures study; G = group data available; I = Individual data points available, numerical and/or graphical; S = statistical comparison(s) made, either between two groups or before and after CI; R = Regression analyses. In the outcome columns, shaded cells indicate that an outcome was measured during the study. White cells indicate that an outcome was not measured. [1]Up to N=43 children using HAs with mild to profound hearing impairment were included in the regression analyses, however N for children with severe hearing impairment was not stated. [2]These data are available from Supplemental Digital Content 2 (http://links.lww.com/EANDH/A165). [3]Children across the wider study received either bilateral CIs, bimodal fitting or unilateral CIs. Fitting cannot be determined for the eligible children with severe hearing loss; however they were all assessed post-operatively via one CI used alone. [4]Children across the wider study received either bilateral CIs or unilateral CIs. Fitting cannot be determined for the eligible children with severe hearing loss; however they were all assessed post-operatively via one CI used alone. [5]Other outcome areas were assessed pre-operatively for some participants but are not included as they were not repeated post-operatively.

Speech perception and speech and language outcome measures are catalogued in detail in Tables 2.4 and 2.5 respectively. In terms of speech perception, most assessments were conducted in quiet (Table 2.4). Fewer studies addressed speech perception in background noise and presentation levels, type of noise, and signal to noise ratios differed. The speech materials used also varied and included phonemes, words, sentences, and supra-segmental features.

Table 2.4: Speech perception outcome measures

	Speech perception in quiet (Presentation level)				Speech perception in noise (Presentation level; Signal to noise ratio)		
Study	**Phonemes**	**Words**	**Sentences**	**Supra-segmental features**	**Phonemes**	**Words**	**Sentences**
Fitzpatrick (2012)	PBK (70 dB SPL)	PBK (70 dB SPL)	HINT-C (70 dB SPL)	-	-	-	HINT-C (+10 dB) HINT-C (+5 dB)
Leigh (2011, 2016)	PBK or CNC (65 dB SPL)	PBK or CNC (65 dB SPL)	BKB (65 dB SPL)	-	-	-	-
Most (2007)	-	-	-	WPPT, HeSPAC, MPT	-	-	-
Lovett (2015)	-	CAPT (50 dB A) CCT (NS)[1]	-	-	-	ATT (pink noise) ATT (babble) (Word level NS; noise levels adaptive)	-
Gantz (2016)	-	CNC (60 dB A)	-	-	-	-	-
Gratacap (2015)	-	-	-	-	-	Fournier or Saussus-Boorsma Lists (65 dB HL; +6 dB SNR)	-
Park (2019)	-	CNC (60 dB A)	-	-	-	-	Baby Bio (60 dB(A); +5 dB)
Meredith (2017)	LNT (45 dB HL) PBK (45 dB HL)	LNT (45 dB HL) PBK (45 dB HL)	-	-	-	-	-

Key to Table 2.4: ATT = IHR-McCormick Automated Toy Discrimination Test; CAPT = Chear Auditory Perception Test; CCT = Consonant Confusion Task; CNC = Consonant-Nucleus-Consonant test; HeSPAC = Hebrew Speech Pattern Contrasts (Intonation and Pattern Contrasts sub-tests); HINT-C = Hearing in Noise Test for Children; MPT = Minimal Pairs Test (extended version); PBK = Phonetically Balanced Kindergarten Test; WPPT = Word Pattern Perception Test; NS = not stated. Presentation levels are described using the units from the original manuscripts.
[1]These data are available from Supplemental Digital Content 2 (http://links.lww.com/EANDH/A165).

Various outcome measures were used to assess speech and language outcome skills (Table 2.5). Speech production was the most frequently assessed speech and language outcome, reported in seven articles. Expressive language was the next most frequently assessed speech and language outcome, reported in five. Phonological skills, receptive language comprehension, and proxy reports of general auditory performance were each reported in more than one article. Only one study assessed voice (Baudonck et al., 2011).

Quality of life scores for 38 individuals using HAs, some of whom had severe hearing-loss, are plotted in a scatterplot alongside a histogram of scores from 22 children with BiCIs in the Supplemental Digital Content 2 file of Lovett et al., (2015). Psychosocial outcomes were reported in one article only (Wong et al., 2017). Personal and group interaction and social behaviours were assessed with the social sub-scale of the Child Development Inventory. The Strengths and Difficulties Questionnaire (Goodman, 1997) was used to assess emotion, conduct, hyper-activity, and peer problems.

Table 2.5: Speech and language outcome measures

Study	Phonological skills	Receptive language / comprehension	Expressive language	Speech production	Voice	Communication / Pragmatics	Auditory Performance
Baudonk (2010)	-	-	-	Consonant production[1]	-	-	-
Baudonk (2011)	-	-	-	Intelligibility	PESP[2]	-	-
Ching (2015)	CTOPP: sound matching	PPVT-4	-	-	-	-	-
Eriks-Brophy (2013)	-	-	-	GFTA KLPA-2	-	-	-
Fitzpatrick (2012)	CTOPP: memory and analysis and rapid naming	CELF: Core language score PPVT-III	CELF: Core language score	GFTA	-	-	-
Hammer (2015)	-	-	Finite verb morphology[3]	-	-	-	-
Jallu (2019)	-	REELS	REELS	-	-	-	CAP
Kawar (2019)			Morpho-syntactic error; Complex sentences; MSAE				
Meister (2015)	-	-	-	-	-	-	FAPCI
Rezaei (2017)	-	-	-	Intelligibility	-	-	-
Lovett (2015)	-	CELF: Standard score[4] PLS-4: Standard score[4]	CELF: Standard score[4] PLS-4: Standard score[4]	-	-	-	-
Gantz (2016)	-	-	CASL	GFTA	-	-	-
Trevisi (2016)	-	-	MSLD	-	-	-	APP
Tzifa (2013)	-	-	-	SIR	-	-	CAP
Wong (2017)	-	PLS-4 (expressive communication)	PLS-4 (auditory comprehension)	-	-	-	PEACH

Key to Table 2.5: APP = Auditory Perceptive Performance; CAP = Categories of Auditory Performance; CASL = Comprehensive assessment of spoken language; CELF = Clinical Evaluations of Language Fundamentals Test; CTOPP = Comprehensive Test of Phonological Processing; FAPCI = Functioning After Paediatric Cochlear Implantation questionnaire [in German]. MSAE = Modern Standard Arabic Expressions assessment; MSLD = Major Stages of Language Development, modified version; PEACH = Parents Evaluation of Aural/Oral Performance of Children; PESP = Perceptual evaluation of speech production; PLS-4 = Preschool Language Scale Fourth Edition; PPVT = Peabody Picture Vocabulary Test; REELS = Receptive-expressive emerging language scale.

[1] Consonant errors, distortions, substitutions and omissions, final consonant deletion, cluster reductions, liquid gliding, stopping, devoicing, assimilation of sounds, substitutions of /n/ by /m/ and of /s/ by /ʃ/

[2] Grade, roughness, breathiness, astenicity, strain, instability, hypernasality, hyponasality and cul-de-sac.

[3] Mean length of utterance, finite verb production, Subject-verb agreement errors / omissions.

[4]These data are available from Supplemental Digital Content 2 (http://links.lww.com/EANDH/A165).

Regarding spatial listening, Lovett et al. (2015) also presented left-right discrimination and sound-source localisation results. Left-right discrimination was assessed using loudspeakers situated at 30 degrees to the left and to the right of the children. Sound-source localisation was assessed using 5 loudspeakers spaced at 30 degree intervals from -60 to +60 degrees azimuth. Localisation accuracy before and after implantation were reported for three eligible individuals in one study only (Gantz et al., 2016), using the Everyday Sounds Localization Test (Dunn et al., 2005), presented at 60 dB(A) from an array of eight loudspeakers arranged in an arc of approximately 108 degrees. Accuracy was determined by calculating the average root mean square error in degrees.

The only educational outcome assessed was literacy, which was reported in two studies. Fitzpatrick et al. (2012) assessed word and pseudo-word reading skills via the Wechsler Individual Achievement Test version 2 (Wechsler, 2005). Outcomes for HA and CI users were also compared on the Gray Silent Reading Test (Wiederholt & Blalock, 2000) and the spelling sub-test of the Peabody Individual Achievement Test (revised) (Markwardt, 2012). Pre- and post-implantation scores on the Woodcock Reading Mastery Test (Woodcock, 1998) were reported by Gantz et al. (2016).

2.3.4. Gaps in the evidence base

Children with additional needs, older children, teenagers, and those using signed communication were sparsely represented in the eligible articles. Other notable gaps were the absence of any comparisons between children using CIs to children with severe hearing loss using HAs related to listening fatigue,

balance, tinnitus, and music perception (Table 2.3). There were also notable gaps in outcomes related to spatial hearing, as no studies reported speech perception in spatially separated noise or spatial release from masking. Within educational outcomes, there were no comparisons of numeracy, or achievement in examinations. Tables 2.4 and 2.5 reveal that other specific aspects of speech perception and speech and language development such as phoneme perception in background noise and pragmatic language skills were also not assessed.

Several further outcomes were measured in only one or two studies. These included quality of life, psychosocial outcomes, sound localisation, perception of supra-segmental features, word or sentence perception in noise, phonological skills, and voice. By cross-referencing Figure 2.2 and Tables 2.2 -2.5 it is possible to identify the populations and interventions involved with the measurement of these sparsely represented outcomes. For example, psychosocial outcomes were only assessed by Wong and colleagues (Wong et al., 2017), in large groups of both CI users and HA users with severe hearing loss, all under approximately 6 years of age. Therefore, there is a gap in our knowledge regarding comparison of psychosocial outcomes in older children with severe hearing loss using HAs and their implanted peers.

Literacy was compared between a group of children with severe hearing loss and HAs to a unilaterally implanted CI group (Fitzpatrick et al., 2012). It was also measured before and after implantation for a small sub-set of individuals with severe hearing loss within a larger study of children who received short electrode arrays to provide electric-acoustic stimulation (Gantz et al., 2016). Figure 2.2 and Table 2.2 show that most children from both studies were

known or presumed to have had hearing loss from an early age. However, they received their CIs later on average than is current standard practice. This is likely because of limited availability of neonatal hearing screening for the cohort of children assessed by Fitzpatrick et al. and the considerable low-frequency residual hearing present in the children studied by Gantz et al. Therefore, there is a gap in the knowledge regarding both literacy and educational attainment for children with severe hearing-loss using HAs compared to children who received early identification and bilateral intervention, with either two CIs or bimodal fitting.

While word perception in quiet was the most frequently assessed speech perception outcome, cross-referencing Table 2.4 with Table 2.3 revealed that only Lovett et al. (2015) reported this outcome for a group of children who all used bilateral CIs. Meredith et al. (2017) reported repeated measures data for six children receiving a unilateral CI. Fitzpatrick et al. (2012) presented group comparisons of only unilateral CI users and most children studied by Leigh et al. (2011; 2016) wore unilateral CIs. Last, Gantz et al. (2016) reported before and after results for children receiving unilateral EAS implants. Therefore, there is a gap in our knowledge regarding word perception in quiet for children using bimodal fitting or bilateral CIs compared to children with severe hearing loss using only HAs, especially for those older than 7 years. Similarly, sentence perception in quiet was reported in three studies (Fitzpatrick et al., 2012; Leigh et al., 2011; Leigh et al., 2016) and in noise by Fitzpatrick et al. (2012); however cross-referencing Table 2.4 with Table 2.3 revealed that between these studies, only five bilaterally implanted children were represented.

2.4. DISCUSSION

Many studies reported outcomes for children using HAs or CIs in isolation over the time period of the literature search. A small minority directly compared children with severe hearing-loss with HAs to children using CIs, an important form of evidence upon which to base clinical guidance. By cataloguing what is known and what is not, this study provided a map of data available to support future meta-analyses and evidence syntheses, aid researchers in planning future studies, and inform families of deaf children, policy makers, and practitioners.

2.4.1. Feasibility of a systematic review

This chapter did not catalogue the outcomes of any statistical comparisons made within the studies. However, the results can be used to determine whether there is sufficient similarity in how outcomes have been assessed to warrant a formal synthesis of evidence being conducted in a subsequent review. The data map (Table 2.3) illustrates that few outcomes have been assessed by multiple studies, limiting the potential scope for such syntheses. Outcomes such as quality of life, psychosocial outcomes, fatigue, balance, tinnitus, and music perception were only reported in one eligible article or none. Localisation accuracy and literacy were both assessed in two articles but, for both outcomes, one study was a repeated measures design and the other a between groups design. Additionally, localisation test methods differed between the two studies in loudspeaker number and separation, and the two

studies were further distinguished by reporting data from different interventions (BiCIs in Lovett et al., (2015); EAS in Gantz et al., (2016)). Hence for these outcomes there are few articles available, and significant methodological differences between studies that limit the potential for evidence synthesis. It is possible that a search strategy designed for a systematic review could find additional articles, but this scoping review suggests that more research needs to be published before evidence synthesis can be meaningful. The remaining outcomes of speech perception and speech and language development are those addressed in the systematic review of group comparison studies by (de Kleijn et al., 2018).

De Kleijn et al. concluded that the heterogeneity of populations studied and inconsistency in the outcome measures used precluded meta-analysis of the comparative studies they identified. While we have identified additional group comparisons published after de Kleijn et al.'s searches (Jallu et al., 2019; Kawar et al., 2019; Meredith et al., 2017b; Park, Teagle, et al., 2019), there is still heterogeneity in the outcome measures and populations studied. We suggest that more consistent approaches to data collection, for example by developing core outcome sets or establishing other forms of international consensus on choice of outcomes, would aid future evidence syntheses (Anne et al., 2022). Greater consistency in how study populations are characterised would also be helpful. Scoping reviews such as this are complementary to systematic reviews such as that of de Kleijn et al. (2018) because they can catalogue a wider range of outcomes and study designs. In doing so, they identify gaps for further research to address and opportunities for formal meta-analyses to be conducted where there is evidence of comparable outcome data.

Despite the large number of records published since 2007 that compared outcomes for children using HAs to children using CIs, only 21 were eligible for inclusion in the review. The most common cause for exclusion was that even large studies rarely reported outcomes for children with average HTLs between 61 to 95 dB HL using HAs in isolation. Grouping these children together with children with better hearing is likely to improve the overall performance of the HA group on aural / oral outcomes, whereas grouping them with children using HAs with profound deafness will likely have the opposite effect. Some studies grouped all HA users' results together across the whole range of hearing loss. We retained these studies if correlations of outcomes with HTLs were used to estimate the average HTL associated with equivalent performance of HAs to CIs (Leigh et al., 2011; Leigh et al., 2016; Lovett et al., 2015). However, most studies where children using HAs had a wide range of losses only presented group summary data. Doing so increases the variance in the HA group data. Each of these group summary approaches decreases the likelihood of identifying differences between HA and CI users that could be relevant and important in defining the audiometric cusp of suitability for CIs. The capacity of future research to contribute to audiometric criteria might also be strengthened by characterising children in terms of device acceptance and daily device use, in addition to age at first fitting. These are known to be associated with children's outcomes (Park, Gagnon, et al., 2019; Wiseman et al., 2021) but were not charted in this review, as data were not available for any of the studies included.

2.4.2. Gaps in the evidence base

The inclusion of a broad range of outcomes enables this review to identify gaps in the evidence base for comparisons of children with average unaided hearing thresholds from 61 to 95 dB HL using HAs and children using CIs. Most studies assessed speech perception, speech production and / or language development. The differences identified in the participant characteristics, interventions, and outcome measures used in these studies may account for some discrepancies in their findings.

2.4.2.1. Gaps in the evidence base: Population

Long-term outcomes for children born profoundly deaf are influenced by age at intervention (Ching et al., 2018; Yoshinaga-Itano et al., 2010, 2018). Some between-groups comparisons, albeit a minority, included congenitally-deaf CI users with a wide range of chronological age at implantation, some as late as during their fourth year of life (Figure 2.2). This likely increased variability in CI group outcomes. Including late-implanted CI users with congenital deafness limits the relevance of these studies to determining candidature for children born with severe hearing loss or children with acquired or progressive losses, who have better access to sound via HAs during their early years. Future research in CI users with acquired or progressive losses could clarify how comparative outcomes are influenced by hearing device type, while minimising the potentially confounding effect of early auditory deprivation. There was little information on the outcomes of older children and teenagers in the studies. Children assessed via repeated measures before and after

implantation tended to be older than those in between groups comparisons (Figure 2.2) but the numbers of children with severe hearing-loss participating in repeated measures studies was small (Table 2.2). It is plausible that outcomes for HA users with severe hearing-loss and CI users might diverge during later childhood and adolescence. Therefore, studies comparing outcomes for older children and teenagers would make an important addition to the literature, and could also improve the availability of some of the missing outcomes that are easier to assess in older children and teenagers such as tinnitus, spatial release from masking, and localisation. Further research into comparative outcomes for children with additional health or learning needs is also needed, since these children were underrepresented in the literature despite representing a higher proportion of children with hearing loss than in the general population (Birman et al., 2012; Szymanski et al., 2012). No records included whole groups of children who used sign language as their primary form of communication.

2.4.2.2. Gaps in the evidence base: Interventions

This review also highlights gaps in the evidence base in terms of interventions. Simultaneous, or short-interval sequential, bilateral cochlear implantation is becoming the standard of children's care in more areas of the world (Ramsden et al., 2012; Teagle et al., 2019) however only five studies clearly stated that any biCI users were included, and seven studies did not describe whether children used one or two CIs. Only one study presented data for a HA group compared to one group of unilateral CI users and a separate group of bilateral CI users (Baudonck et al., 2011). There is a need for more studies comparing

outcomes for HA users with severe hearing loss to bilaterally implanted and bimodal CI users. Only one study reported outcomes on EAS (Gantz et al. 2016), likely because EAS is commonly used for steeply sloping losses where thresholds averaged across the frequency range often fall within the moderate range. This review did not address comparative outcomes for all children outside traditional audiometric CI candidacy. Some repeated measures studies we excluded from the current review included children whose hearing loss in the implanted ear was moderate pre-operatively, rather than severe, and who therefore did not meet our inclusion criteria. This led to the exclusion of some studies evaluating EAS in children with good low-frequency hearing.

2.4.2.3. Gaps in the evidence base: Outcomes

Data over a wide range of outcomes were charted. It is possible that the relative importance of outcomes may vary between and among children, parents, clinicians, researchers and funders. However, our stakeholder consultation indicated that each outcome domain included in this review is valid in this population and relevant to the question of comparative outcomes with HAs or CIs. It is notable that this scoping review identified similar gaps in knowledge as those identified by Bond, Mealing, et al., (2009) when synthesising evidence for the candidature of children with profound hearing loss for CIs. These gaps included measures of quality of life, educational attainment, and outcomes for children with additional needs. These gaps now exist about the candidature of children with severe hearing loss. With more residual hearing, more complex outcomes also become increasingly relevant including spatial listening, speech perception in noise, voice, communication,

pragmatic skills, and music perception. An awareness of comparative outcomes in listening fatigue, tinnitus, and balance would also provide a more holistic background against which candidature decisions could be made. While all these outcomes were investigated across the many records captured by our initial literature searches, none were eligible for inclusion as they did not include groups of HA users with severe hearing loss in isolation.

2.4.3. Stakeholder consultation

Our stakeholders confirmed that the range of outcomes included are relevant and meaningful to families of children with hearing loss. They also suggested ways in which our interpretation of the data could be improved. First, by discussing the size, number, variation in methods, and lack of standardisation of outcome measures and result reporting for studies in this field, and the implications of this for evidence synthesis. Second, they suggested more discussion of factors such as early device acceptance and daily usage on children's outcomes, in addition to the factors we had charted. Last, they alerted us to extra data available via the Digital Supplemental Content files of (Lovett et al., 2015).

2.4.4. Limitations

A limitation of this study was the financial constraints that prevented access to translation services, necessitating the exclusion of studies without full texts in English. Because of this, we may have omitted studies written in other languages that are relevant to the objectives of this review.

2.5. SUMMARY

This scoping review catalogued recent literature comparing outcomes for children with severe hearing-loss using HAs to those of children using CIs. While several studies were eligible for inclusion, there remain significant gaps in the evidence base for comparative outcomes in these groups. To address these gaps, more studies are needed that include descriptions of outcomes for HA users with severe hearing-loss in isolation from those with other degrees of hearing loss and compare the outcomes of these children to outcomes for children using CIs who have had more comparable early auditory experience. Studies using interventions such as bilateral digital HA fitting, bilateral CIs and bimodal fitting would ensure relevance to current best practice. Further research is also needed to compare a broader set of outcomes for children with severe hearing loss to children with CIs, including speech perception in complex background noise, spatial listening, quality of life, listening effort, balance, dizziness, tinnitus, voice, communication, pragmatic language skills, music perception, and educational attainment.

The results of this review informed the study design behind the rest of this thesis. Behavioural assessments of speech perception in complex background noise and spatial listening were planned. Children with additional needs were not excluded from care-giver reported outcome measure collection. Collection of data regarding tinnitus and listening fatigue was designed. Children of any age were recruited, and children using CIs were recruited from a cohort known to mostly use bilateral implants. Educational outcome data was sought from an

existing research dataset. Chapter 3 will describe the development of the study design in more detail.

CHAPTER 3: STUDY DESIGN AND COVID-19 IMPACT

3.1. INTRODUCTION

The scoping review in Chapter 2 identified several gaps in the evidence base for studies comparing children with severe hearing loss using HAs with children using CIs, and outcomes for which only sparse evidence was available. This project therefore planned to compare outcomes from children with severe hearing loss using HAs and children using CIs, recruited from a representative sample of children with hearing loss. Prior to the pandemic I aimed to produce novel evidence across several of these domains. This included measures to be recorded directly from the children including speech perception in noise, sound-source localisation, self-reported tinnitus, and self-reported need for recovery from listening. It was also to include caregiver-reported listening, tinnitus, and need for recovery outcomes along with parent satisfaction with, and child acceptance of, HAs or CIs. Families were to be recruited from whole hospital caseloads, to maximise the clinical applicability of the findings. To accurately characterise the participants, and to be able to control for potentially confounding differences between the HA and CI users, demographic, medical, and audiological data were to be collected from caregiver reports and children's hospital notes. This primary study was named "Comparative oUtcomes for children with Severe and Profound hearing loss" (CUSP). This chapter begins with a high-level overview of the CUSP study as originally designed to meet these objectives. It also introduces the secondary

project that will be presented in Chapter 6, a collaboration with the Born in Bradford cohort study.

The onset of the COVID-19 pandemic in my second year of study affected the period during which I needed to carry out recruitment and data collection. This necessitated changes to the protocol. This chapter concludes with a description of the impact of the pandemic on the study design and an overview of the adaptations that were made. For ease of reference, detailed methods for the selection of individual outcome measures used in the final study and their analyses will be provided in subsequent chapters.

3.2. OVERVIEW OF THE ORIGINAL STUDY DESIGN

Building on the scoping review and in consultation with clinicians, the Research Design Service, University of Nottingham Sponsor, and Clinical Research Network, I developed an observational study protocol that gained ethical approval[1]. Figure 3.1 gives an overview of the original project design, from eligibility screening and recruitment through to dissemination.

[1] Study title: Comparative outcomes for children with and without access to cochlear implantation. IRAS Project ID: 261824. Protocol Number: 19035. Research Ethics Committee reference: 19/EM/0302. Sponsor: University of Nottingham. Nottingham 2 Research Ethics Committee.

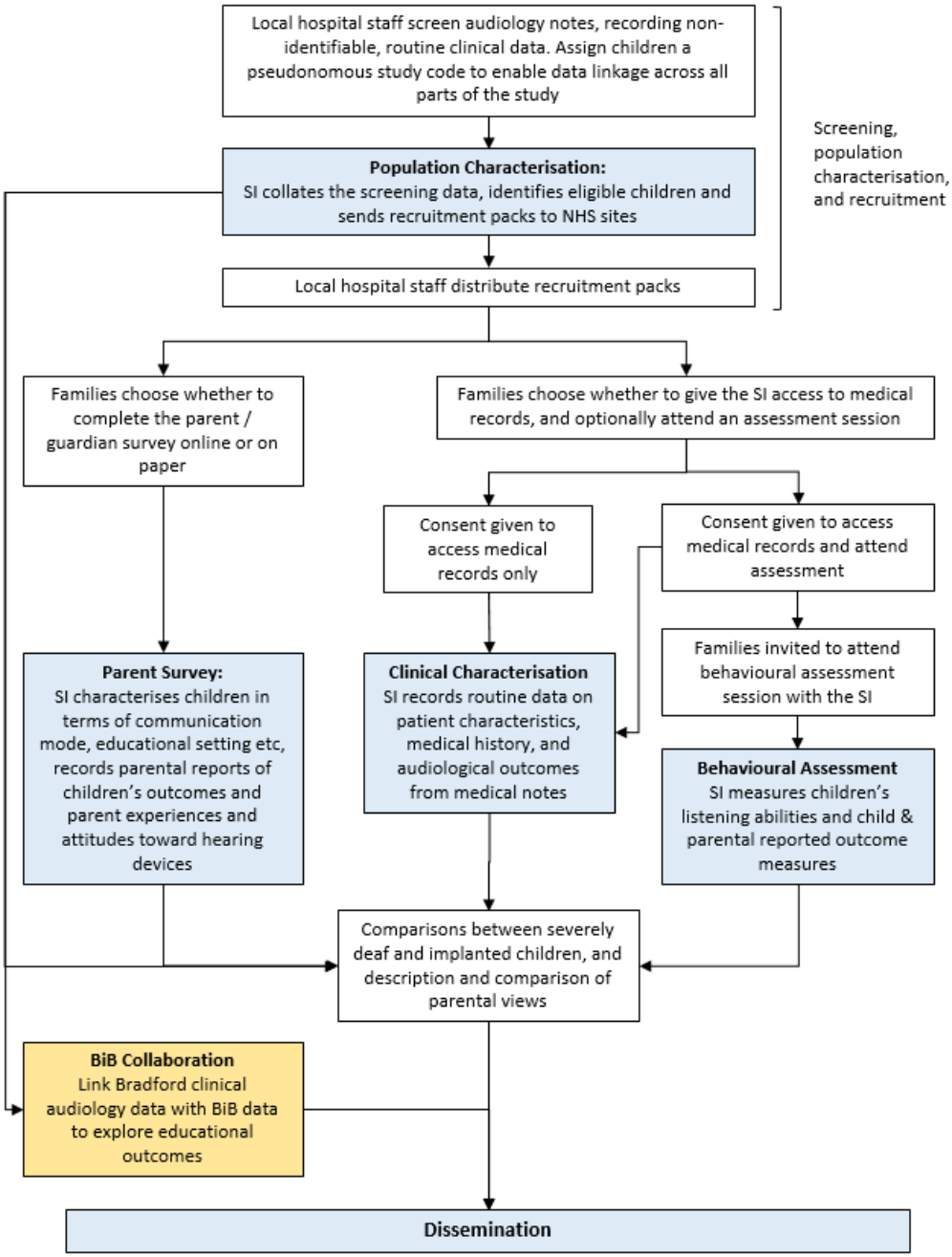


Figure 3.1. Original project design overview

SI = Student Investigator, BiB = Born in Bradford cohort study. Key stages of the CUSP study are highlighted in pale blue. The BiB collaboration is highlighted in pale yellow. Arrows indicate the flow of participants through the study and show how data collected at each stage were intended to be linked for analyses by using the pseudonomised participant identification codes.

3.3. RATIONALE FOR PRIMARY OUTCOME MEASURE SELECTION AND POWER CALCULATION

The primary outcome measure for the original study design was to be the speech reception threshold from the McCormick Toy Discrimination test in adaptive pink noise, collected in-person at behavioural assessments. This section gives the background to this choice of test, hypothesis formation, and the power calculation used to determine target sample size for the behavioural assessments.

Speech perception is the foundation upon which speech production, language, psychosocial and educational outcomes can develop and is therefore a key aspect of CI candidacy selection (Warner-Czyz et al., 2022). I therefore chose this to be the primary outcome for the CUSP study. The next step was to choose the type of speech stimulus to use and its presentation method. Adult CI candidacy is determined based on speech perception in quiet, i.e. with no background noise (NICE, 2019). However, children spend significant amounts of their time in noisy classrooms, dining-rooms, playgrounds and homes (Busch et al., 2017) and speech perception in noise has been recommended in the most recent ACIA task force guidelines for determining CI candidacy in children within a wider battery of tests (Warner-Czyz et al., 2022).

The scoping review in Chapter 2 identified that four studies published since 2007 had compared speech perception in background noise for children using HAs with average HTLs between 61 to 95 dB HL to children using CIs (Fitzpatrick et al., 2012; Gratacap et al., 2015; Lovett et al., 2015; Park, Teagle, et al., 2019). There was heterogeneity in the types of implantation assessed (full-length versus short electrode arrays, uniCI versus biCI), the type

of test stimuli used, and the findings in terms of whether HAs or CIs were associated with better outcomes. Hence more data comparing speech perception in noise for a large, representative group of children using HAs to children using biCIs would be a valuable addition to the literature.

Test choice was limited to material in the English language with an English accent. The target population was children whose HTLs were close to NICE audiometric candidacy guidance and I planned to include children with additional difficulties, who had been sparsely represented in studies found by the scoping review. I was aware that children with severe hearing loss constituted the smallest proportion of HA caseloads (CRIDE, 2021; Fortnum, Marshall, & Summerfield, 2002). Therefore, I wished to choose an outcome measure appropriate for children across a range of ages and developmental abilities, to help me to recruit enough participants and to be as inclusive as possible. Sentence perception can be influenced by a child's working memory, language development and vocabulary, as these affect their ability to guess words they might not have heard based on the context provided by other words in the sentence (Montgomery, 2000). Word perception tasks are less reliant on these skills because they include less redundant information. One English language word test with simple vocabulary and accessible to children with less-developed language skills was the McCormick Toy Discrimination Test, where children choose from a group of toys made up of similar-sounding pairs (e.g. 'cup'/'duck' and 'shoe'/'spoon') (McCormick, 1977).

In term of competing noise, speech-based signals such as the babble used by Lovett et al. (2015) and Park, Teagle et al. (2019), are more representative of real-world conversational background noise than computer-generated, steady-

state noise. However, another important consideration was the need to collect data from enough children to detect any difference between the HA and CI groups. I had already amassed data from 91 children with biCIs who had participated in the McCormick Toy Discrimination test with adaptive pink noise, collected by myself in clinic prior to beginning my PhD. The equipment I had used for that clinic was available for my research use at Bradford Royal Infirmary and an identical kit was also available at the University of Nottingham (Kitterick et al., 2011). This outcome measure would therefore allow me to focus on recruiting children using HAs, knowing a large comparator group was already available in case it was not possible to recruit enough CI users within the time available. Also, I could offer families two testing locations if I needed to recruit from a wide geographical area. Floor or ceiling effects were unlikely to arise for this test in this population (Killan et al., 2015; Lovett et al., 2015). The signal to noise ratio (SNR) scores were known to correlate with unaided HTLs (Lovett et al., 2015), and to be sensitive to improvement post-biCI for children meeting the 2009 NICE candidacy guidance (Killan et al., 2018).

Based on these studies, I hypothesised that children using HAs who were newly audiometrically-eligible for CIs under TAG566 (NICE, 2019) would have higher SNR scores than children using CIs on average. In other words, that children using CIs would be able to discriminate the words in the presence of more background noise than the children using HAs. The clinical data revealed a mean difference of 6.0 dB SNR between children using HAs under assessment for CI candidacy, referred under TAG166 (NICE, 2009), and a group of implanted children. This corresponded to an effect size of 0.8 SD.

Because the present study would be recruiting children using HAs with better hearing, who therefore might have performed more similarly to the children with CIs, the study was powered to detect an effect as small as 0.5 SD. To detect an effect of that size with alpha = 0.05 and a power of 80% required data from at least 27 children with severe hearing loss using HAs. It is not uncommon for some children to withdraw co-operation before all data are collected nor for appointments to be cancelled due to ill health or unavailability. I therefore aimed to recruit at least 35 children using HAs to the behavioural assessments. I also planned to apply secondary outcome measures to assess sound-source localisation, child-reported listening, tinnitus, and listening fatigue at these appointments. These outcome measures will not be described in detail since behavioural assessments ultimately did not take place, as will be explained in section 3.4.

No power calculations were applied to the other areas of the study. It was not known what proportion of families approached with in the survey would consent to attend behavioural assessments, nor what proportion would have children aged 5 or older who were developmentally able to complete the McCormick Toy Test. I aimed to characterise as large a cohort as possible for whole caseloads' audiometric eligibility for CIs and the parent survey, so no targets or maximum recruitment limits were set for these activities.

3.4. IMPACT OF COVID ON THE ORIGINAL CUSP STUDY DESIGN

3.4.1. Impact of COVID-19 on population characterisation

This part of the CUSP study aimed to assess the impact of TAG566 (NICE 2019) on children's eligibility for CIs, by studying whole caseloads of children

using HAs. This was to be achieved by analysing the data collected during eligibility screening to find children eligible for the parent survey and behavioural assessments. For this reason, participating children's HA services were located within reasonable distance of Bradford Royal Infirmary and Ropewalk House in Nottingham, where the test facilities had been arranged to carry out speech perception and sound-source localisation tests. Following several iterations of study design to satisfy the University of Nottingham sponsor and Health Research Authority, I gained ethical approval on 24th October 2019. Eligibility screening began at Bradford in November 2019, at Nottingham in December 2019, at Huddersfield in January 2020, and at Calderdale in March 2020. Soon afterwards the COVID-19 pandemic began to impact the study.

In spring 2020, eligibility screening was brought to an abrupt halt. All NHS Trusts reacted to COVID-19 by applying a blanket halt to any non-COVID research activity. This was frustrating because there were audiologists who could not clinically practice at this time, due to insufficient infrastructure to provide remote care, and who were not redeployed. Some sites reported this was an ideal time for their staff to carry out screening but their research departments would not allow them to. I felt fortunate that I had a pre-existing working relationship with the research department at Bradford Teaching Hospitals. I kept in close contact and they allowed me to recommence screening 2 months later. Without this, it would not have been possible for me to complete my own screening and support the other sites with recruitment within the data collection time window. It took 5 months for screening to resume at Nottingham and Huddersfield, and 8 months at Calderdale.

Ultimately sites reported that all children prescribed HAs had been screened, with the process continuing until the final month of data collection at the end of the 3-year funded period in March 2021. Results of this part of the study will be presented in Chapter 4.

3.4.2. Impact of COVID-19 on CUSP parent survey

The CUSP parent survey aimed to compare parent-reported functional listening, tinnitus, and listening fatigue outcomes for children using CIs to children using HAs with hearing close to or within CI eligibility. The survey was designed to be very inclusive, to represent the clinical population. This involved recruiting families with children of all ages whose hearing placed them on the cusp of CI eligibility, including those with and without additional needs, and families who were or were not proficient in spoken or written English. It was planned to recruit English speaking families via postal packs, with the option to complete the survey online or on paper so that digital poverty would not preclude participation. For families not proficient in English, recruitment was to take place during routine clinical appointments in the presence of an interpreter. Some recruitment packs had been distributed from Bradford and Nottingham in February 2020. Six consent forms and surveys had been returned before research activity was ceased, but no hospital note searches for clinical characterisation, or behavioural assessments, had taken place.

To resume recruitment, it was necessary for the CRN Audiologist based at the Biomedical Research Centre (BRC) in Nottingham to have time assigned to non-COVID research and physical access to BRC premises to print, collate and

distribute packs to the local sites. Hospital sites needed to have staff available to edit in children's names and addresses to the required covering letter, staff with physical access to facilities to print covering letters and post the packs, and permission to conduct non-COVID research activity. The earliest sites to recommence recruitment were Bradford and Huddersfield, in October 2020, and the latest at Calderdale in February 2021. It was possible to begin recruiting from additional HA sites and CI centres in November to December 2021. Neither screening of whole caseloads nor recruitment pack distribution were completed until the final month of the PhD's 3-year funded period in March 2021.

As well as lost time, COVID-19 caused other difficulties. Very few in-person clinical audiology appointments were taking place and there were strict limits on interactions between people and the number of people who could be together in rooms. This meant it was not feasible for recruitment packs to be handed out in clinics and very few families who needed a spoken language interpreter could be approached to participate overall. However, by March 2021 every eligible child identified had had a recruitment pack created and sent to their local site for distribution, and ultimately the caregivers of 109 children participated. The results of the CUSP survey will be presented in Chapter 5.

3.4.3. Impact of COVID-19 on CUSP clinical characterisation

When returning their survey, caregivers were given the option to also consent for the student investigator to travel to their child's local audiology department and extract data from the audiology records. This option was retained in the

reduced protocol following the onset of the pandemic and most parents who participated in the survey also gave their consent for this hospital notes search. However, a combination of factors including time-delays, travel restrictions, volume of work in survey recruitment, and my personal circumstances, meant that it was not feasible to carry out these visits. As a result, it was only possible to characterise children based on eligibility screening data and answers to questions in the parent survey. This prevented the accurate characterisation of children in terms of exact date of first diagnosis of hearing loss, first fitting of hearing devices, incidence of ear infections and middle ear effusion, and changes in HTLs over time. The implications of this for analysing and interpreting the CUSP study findings will be discussed in each relevant chapter.

3.4.4. Impact of COVID-19 on CUSP behavioural assessments

The CUSP study behavioural assessments aimed to prospectively collect word discrimination in noise and sound-source localisation data from children, so that the outcomes of children with severe hearing loss using HAs and children using CIs could be compared. They were also to include the collection of self-report measures from children direct, to most accurately represent their experiences of tinnitus and need for recovery from listening. The original CUSP study recruitment pack included information sheets for parents and eligible children describing these appointments. As the pandemic progressed it became apparent that it would not be possible to carry out enough assessments to test the number of children required for the primary outcome measure (word perception in background noise). This was due to a combination of restricted

access to test facilities, travel restrictions, bans on non-COVID in-person research activity, and personal circumstances. The information packs were therefore rewritten, removing any mention of behavioural assessments, and a protocol amendment was submitted for a reduced study design. This reduced the length of the parents' information sheet, and information sheets for children aged under 16 years were no longer needed as the study would now only collect data from caregivers via the survey. Interestingly, while this felt like a set-back, the proportion of families approached who chose to complete the survey increased following the simplification of the recruitment pack. There is therefore no chapter on behavioural outcomes within this thesis. However, suggestions for incorporating behavioural testing into future research are made in Chapter 7.

3.5. BORN IN BRADFORD COLLABORATION

The Born in Bradford (BiB) cohort study recruited families of babies born at Bradford Royal Infirmary between 2007 and 2011 to explore how medical, environmental, behavioural, and social factors affect children's health and development (Wright et al., 2013). I was aware of the BiB study prior to my PhD, as it was founded from the Trust where I was employed prior to and during my PhD data collection period. No previous BiB studies had looked at hearing loss. The impact of visual impairment on children's educational outcomes had been assessed using their dataset (Bruce et al., 2016) which suggested that a BiB collaboration might address the gap in the knowledge base identified in Chapter 2 for comparative educational outcomes for children with severe hearing loss using HAs to children with CIs. So, during my second

PhD year, a proposal to access BiB data was submitted in consultation with the BiB support team, my primary supervisor, and a statistician, in the months preceding the pandemic. The proposal was accepted by the BiB executive group just before they closed their doors to non-COVID-19 projects for a period of several months.

Parents of children in the BiB cohort had already consented for BiB research data to be linked to all routine clinical data held at Bradford Teaching Hospitals, and so no additional consent was needed for me to link the eligibility screening data or other data held in children's audiology notes to BiB research outcomes. Throughout my PhD I continued to be a part-time employee of Bradford Teaching Hospitals NHS Trust and part of the local clinical care team, meaning that I had access to children's audiology records for data extraction. An amendment to add this collaboration to the CUSP study protocol was accepted by the Health Research Authority.

The BiB data dictionaries held early years educational outcome data. Initial informal correspondence with the BiB team led me to estimate that approximately 120 children using hearing devices could have also enrolled into BiB and that a large proportion of these would have educational outcomes available. Unfortunately, the total number of children who were both enrolled in BiB and using hearing devices on the Bradford audiology and YAIS caseloads was smaller than expected and included too few children using CIs for statistical comparison with children using HAs. Nor were there enough children with severe hearing loss using HAs to form a comparator group. However, few publications on the effect of any degree of hearing loss on early years educational outcomes in the UK were identified. The richness of the data

held by BiB meant the children could be characterised well. This revealed that both the cohort demographics and outcome measures available were different to those of other UK studies linking HTLs to educational outcomes. It was therefore possible to address a gap in the knowledge base by measuring the impact of unilateral, mild, and moderate hearing loss on children's early years educational outcomes. The results will be presented in Chapter 6.

On receipt of the linked BiB datasets, I discovered that there was a discrepancy of up to several years between the dates of the audiograms collected for CUSP study eligibility screening and the dates of the educational assessments. This meant it was necessary to extract additional, older audiograms and device use data to have an accurate reflection of children's hearing at the time of their educational outcome assessments. Unfortunately, changes to working practice in response to the COVID-19 pandemic at this time meant I did not have access to audiology records, as I was working off-site without remote access to hospital IT systems. I therefore recruited the help of a specialist registrar with an interest in the topic to extract the additional audiometry data, with the support of local ENT consultants. Because of this support, the impact of the COVID-19 pandemic on this part of the study was minimal.

3.6. OVERVIEW OF THE FINAL STUDY DESIGN

The result of the protocol adaptations is shown in Figure 3.2, an overview of the final project design.

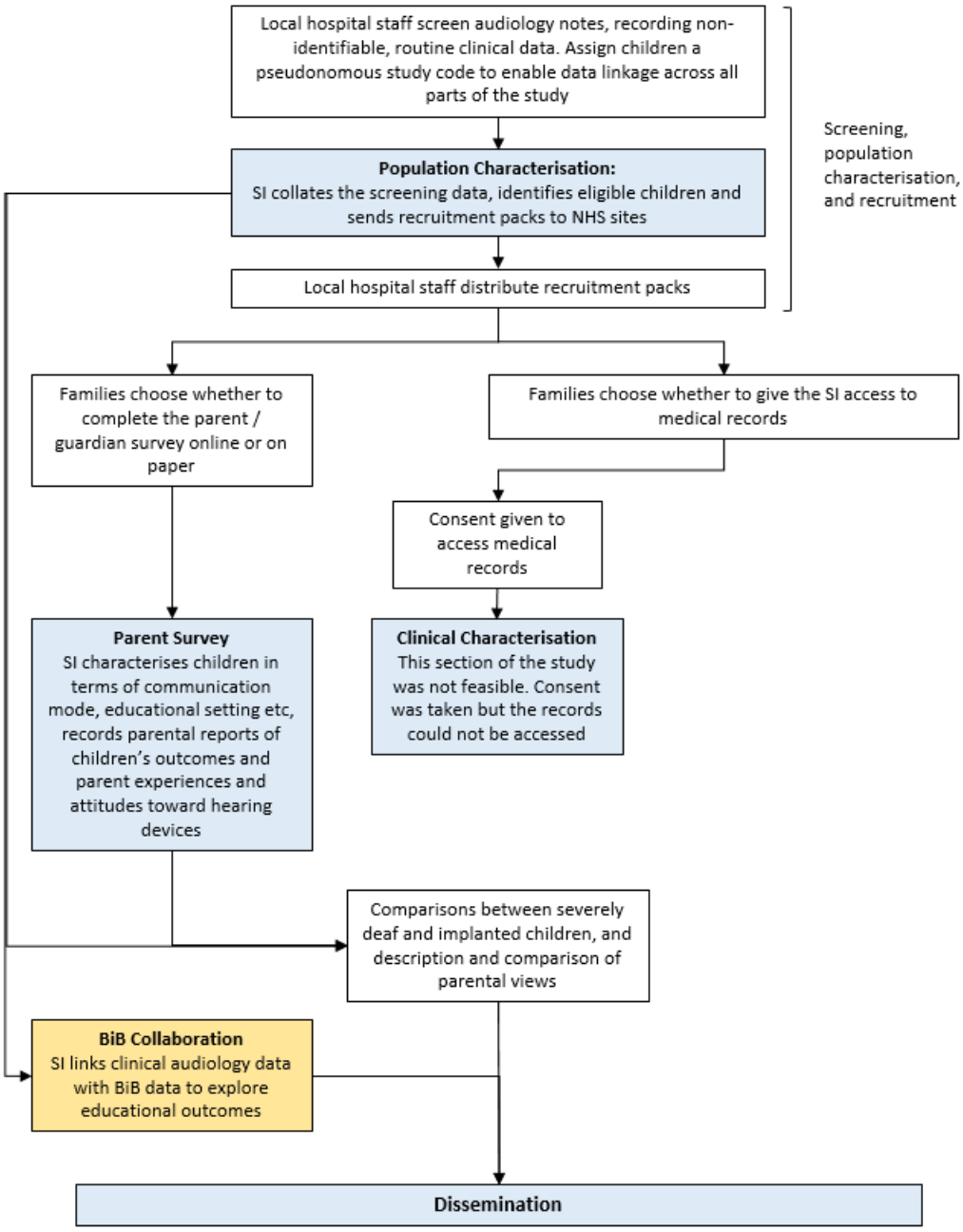


Figure 3.2: Final project design overview
SI = Student Investigator, BiB = Born in Bradford cohort study, CUSP = Comparative oUtcomes for Severely and Profoundly deaf children. Key stages remaining from the original CUSP study protocol are highlighted in pale blue, and the BiB collaboration in pale yellow. SI = Student Investigator.

CHAPTER 4: ASSESSING THE POTENTIAL IMPACT OF THE 2019 NICE GUIDANCE CHANGE BY CHARACTERISING CHILDREN WITHOUT ACCESS TO COCHLEAR IMPLANTATION

4.1. INTRODUCTION

Chapter 1 established that the revised NICE guidance TAG566 (2019) changed children's audiometric eligibility for CIs in two ways. First, the HTL level was revised from 90 dB HL to 80 dB HL. This was intended to increase the number of children eligible for cochlear implantation by including children with better hearing. Second, rather than only HTLs 2 and 4 kHz being needed to establish eligibility as in TAG166 (NICE, 2009), the new guidance stated that eligibility could be determined by HTLs at any two audiometric frequencies including 0.5, 1, 2, 3, and 4 kHz. This was intended to widen access to cochlear implantation for children where their low-frequency hearing might be worse than their mid- to high-frequency hearing. Therefore, under TAG166 (NICE, 2009) eligibility could be established by measuring four HTLs. Under TAG566 (NICE, 2019) up to ten HTLs could need to be measured to ensure that the eligibility of a child was determined definitively.

The guidance change could therefore be expected to affect two distinct groups. First were those newly eligible for CIs, with HTLs falling outside TAG166 (NICE, 2009) but within TAG566 (NICE, 2019). Second were those with missing audiometric data who were at risk of their eligibility being unknown due to the greater number of HTL measurements needed for TAG566 (NICE, 2019) compared to TAG166 (NICE, 2009). A third distinct group of HA users also exists, those who had HTLs within the prior 2009 eligibility guidance, but

who did not have access to cochlear implantation. This group was unlikely to be affected by the guidance change. However, any differences in their characteristics compared to the wider population of children using HAs might reveal barriers to accessing CIs for children in the UK that could affect the first two groups also.

It was therefore important to assess the prevalence and characteristics of children affected by TAG566 (NICE, 2019). This information would allow CI services to understand the additional children they could expect to be referred for CI candidacy assessments. Results could serve as a benchmark against which CI services could compare their actual additional referrals. It could also increase awareness among HA and CI service providers of the number and types of children whose eligibility for CIs was not known and needed to be clarified.

However, it was not possible to estimate the prevalence and characteristics of children affected based on previously published data. Two key studies were identified. The first was a large epidemiological study of children with permanent moderate, severe, or profound hearing loss (Fortnum et al., 2001; Fortnum, Marshall, & Summerfield, 2002) and the second was the Consortium for Research in Deaf Education (CRIDE) annual report (CRIDE, 2021). The relevant findings of these studies are summarised below, along with other publications that identified important issues around HAs and CI candidacy provision, including ethnicity and additional needs.

Fortnum et al (2001, 2002) carried out the most recent epidemiological study of children using HAs in the UK that collected their HTLs. This study was comprehensive, with total ascertainment of children with permanent bilateral

hearing loss of >40 dB HL averaged across 0.5, 1, 2 and 4 kHz in the UK; every case known, including children born between 1980 and 1995, was reported for every one of the 122 UK postcode areas, with data collected from both healthcare and education professionals for all but two of these areas. Of the profoundly deaf children, 22.9% had a CI. There has been significant change in clinical practice in the UK since 1998, including greater provision of CIs (BCIG, 1995, 2012, 2014; Cullington, 2021). However, internationally there is evidence that not all children have received equal access to cochlear implantation. In a USA study from 1997, a greater proportion of profoundly deaf children of White or Asian ethnicity received CIs than Hispanic or Black children (Stern et al., 2005). A later study found a narrowing but persistent racial disparity in children's CI provision in the USA (Tampio et al., 2018). Fortnum et al. (2002, 2006) found that profoundly deaf children with CIs in the UK had fewer additional disabilities and came from more affluent families than profoundly deaf children using HAs. It is not known to what extent these trends may exist in present-day CI provision in the UK. Children's age is one further factor known to influence access to CIs, because for children with congenital hearing loss, older age can be a contraindication for implantation due to the adverse effects of auditory deprivation on children's ability to benefit from CI stimulation (Nikolopoulos et al., 2004).

Current summaries of children with hearing loss are published in the CRIDE annual reports. These include all children reported to receive specialist educational support by education providers, including children with unilateral or mild losses excluded by Fortnum et al. (2001, 2002). The 2021 CRIDE report estimated that roughly 77% of profoundly deaf children in England now

have at least one CI. However, CRIDE data cannot be used to identify or estimate the number of children affected by the 2019 NICE guidance change, because children's degree of hearing loss is classified solely by BSA categories and individual HTLs are not collected.

A last consideration for children newly eligible for cochlear implantation is that the proportion of them with asymmetric hearing loss is not known. As described in Chapter 1, long-duration profound hearing loss of the worse-hearing ear risks the presence of aural preference syndrome limiting the potential of simultaneously-implanted bilateral CIs to provide optimal spatial listening skills such as speech perception in complex background noise and sound localisation. As the CRIDE report characterises children by their better-hearing ear alone, this also cannot be derived from existing datasets.

This chapter therefore aimed to assess the impact of TAG566 (NICE 2019) on children's eligibility for CIs, by studying children without access to cochlear implantation. This was to be achieved via meeting the following objectives:

(i) Document demographic and audiological characteristics of whole caseloads of children using HAs.

(ii) Calculate the prevalence of children using HAs who were: newly eligible for CIs; with unknown eligibility for CIs; and who had been previously eligible for CIs.

(iii) Document the demographic and audiological characteristics of children using HAs who were: newly eligible for CIs; with unknown eligibility for CIs; and who had been previously eligible for CIs. Children newly eligible for CIs to be audiologically described in terms of both their better- and worse-hearing ears.

Ultimately a child's candidature for cochlear implantation is determined on a case-by-case basis by a CI team, and children must satisfy the language development clause of TAG566 (NICE, 2019) as well as the multidisciplinary assessment process. However, for the purposes of this chapter, children will be described as 'eligible' for cochlear implantation if they meet the audiometric criterion within NICE guidance on the basis of their HTLs, because this was the information that was available.

Another purpose for collecting these data was to identify children who were eligible for the survey that will be presented in Chapter 5, and to allow the survey participants to be compared to the eligible population.

4.2. METHODS

Data were collected by a form that will be described as an eligibility screening form, since it was used to determine children's eligibility for the CUSP study in addition to assessing the impact of TAG566 (NICE, 2019). This section describes the development of the eligibility screening method used.

4.2.1. Recruitment sites

Full paediatric HA caseload screening was carried out at Bradford Royal Infirmary, at Ropewalk House in Nottingham, and at Huddersfield & Calderdale NHS Trust across two hospital sites. Screening was conducted by qualified audiologists employed by the local NHS Trust.

4.2.2. Ethical considerations

During consultation with stakeholders, the heads of Audiology services had expressed a strong preference for eligibility screening to be carried out by myself rather than their own staff, due to capacity concerns. Permission to apply for the required Confidentiality Advisory Group approval was not granted. Therefore, the data collection method was refined to present as little burden as possible on the staff carrying out screening at participating sites. Permission was sought and granted to receive screening forms for whole caseloads of NHS patients, without families' consent for the purposes of the current study, because specific measures were put in place to ensure the children would not be identifiable using the data to be shared. A screening form was completed for each child by the local hospital staff and coded with a pseudonomised identification code, the key to which was held at the hospital site. The identification code encoded the hospital site, so children could be classified by hospital without being identifiable to the research team. Data extraction was limited to that needed to identify potentially eligible participants, to know which parts of the study they would be eligible for, to understand whether interpreters would be needed to facilitate their recruitment and participation, and basic characteristics such as sex, and age in years, that could be used to cross-check identity. The study inclusion and exclusion criteria to identify potentially eligible participants are summarised in section 4.2.3.

4.2.3. Inclusion / Exclusion criteria

General Inclusion criteria:

- Child aged less than 18 years at the time of eligibility screening
- Current prescription for at least one acoustic hearing aid
- Two or more unaided hearing thresholds out of 0.5, 1, 2, 3, or 4 kHz equal to or greater than 60 dB HL in both ears

General Exclusion criteria

- Child using any bone-conduction hearing device

Additional exclusion criteria for in-person behavioural assessment

- Child aged under 5 years at the time of eligibility screening
- Child has additional needs other than hearing loss that affects their ability to participate in hearing tests

4.2.4. Eligibility screening form development

The eligibility screening form was developed in consultation with the CRN research audiologist at Nottingham. It was, and was implemented electronically to make the collation of data from a large number of children as efficient as possible and to minimise the need for re-entry of data, which could have introduced errors. Tables within the form used pre-defined fields created with the Developer tab within Microsoft Word. This enabled drop-down options for audiologists to select for site code, age, yes/no responses, transducer, decibel scale, date, device type and audiometric thresholds, to minimise transcription errors This section catalogues the items included on the HA eligibility screening form, which can be seen in Appendix A.

4.2.5. Screening form items

CUSP ID code: Each child screened was assigned a pseudonymous code, comprising four letters to denote their hospital site, a three-digit number, and two letters to denote whether they had been identified via a hearing aid or cochlear implant caseload list. Local sites were given a template key document, with CUSP ID codes pre-populated on a Word form. This document linked the CUSP ID code with children's names, dates of birth, hospital number and date screened, entered by the audiologist to ensure children could be identified locally.

Sex: This was recorded to be used as a cross-check of children's identity against other records as a quality control measure for returned surveys.

Age: The child's age was included in years and months to check their eligibility for the study.

Additional needs: For children aged 5 years or older, the audiologist selected "Yes" or "No" regarding the presence of any information within the audiology notes consistent with the child having additional needs other than hearing loss that might affect their ability to participate in hearing tests. If the audiologist answered "Yes", they were asked to give non-identifiable details via a free-text box. It is acknowledged that this does not provide as accurate a measure of any additional difficulties in the way that could be known via General Practitioner or Paediatrician records, or by directly seeking information from caregivers.

However, it was intended to be a characteristic that would be comparable between the different CI eligibility groups and the wider cohort.

Interpreter needs: Eligible families needing interpreters were to be recruited via a local audiologist at clinical appointments where, as routine, an interpreter would be present to assist them. Families who did not need an interpreter could receive the survey in English via the post. The screening form therefore asked the audiologist to specify "Yes" or "No" to whether the family needed an interpreter for audiology appointments. If they chose "Yes" they were asked to specify which language(s) in a free-text box. This information, while not providing an accurate measure of children's ethnicity, was intended to give an indication of whether a language barrier was present that might cause a potential barrier to accessing healthcare.

Hearing device(s) used: The form required information on both right and left ear device use, to distinguish children using only HAs from those who were unaided or used other devices. Children using CIs needed to be identified, so that they could be approached for the CI version of the survey described in chapter 5. All children with a current prescription for a HA were included regardless of the consistency of their HA use, but those known to have returned their HA to become formal non-users were not required to be screened.

If a patient was found to be ineligible after the fields above were entered, for example if they were over 18 years of age or used a bone-conduction device, the form could be returned without extracting any further data. However, if the

child met the age and device use inclusion criteria, audiologists were asked to complete the rest of the form:

Audiometric thresholds: The form requested the most recent unaided audiogram, including air-conduction thresholds at 0.25, 0.5, 1, 2, 3 and 4 kHz in both ears. If there were missing data at any of these frequencies in either ear, audiologists were asked to refer to the preceding unaided audiogram, and to extract all thresholds from this if it supplied any information missing from the most recent. Test date(s), air-conduction transducer, and decibel unit were requested. Audiologists recorded whether a true HTL had been recorded at the level stated, or was thought to be at a softer or louder level.

Other information: The final, optional, free-text cell asked audiologists to "Type any comment here that is needed to interpret the test results above and to establish eligibility, if needed." Audiologists were encouraged to record here any general comments such as whether behavioural test results had been recorded as being of questionable reliability. The form requested that no information be shared that might allow any child to be identified.

4.2.6. Implementation of eligibility screening

Hospitals varied in the administrative methods used to manage their paediatric audiology caseloads. Nottingham's records were paper-based, other sites used electronic records. Some sites' paediatric caseload lists included adults with learning difficulties and some did not. At some sites, children using bone conduction aids rather than acoustic HAs were managed via separate systems

whilst at other sites caseload lists were combined. As there was no overlap between the audiometric fitting criteria for CIs and bone conduction aids, and as the use of a bone conduction aid was an exclusion criterion for participating in the study, sites were not required to screen all children using bone conduction devices. However, if a form was returned for a child using a bone conduction aid, that form was processed and documented within this chapter among cases screened who were not eligible for inclusion in analyses.

It was suggested that sites capture a list of their whole paediatric HA caseload from one moment in time at the start of screening and try to complete data extraction over as short a period as possible. The purpose of this was to capture a picture of children using HAs as soon as possible following the introduction of the new NICE guidance which was to be enacted from June 2019. At Bradford, a list of the paediatric HA caseload was generated that represented every child under the service in November 2019. Due to differences in record-keeping, the other sites screened children on a rolling basis until it was decided that all children had been checked. Sites were encouraged to prioritise screening children with more severe and profound losses earlier on, so that survey data collection could take place as rapidly as possible. Another potential benefit of prioritising those with greater hearing losses was that fewer children within or close CI eligibility would have become older than 18 years, or been referred for CI assessment, before a screening form was completed for them. Figure 4.2 illustrates the time-course of eligibility screening for all four sites, which ran from January 2019 to March 2021.

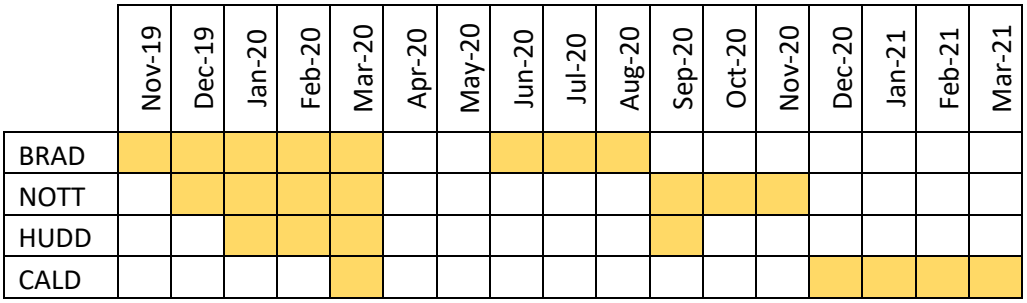


Figure 4.1: Time-course of HA eligibility screening by site

Coloured cells indicate that some eligibility screening was carried out for children using HAs during that month. Blank cells indicate no eligibility screening took place during that month. BRAD = Bradford; NOTT = Nottingham; HUDD = Huddersfield; CALD = Calderdale

All four sites reported that all children using at least one acoustic HA had been screened, however this could not be verified.

4.2.7. Analysis of audiometric data

In line with BSA guidelines (BSA, 2018) a value of 130 dB HL was assigned to any audiometric frequency where the true threshold was not reached up to the limits of the audiometer. Group analyses were carried out to identify the proportion of missing threshold data for each audiometric frequency, based on both the most recent audiogram and data from this and the preceding audiogram combined. As described in Chapter 1, the BSA recommends averaging HTLs over the five frequencies 0.25, 0.5, 1, 2 and 4 kHz to classify hearing losses by its descriptors of mild, moderate etc. (BSA, 2018). However, due to the amount of missing HTLs at 0.25 kHz, four-frequency averages of thresholds across 0.5, 1, 2 and 4 kHz were calculated for right and left ears where available, or for binaural thresholds measured in soundfield. The ear with the lower 4-frequency average HTL was classed as the better-hearing ear. This was used to classify children by the BSA descriptors (BSA, 2018). Four-

frequency averages are also widely recognised internationally, facilitating wider interpretation of this dataset.

Where no frequency-specific thresholds were available, children were classified according to the lowest level of environmental sounds needed to elicit behavioural responses. For cases where not all four frequencies were measured, the mean of the available thresholds was calculated. If none of the four thresholds were available, nor any quantified behavioural observation to environmental sounds, children's degree of hearing loss was described as 'could not classify'.

Children were also classified according to the audiometric criteria of both TAG166 (NICE, 2009) and TAG566 (NICE, 2019). The 2009 NICE guidance specified that in both right and left ears, unaided air-conduction thresholds must be ≥ 90 dB HL at both 2 kHz and 4 kHz. Accordingly, if any threshold fell at ≤ 85 dB HL at either of those two frequencies in either ear, it could be confirmed that the child did not meet the 2009 NICE guidance for cochlear implantation. If no HTL was ≤ 85 dB HL at 2 or 4 kHz, and any HTL for 2 kHz or 4 kHz was missing in either ear, it could not be determined whether a child met this guidance. The 2019 guidance specifies that HTLs be ≥ 80 dB HL for at least two frequencies including 0.5, 1, 2, 3 or 4 kHz in both ears. These HTLs need not be at the same frequencies for the right and left ears. Therefore, if a child had four or more thresholds at ≤ 75 dB HL across that frequency range in either ear, it could be confirmed that they did not meet this guidance. If they had only one threshold at ≥ 80 dB HL and up to three thresholds at ≤ 75 dB HL with missing data for the remaining frequency / frequencies in either ear, it could not be determined whether they met this guidance. If they had two

or more missing HTLs at 0.5, 1, 2, 3 or 4 kHz in either ear, it could not be determined whether they met the 2019 guidance. If there were too few thresholds on the most recent audiogram to determine eligibility for either guidance, the preceding audiogram was also reviewed, and HTLs values from this imputed for the values missing from the most-recent. In some cases whose eligibility remained unknown, classification could be made with the help of comments in the "any other comments" box. Sometimes these described HTLs not included within the table in the form, or revealed profound hearing loss in an ear, based on audiograms preceding the two most recent. For each child, it was determined if their HTLs placed them within or outside NICE TAG166 (NICE, 2009) and NICE TAG566 (NICE, 2019) or there was insufficient audiometric data to establish their eligibility by either guidance.

4.3. RESULTS

This section will begin by presenting the results of identifying the eligible cohort. This will be followed by results that map to the chapter objectives, including how many children were eligible, of unknown eligibility, or previously eligible for CIs by TAG166 (NICE, 2009) and TAG566 (NICE, 2019). Last, the demographic and audiological characteristics of these children will be presented, both for the overall cohort and individual NICE eligibility groups.

4.3.1. Cohort identification

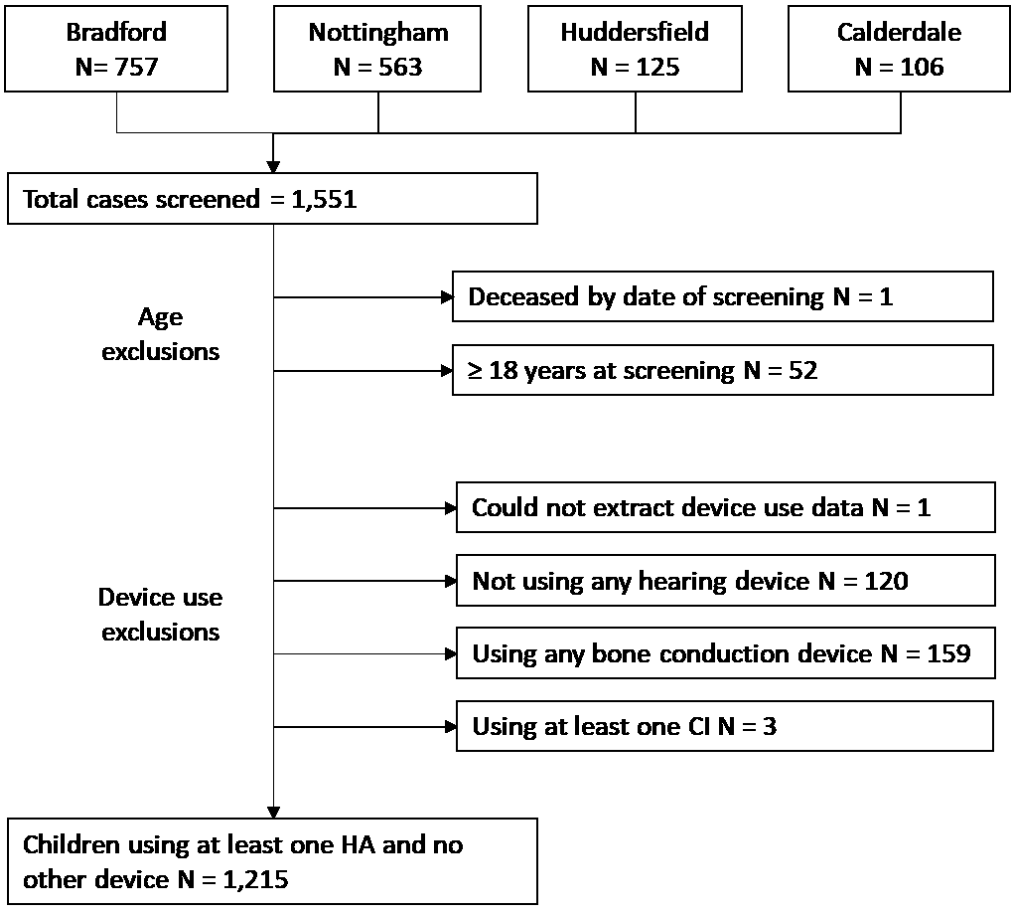


Figure 4.2: Flow diagram of whole caseload eligibility screening
N = number of children; CI = cochlear implant; HA = hearing aid.

Of 1,551 cases screened, 1,215 were children under 18 years of age using at least one HA. The 52 patients who were aged 18 years or older at the time of screening included adults with learning difficulties who were managed by the paediatric audiology team, teenagers who had turned 18 years old during the period of eligibility screening, and teenagers who were on the paediatric hearing aid caseloads but had failed to attend appointments to be discharged to adult services. Of the 120 children not using any hearing device, some were waiting for the first fitting of devices. Others had been issued devices in the

past for conditions such as temporary or fluctuating conductive losses and were on waiting lists for review but were not currently fitted with aids. Tables 4.1 and 4.2 show exclusions overall and by site for age and device criteria respectively.

Table 4.1: Age exclusions by site

	All	Bradford	Nottingham	Huddersfield	Calderdale
N screened	**1,551**	**757**	**563**	**125**	**106**
Deceased	1 (<1%)	1 (<1%)	—	—	—
>= 18 years	52 (3%)	46 (6%)	—	1 (<1%)	5 (5%)

Table 4.2: Device exclusions by site

	All	BRAD	NOTT	HUDD	CALD
Total N <18 years	**1,498**	**710**	**563**	**124**	**101**
No device data	1 (<1%)	1 (<1%)	—	—	—
Device returned	120 (8%)	32 (5%)	76 (13%)	2 (2%)	10 (10%)
BC aid	159 (11%)	105 (15%)	36 (6%)	4 (3%)	14 (14%)
CI	3 (<1%)	1 (<1%)	2 (<1%)	—	—
N not excluded	**1215**	**571**	**449**	**118**	**77**

BRAD = Bradford, NOTT = Nottingham, HUDD = Huddersfield, CALD = Calderdale; BC = bone conduction; CI = cochlear implant

Table 4.2 reveals variation in device exclusions between sites. This is likely because caseloads are managed in different ways between hospitals. The largest number of children using bone conduction aids were screened at Bradford, likely because bone conduction aid users were managed via the same waiting list system as children using HAs. Few children using CIs were found on local audiology caseloads. The child at Bradford with CIs had become a CI user after the caseload list was downloaded and before the screening form was completed. The two children at Nottingham each used one CI, managed by a regional CI service, with a contralateral HA that continued to be managed by the local audiology team.

The remaining children who were not excluded represented complete cohorts of children aged under 18 years using at least one acoustic hearing aid and no other type of device across four hospital sites.

4.3.2. Children's characteristics overall and by hospital site

This section will achieve the objective of documenting the demographic and audiological characteristics of whole caseloads of children using HAs. Characteristics including age distribution, family need for interpreters and the prevalence of additional needs affecting children's ability to participate in audiology appointments will be shown overall and by hospital site. Table 4.3 shows descriptive group summary data for sex, age, and HA fitting.

Table 4.3: Cohort demographics and HA fitting overall and by hospital site

	All Sites N = 1215	**BRAD N = 571**	**NOTT N = 449**	**HUDD N = 118**	**CALD N = 77**
Female (%)	590 (49)	278 (49)	216 (48)	61 (52)	35 (45)
Male (%)	625 (51)	293 (51)	232 (52)	57 (48)	42 (55)
Sex missing (%)	1 (<1)	—	1 (<1)	—	—
Median age (years)	10	10	9	10	11
IQR age (years)	7 to 13	7 to 14	7 to 13	7 to 12	6 to 14
Bilateral HAs (%)	871 (72)	431 (75)	302 (67)	81 (69)	57 (74)
Unilateral HA (%)	344 (28)	140 (25)	147 (33)	37 (31)	20 (26)

HA = Hearing aid, IQR = Interquartile range, BRAD = Bradford, NOTT = Nottingham, HUDD = Huddersfield, CALD = Calderdale

4.3.2.1. Children's audiological characteristics overall and by hospital site

Table 4.4 shows the distribution of children's hearing loss based on BSA descriptors, compared to the distribution of children's hearing loss reported in the CRIDE report for 2020-2021 (CRIDE, 2021).

Table 4.4: CUSP study and CRIDE report cohorts by hearing loss classifications

BSA Hearing loss classification	CUSP HA cohort N = 1,215	CRIDE 2020/21 N = 44,198
Normal (≤20 dB HL)	349 (28.7%)	9,365 (22%)
Mild (21 – 40 dB HL)	373 (30.7%)	10,917 (26%)
Moderate (41 – 70 dB HL)	384 (31.6%)	13,265 (31%)
Severe (71 – 95 dB HL)	75 (6.2%)	3,546 (8%)
Profound (>95 dB HL)	29 (2.4%)	5,064 (12%)
Could not classify	5 (0.4%)	2,041 (5%)

BSA = British Society of Audiology, CUSP = Comparative oUtcomes for Severely and Profoundly deaf children, CRIDE = Consortium for Research in Deaf Education

Figure 4.3 illustrates the distribution of 4-frequency average HTLs in the better-hearing ear for children using at least one HA by NHS Trust. Data were normalised by plotting the proportion of each caseload falling within each 10 dB range. Given the smaller numbers of children, and that they were two sites of the same NHS Trust under the same Audiology management, data from Huddersfield and Calderdale were combined. The closest agreement between sites was the proportion of children with losses between 50 and 90 dB HL. Bradford had a greater proportion of children with losses above 90 dB HL.

Nottingham had a greater proportion of children with losses between 10 and 30 dB HL in the better-hearing ear.

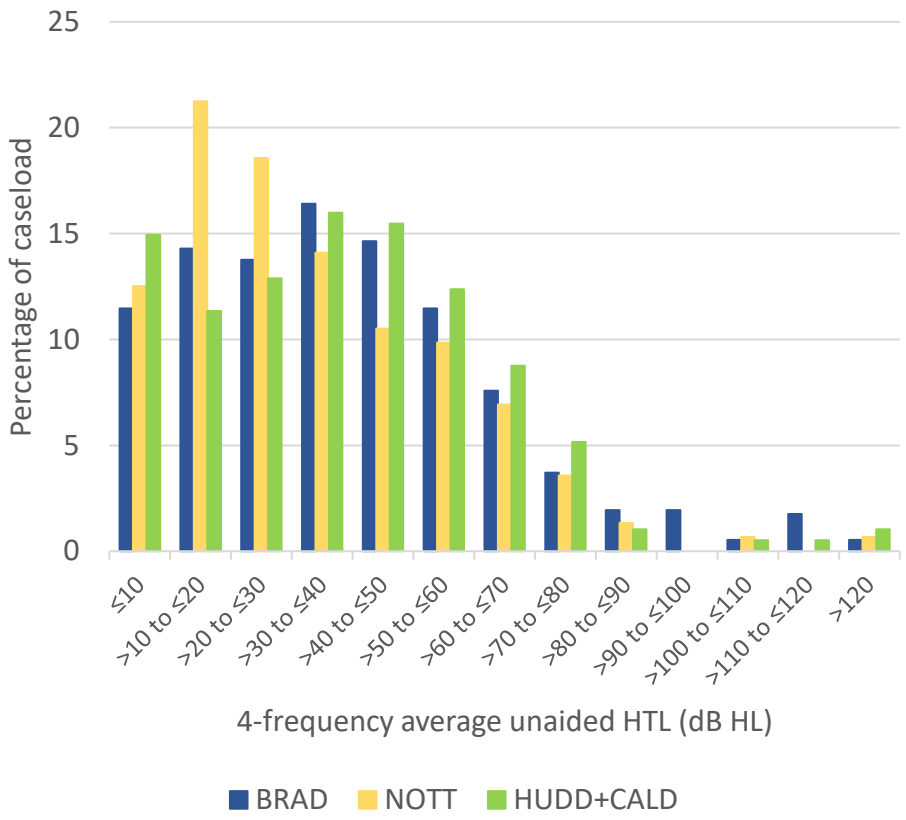


Figure 4.3: Distribution of average unaided 4-frequency HTLs by NHS Trust

BRAD = Bradford, NOTT = Nottingham, HUDD+CALD = Huddersfield & Calderdale, 4-frequency average unaided HTL = Unaided hearing threshold level averaged over 0.5, 1, 2 and 4 kHz

4.3.2.2. Children's age distribution overall and by hospital site

Figure 4.5 shows the age distributions of caseloads by NHS Trust. Data from Huddersfield and Calderdale were combined as the numbers of children at each site were small and together represented the children under one Audiology manager and NHS Trust.

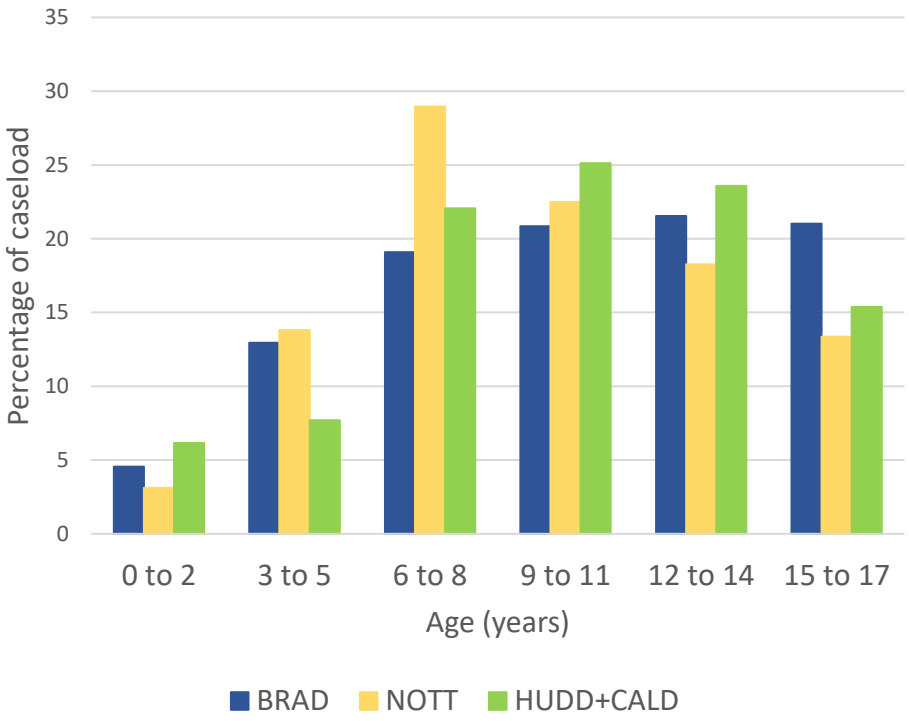


Figure 4.4: HA caseload age distributions by hospital site

BRAD = Bradford, NOTT = Nottingham, HUDD+CALD = Huddersfield & Calderdale.

The proportion of caseloads within each age group increased from birth to 9-to-11 year-olds for Bradford and Huddersfield & Calderdale, decreasing for 15-to-17 year-olds at Huddersfield & Calderdale and plateauing at Bradford. In contrast, the modal age of children at Nottingham was the 6-to-8 year-old category, with the proportion declining for each older age group.

Figure 4.5 is a heat map illustrating the numbers of children in each 10 dB 4-frequency average unaided threshold band by age group in years. Numbers of children in each category are shown rather than percentages, due to the small percentages involved for individual cells. The darker the shading the greater the proportion of the whole caseload within each cell. The modal average threshold for infants fell within the moderate range, whereas the modal losses

for older children and teenagers fell within the normal to mild ranges, by the BSA classification system (BSA, 2018).

		Age (years)					
		0 to 2	3 to 5	6 to 8	9 to 11	12 to 14	15 to 17
4-freq HTL (dB HL)	<=10	0	6	23	58	35	28
	>10 to <=20	0	22	51	42	45	38
	>20 to <=30	6	23	54	38	35	30
	>30 to <=40	5	28	47	37	33	37
	>40 to <=50	11	21	39	38	31	20
	>50 to <=60	11	18	32	24	31	17
	>60 to <=70	9	15	16	18	21	12
	>70 to <=80	3	12	7	8	6	11
	>80 to <=90	1	3	2	1	5	7
	>90 to <=100	2	0	3	1	4	1
	>100 to <=110	0	1	1	1	0	4
	>110 to <=120	0	0	3	3	1	4
	>120	2	0	2	0	3	1
	total	**50**	**149**	**280**	**269**	**250**	**210**

Figure 4.5 Distribution of whole cohort by 4-frequency unaided HTLs and age

4.3.2.3. Family need for interpreters overall and by hospital site

Table 4.4 shows the number and proportions of families who needed no interpreter, any signed interpreter, or any spoken interpreter, with the type of spoken language interpreter also classified into the two most frequently needed geographical regions (South Asian and Eastern European), any other spoken language, or an unknown language, for the cohort overall and by hospital site.

Table 4.5: Family need for interpreters at audiology appointments overall and by hospital site

	All sites N = 1215	BRAD N = 571	NOTT N = 449	HUDD N = 118	CALD N = 77
Type of interpreter					
None (%)	1069 (88)	456 (80)	424 (94)	116 (98)	73 (95)
Any signed (%)	29 (2)	26 (5)	3 (<1)	—	—
Any spoken (%)	129 (11)	100 (18)	23 (5)	2 (2)	4 (5)
Language(s) needed					
BSL only (%)	17 (1)	15 (3)	2 (<1)	—	—
S. Asian and BSL (%)	3 (<1)	3 (<1)	—	—	—
E. European and BSL (%)	9 (<1)	8 (1)	1 (<1)	—	—
S. Asian only (%)	46 (4)	42 (7)	4 (<1)	—	—
E. European only (%)	52 (4)	36 (6)	12 (3)	—	4 (5)
Other spoken (%)	13 (1)	6 (1)	6 (1)	1 (<1)	—
Unknown spoken (%)	6 (<1)	5 (<1)	—	1 (<1)	—

BRAD = Bradford, NOTT = Nottingham, HUDD = Huddersfield, CALD = Calderdale, BSL = British sign language; S. Asian = South Asian, E. European = Eastern European.

The eligibility screening form did not require audiologists to specify which family member(s) the interpreters were needed for. However, on some forms this information was provided in the free-text box supplied to name the language(s). In several cases where two interpreters were needed, the spoken-language interpreter was for a parent and the sign-language interpreter for the child. In other cases, a sign-language interpreter was needed for a parent, or a sign-language interpreter was requested for one parent, and a spoken-language interpreter for the other parent. The proportion of families who needed any interpreter varied significantly between with the greatest need at Bradford. Bradford's caseload included all but one family where both signed- and spoken-language interpreters were needed and had similar proportions of families who needed Eastern European or South Asian interpretation. Eastern European languages were the most often requested at Nottingham and

Calderdale. Huddersfield had the smallest proportion of families who needed interpreters.

Figure 4.6 shows the proportions of families needing spoken and/or signed interpreters by degree of hearing loss in the child's better-hearing ear.

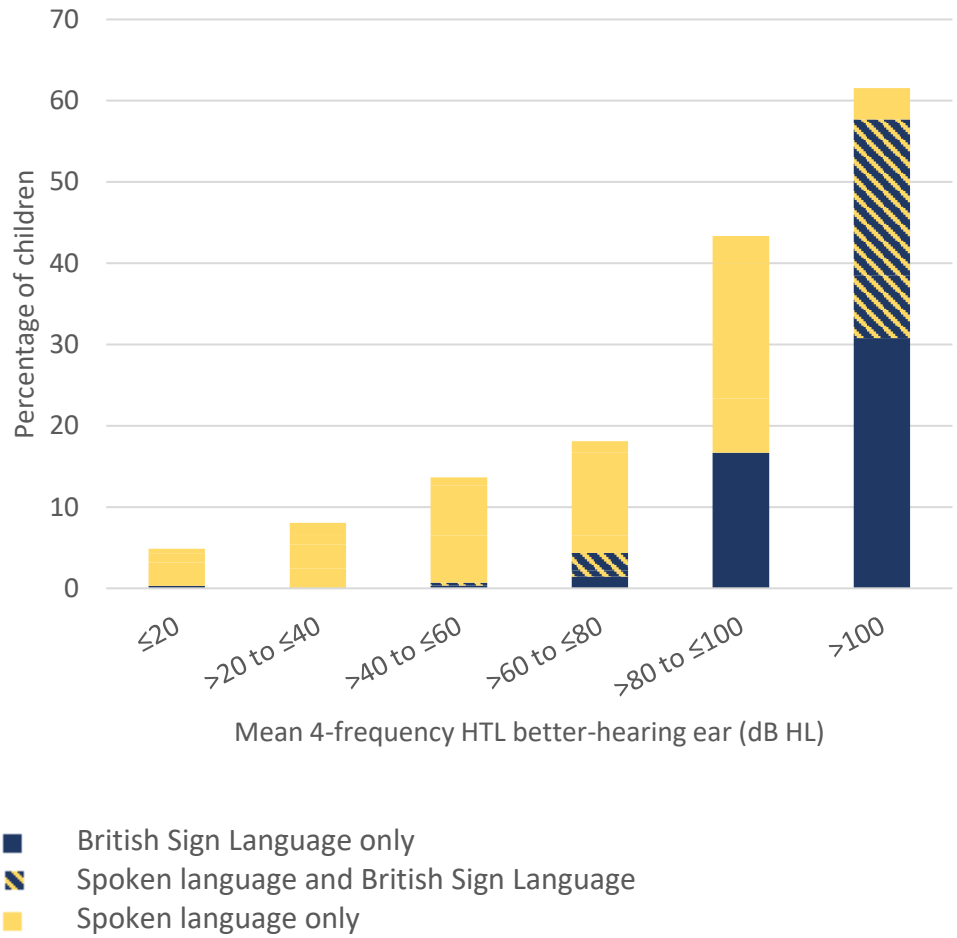


Figure 4.6: Proportion of families needing interpreter(s) by 4-frequency average HTLs

4-frequency HTL = mean of available hearing threshold levels at 0.5, 1, 2 and 4 kHz.

Due to the smaller numbers of children with severe and profound losses, this figure shows children grouped into hearing loss categories of 20 dB rather than 10 dB range. Over half the families of children with hearing losses greater than

100 dB HL needed a BSL interpreter and most of the families who needed both spoken and signed interpreting also fell within this group.

4.3.2.4. Children's additional needs overall and by hospital site

Data on additional needs was only extracted for children aged 5 years or older due to ethical restrictions. Therefore, data on the presence of additional needs was not available for children aged <5 years. Table 4.6 shows the number and proportion of children aged ≥5 years with or without additional needs or with missing data, and those aged <5 years, overall and by hospital site.

Table 4.6: Prevalence of additional needs overall and by hospital site.

	All Sites N = 1215	Bradford N = 571	Nottingham N = 449	Huddersfield N = 102	Calderdale N = 77
≥ 5 years, no additional needs (%)	861 (71)	407 (71)	290 (65)	102 (86)	62 (81)
≥ 5 years, additional needs (%)	182 (15)	80 (14)	93 (21)	3 (3)	6 (8)
≥ 5 years, data missing (%)	36 (35)	10 (2)	24 (5)	2 (2)	—
Under 5 years (%)	136 (11)	74 (13)	42 (9)	11 (9)	9 (12)

Classing children aged ≥5 years with missing data regarding additional needs as having no additional needs, the proportion of children aged ≥5 years with additional needs that affected their ability to participate in hearing tests was 20%. There were differences between sites in the prevalence of additional needs, with the larger sites of Bradford and Nottingham having the greatest proportions.

Table 4.7 shows the proportion of the whole cohort grouped by whether they had additional needs that affected their ability to participate in hearing tests. 20-dB bins were used due to small sample numbers and the data reflect children aged ≥5 years only as data on additional needs was not obtainable for those <5 years.

Table 4.7: Proportion of children with additional needs by 4-frequency average unaided HTLs

	4-frequency average hearing threshold level of the better-hearing ear (dB HL)					
	≤20	**>20 to ≤40**	**>40 to ≤60**	**>60 to ≤80**	**>80 to ≤100**	**>100**
Proportion of children with additional needs	13%	17%	20%	20%	32%	23%

4.3.3. Prevalence of children in each NICE eligibility group

This section will achieve the objective of calculating the prevalence of children using HAs who were newly eligible for CIs, of unknown eligibility for CIs, and previously eligible for CIs following TAG566 (NICE, 2019).

Table 4.11 shows the number and proportions of children overall and by site who fell outside, within, and who could not be classified by both TAG166 (NICE, 2009) and TAG566 (NICE, 2019).

Table 4.8: Audiometric CI eligibility classifications overall and by hospital site

	All Sites N = 1215	BRAD N = 571	NOTT N = 449	HUDD N = 118	CALD N = 77
			TAG566 (NICE, 2019)		
Outside (%)	1039 (86)	479 (84)	392 (87)	102 (86)	67 (87)
CND (%)	85 (7)	38 (7)	35 (8)	4 (3)	7 (9)
Within (%)	91 (7)	54 (7)	22 (5)	12 (10)	3 (4)
			TAG166 (NICE, 2009)		
Outside (%)	1152 (95)	532 (93)	434 (97)	111 (94)	76 (99)
CND (%)	26 (2)	13 (2)	7 (2)	4 (3)	1 (1)
Within (%)	37 (3)	26 (5)	8 (2)	3 (3)	—

CND = could not determine eligibility, TAG = Technology Appraisal Guidance, NICE = National Institute for Health and Care Excellence, BRAD = Bradford, NOTT = Nottingham, HUDD = Huddersfield, CALD = Calderdale

Figure 4.7 is a Sankey diagram that shows more dynamically how children's audiometric eligibility status changed following the revised guidance.

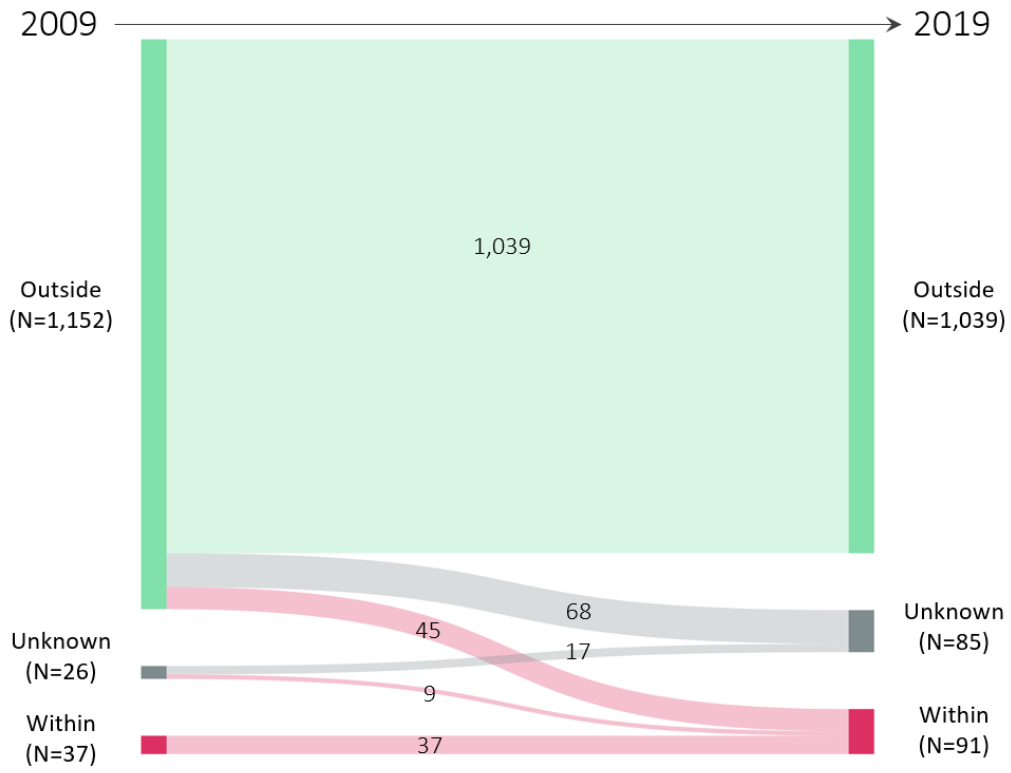


Figure 4.7: The impact of the revised NICE guidance on the cohort's audiometric eligibility for CIs

For both the 2009 (left axis, National Institute for Health and Care Excellence Technology Appraisal Guidance 166) and 2019 (right axis, National Institute for Health and Care Excellence Technology Appraisal Guidance 566) guidance, the numbers of children who fell within, outside, or whose eligibility was unknown due to missing HTLs are represented by red, green, and grey vertical bars respectively. The numbers in the centre of the figure describe the number of children whose status did or did not change from and to each category.

All 37 children who met the 2009 guidance also met the 2019 guidance, which is to be expected since all children with thresholds of ≥90 dB HL at 2 and 4 kHz bilaterally by definition also have at least two thresholds ≥80 dB HL at two or more frequencies including 0.5, 1, 2, 3 and 4 kHz bilaterally. The remaining 54 children who met the 2019 guidance, those newly eligible, included some whose hearing fell outside 2009 guidance (N=45) and some whose candidacy via the 2009 guidance could not be determined (N=9). There

were 85 children whose 2019 eligibility was not known, due to missing HTLs. Most of these (N=68) had been ineligible for CIs under the previous guidance and the remainder (N=17) were of unknown eligibility previously also.

Table 4.9 shows the number and proportions of children overall and by hospital site falling into each TAG566 (NICE, 2019) CI eligibility group; those newly eligible, those whose eligibility was unknown, those who had been previously eligible under TAG166 (NICE, 2009) and those who remained ineligible for CIs.

Table 4.9: Number and proportions of children in each NICE CI eligibility group overall and by site

TAG566 (NICE, 2019) CI eligibility group	All Sites N = 1215	Bradford N = 571	Nottingham N = 449	Huddersfield N = 118	Calderdale N = 77
Newly eligible	54 (4%)	28 (5%)	14 (3%)	9 (8%)	3 (4%)
Eligibility unknown	85 (7%)	38 (6%)	36 (8%)	4 (3%)	7 (9%)
Previously eligible	37 (3%)	26 (5%)	8 (2%)	3 (3%)	—
Not eligible	1,039 (86%)	479 (84%)	391 (87%)	102 (86%)	67 (87%)

TAG = Technology Appraisal Guidance, NICE = National Institute for Health and Care Excellence

In summary, this data confirms that the change in NICE guidance had achieved its goal of widening access to CIs because 54 (4.4%) of the cohort had become newly eligible for cochlear implantation. It also confirms that increased number of measured HTLs needed to establish a child's eligibility under TAG566 (NICE, 2019) increased the number of children for whom eligibility was unknown, from 26 (2.1%) to 85 (7.0%).

4.3.4. Children's characteristics by eligibility for cochlear implantation

This section will achieve the final chapter objective by documenting the demographic and audiological characteristics of children using HAs who were: newly eligible for CIs; of unknown eligibility for CIs; and who had been previously eligible for CIs. Each group will be described in terms of the characteristics suspected to influence access to cochlear implantation, as outlined in the chapter introduction 4.1. These include: age; family need for interpreting services; additional needs; and missing audiometric data. The group newly eligible for CIs will be audiologically characterised in the most detail, to establish to what extent any asymmetry in their better- and worse- ear HTLs might be expected to impact their benefit from bilateral CIs should they receive them in the future.

Table 4.10 shows group summary characteristics by NICE eligibility group including sex, age, unilateral or bilateral HA fitting, 4-frequency average unaided HTLs and the stimulus presentation method used for their most recent audiogram.

Table 4.10: Children's summary characteristics by NICE eligibility group

	Newly eligible N=54	Eligibility unknown N=85	Previously eligible N=37	Not eligible N=1,039
Female (%)	24 (44)	45 (53)	17 (46)	504 (49)
Male (%)	30 (56)	40 (47)	20 (54)	534 (51)
Sex missing (%)	—	—	—	1 (<1)
Median age (years)	10	5	13	10
IQR age (years)	6.25 to 13	3 to 10	10 to 16	7 to 13
Bilateral HAs (%)	53 (98%)	78 (92%)	35 (96%)	707 (68%)
Unilateral HA (%)	1 (2%)	7 (8%)	2 (4%)	332 (32%)
Median 4-freq HTL (dB HL)	77.5	60.0	102	30
IQR 4-freq HTL (dB HL)	71.3 - 82.5	45.0 – 69.4	90.0 - 113.8	16.3 – 43.8

IQR = Interquartile range, HA = Hearing aid, 4-freq HTL = unaided hearing threshold levels averaged over 0.5, 1, 2 and 4 kHz or as many of these thresholds as was available.

4.3.4.1. Age distributions

The age distributions of each NICE eligibility group are plotted in Figure 4.8.

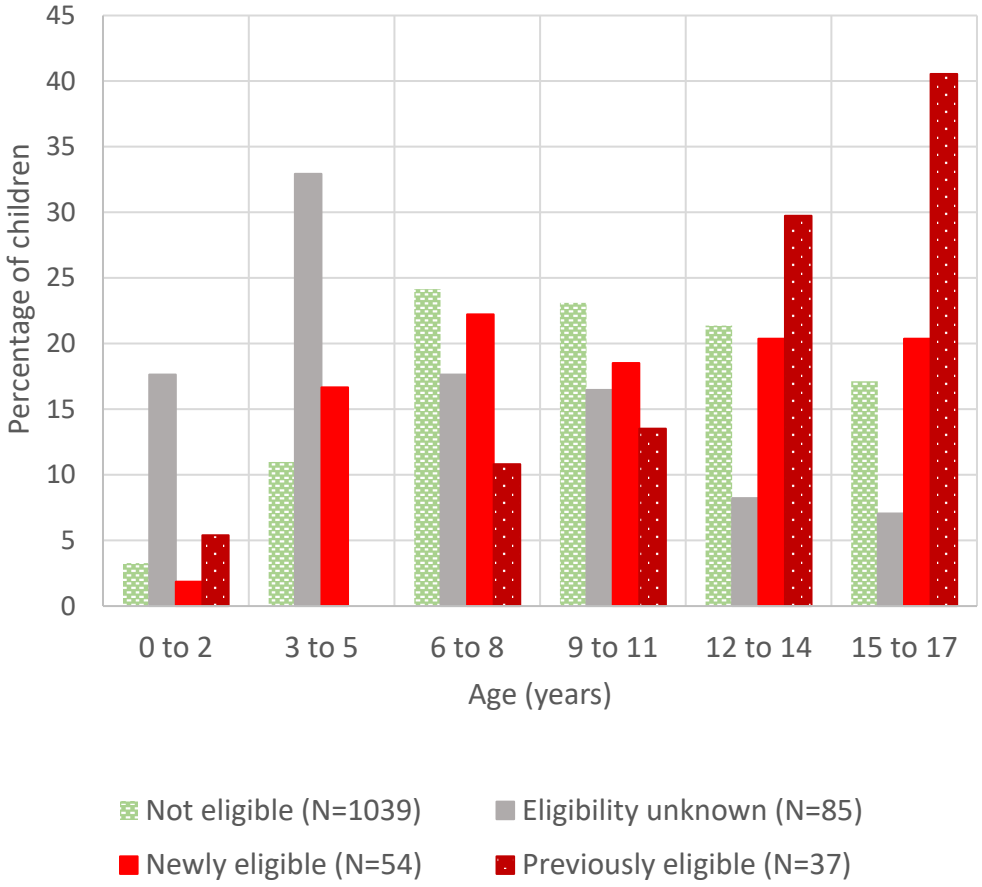


Figure 4.8: Age distributions by NICE guidance audiometric eligibility group.

Children are grouped by their audiometric eligibility for cochlear implantation under the National Institute for Health and Care Excellence Technology Appraisal Guidance 155 (NICE, 2019).

4.3.4.2. Interpreter needs and Additional needs

To explore differences in the characteristics of children within each NICE eligibility group, Tables 4.11 to 4.14 show the numbers and proportions of

children within each group who had additional needs and/or family need for interpreter(s).

Table 4.11: Family need for interpreters and additional needs of children previously eligible for CI under TAG166 (2009)

Interpreter	Under 5 years	5 years or older		Row Totals
		No additional needs	Additional needs	
None	2 (5%)	9 (24%)	4 (11%)	**15 (41%)**
Any	—	18 (49%)	4 (11%)	**22 (59%)**
Totals	**2 (5%)**	**27 (73%)**	**8 (22%)**	N=37

Table 4.12: Family need for interpreters and additional needs of children newly eligible for CI under TAG566 (2019)

Interpreter	Under 5 years	5 years or older		Row Totals
		No additional needs	Additional needs	
None	3 (6%)	29 (54%)	7 (13%)	**39 (72%)**
Any	1 (2%)	11 (20%)	3 (6%)	**15 (28%)**
Totals	**4 (7%)**	**40 (74%)**	**10 (19%)**	**N=54**

Table 4.13: Family need for interpreters and additional needs of children of unknown eligibility for CI under TAG566 (2019)

Interpreter	Under 5 years	5 years or older		Row Totals
		No additional needs	Additional needs	
None	32 (38%)	17 (20%)	30 (35%)	**79 (93%)**
Any	3 (4%)	2 (2%)	1 (1%)	**6 (7%)**
Totals	**35 (41%)**	**19 (22%)**	**31 (36%)**	**N=85**

Table 4.14: Family need for interpreters and additional needs of children not eligible for CI under TAG566 (2019)

Interpreter	Under 5 years	5 years or older		Row Totals
		No additional Needs	Additional needs	
None	84 (8%)	729 (70%)	124 (12%)	**937 (90%)**
Any	11 (1%)	81 (8%)	10 (<1%)	**102 (9%)**
Column	**95 (9%)**	**810 (78%)**	**134 (13%)**	**N=1,039**

For Tables 4.11 to 4.14, children aged ≥5 years with missing data regarding their additional needs were classified as not having additional needs and

children with missing data regarding interpreters were classified as not needing an interpreter. These tables allow the numbers and proportions of children in each NICE eligibility group to be clearly accessible. For ease of visualisation between groups, Figure 4.9 shows the data from Tables 4.11 to 4.14 in graphical form, with the composition of each group based on the proportions in the matrices tables shown as cumulative bar charts.

Tables 4.11 to 4.14 and Figure 4.9 show that the group who could not be classified by the 2019 guidance differed from the other groups, having greater proportions of children aged under 5 years and children with additional needs. From the groups of children who could be classified, the proportion of families who needed any interpreter was least for those not eligible for CIs, greater for those newly eligible, and greatest for those previously eligible. The group with the highest proportion of children with neither additional needs nor family need for an interpreter was those not eligible for CIs.

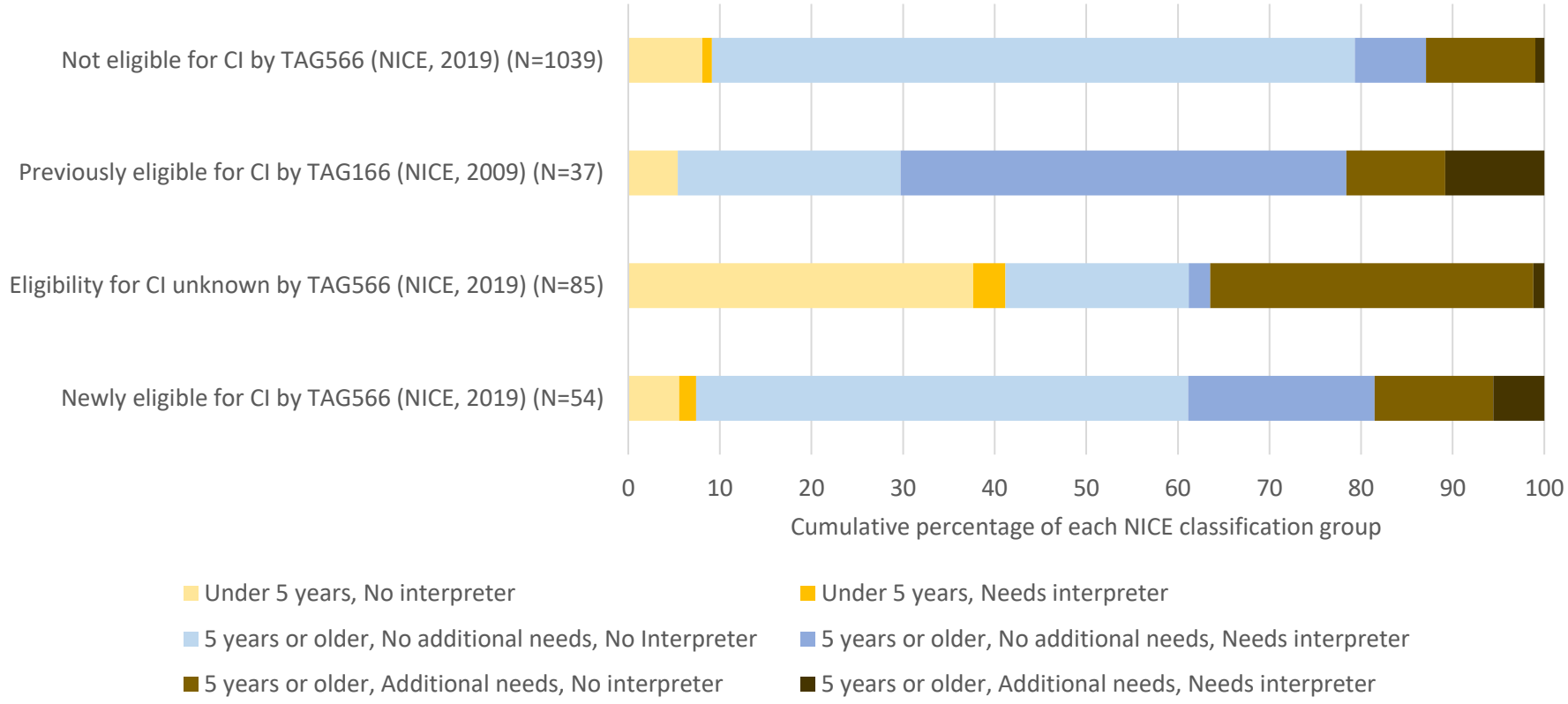


Figure 4.9: Children's age category, family need for any interpreter and additional needs by NICE eligibility group
TAG = Technology Appraisal Guidance, NICE = National Institute for Health and Care Excellence, CI = cochlear implantation

Table 4.15 documents the need for different types of interpreters for each of the four NICE eligibility groups in more detail

Table 4.15: Type of interpreter(s) needed by NICE eligibility group

	Newly eligible N = 54	Eligibility unknown N = 85	Previously eligible N = 37	Not eligible N = 1,039
No interpreter (%)	39 (72)	79 (93)	14 (38)	937 (90)
BSL only (%)	2 (4)	—	12 (32)	3 (<1)
South Asian and BSL (%)	1 (2)	—	2 (5)	—
Eastern European and BSL (%)	3 (6)	—	4 (11)	2 (<1)
South Asian only (%)	2 (4)	2 (2)	2 (5)	40 (4)
Eastern European only (%)	2 (4)	2 (2)	2 (5)	43 (4)
Other spoken (%)	2 (4)	2 (2)	1 (3)	8 (<1)
Unknown spoken (%)	—	—	—	6 (<1)
Any spoken (%)	13 (24)	6 (7)	11 (30)	99 (10)
Any South Asian (%)	3 (6)	2 (2)	4 (11)	40 (4)
Any Eastern European (%)	5 (9)	2 (2)	6 (16)	45 (4)
Any BSL (%)	6 (11)	—	18 (49)	5 (<1)

BSL = British Sign Language

4.3.4.3. Missing audiometric data

Figure 4.10 plots the percentage of audiometric frequencies for which HTLs were measured considering children's two most-recent audiograms combined.

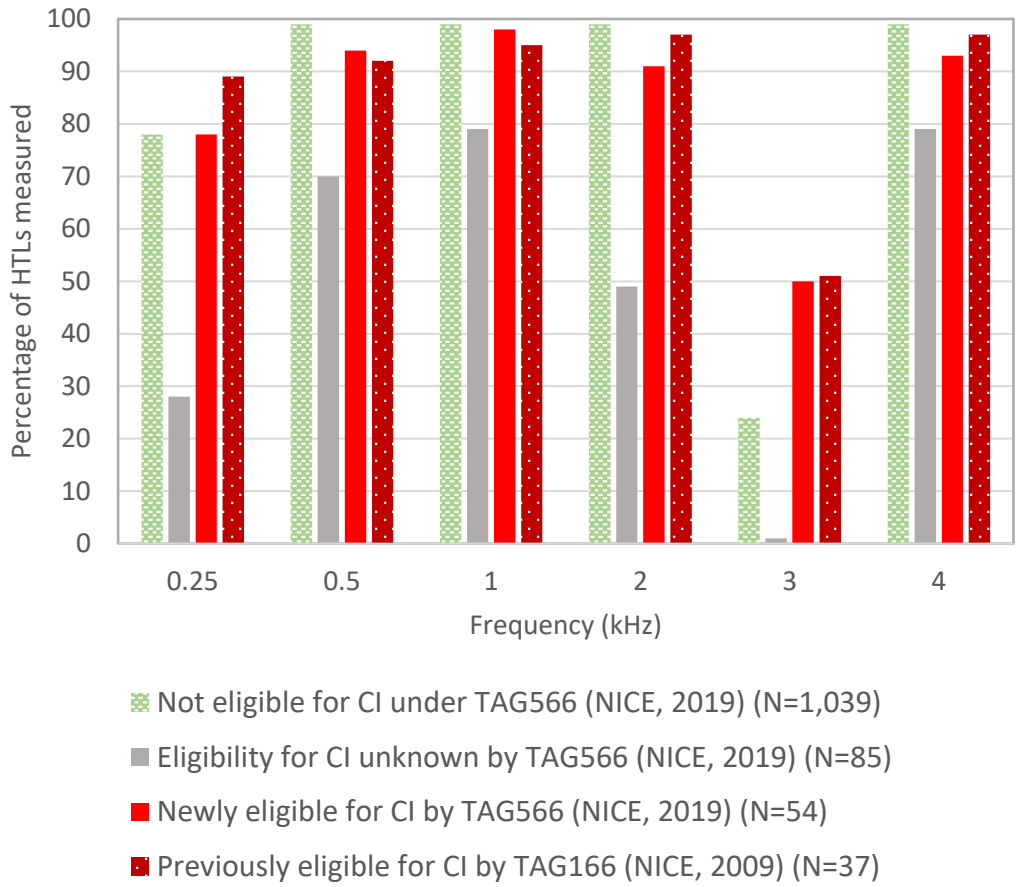


Figure 4.10: Percentage of audiometric thresholds measured by NICE eligibility group

CI = Cochlear implantation, TAG = Technology Appraisal Guidance, NICE = National Institute for Health and Care Excellence, HTLs = Hearing threshold levels.

For children known to be not eligible for CIs, almost all (99%) had HTLs measured at 0.5, 1, 2, and 4 kHz. For children whose eligibility for CIs was unknown, the most often measured HTLs were at 1 and 4 kHz, although these were available for under 80% of children. The groups who were eligible for

CIs, before or following the NICE guidance change, were the most likely to have had HTLs measured at 3 kHz, and those who could not be classified were the least likely to have had their hearing tested at that frequency.

4.3.5. Better- and worse-ear HTLs of children newly eligible for CIs

This section fulfils the objective of describing in detail the audiological characteristics of children newly eligible for cochlear implantation under TAG566 (2019). Forty-eight (89%) of the 54 children in this group had ear-specific unaided HTLs available at 0.5, 1, 2, or 4 kHz.

Figure 4.11 shows the 4-frequency average unaided thresholds of the better and worse-hearing ears. These are plotted for each child for both ears. Different symbols differentiate between children whose HTLs fell outside or could not be classified by TAG166 (2009).

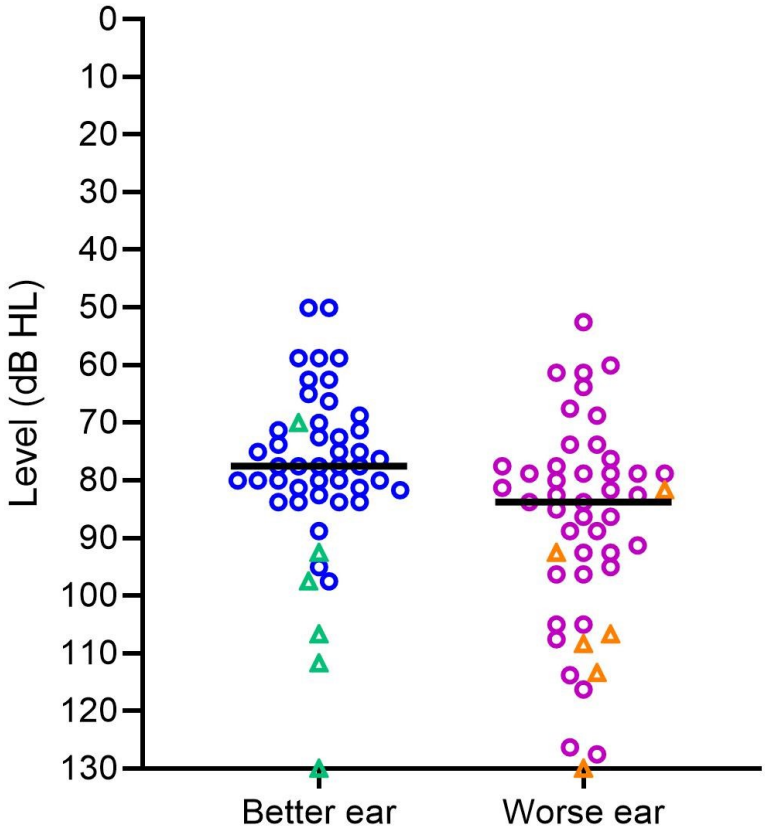


Figure 4.11: 4-frequency average HTLs for better- and worse-hearing ears for children newly confirmed to be audiometrically-eligible for CIs by TAG566 (2019)

Horizontal black lines show median 4-frequency average HTLs. Individual data points are plotted as circles for children who fell outside TAG166 (2009) and within TAG566 (2019). Individual data points are plotted as triangles for children who could not be classified by TAG166 (2009) and fell within TAG566 (2019).

Figure 4.11 reveals that children who could not be classified by TAG166 (2009) due to missing HTLs at either 2 and/or 4 kHz tended to have worse hearing in their better-hearing ear than children who were confirmed to have fallen outside TAG166 (2009) and within TAG566 (2019).

Figure 4.12 plots the distribution of HTLs measured at 0.5, 1, 2 and 4 kHz for the better- and worse-hearing ears of this group.

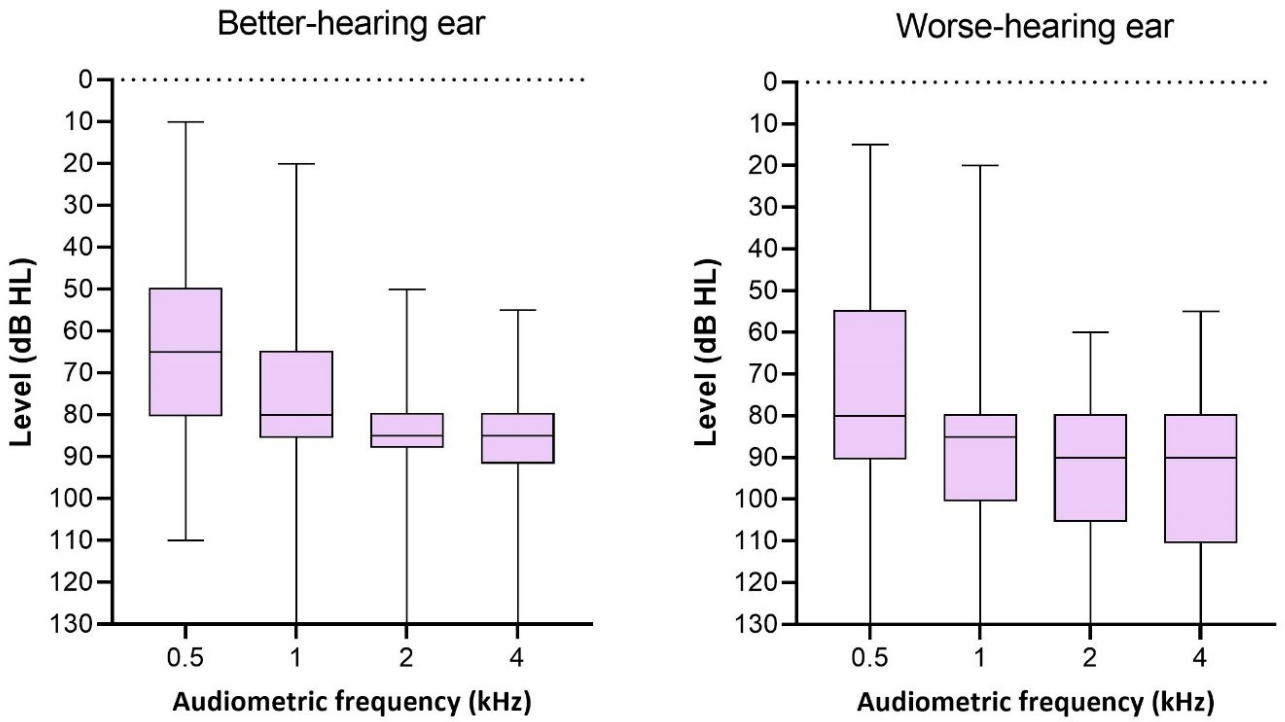


Figure 4.12: Distributions of better- and worse-hearing ear HTLs by audiometric frequency
For each audiometric frequency, the median HTL is shown by the horizontal black line. The 25th to 75th centiles are shown via the boxes and the whiskers represent the range of HTLs.

A greater divergence in HTLs between better- and worse-hearing ears can be seen with increasing audiometric frequency, for those with HTLs below the median.

Figure 4.13 shows the difference in decibels between the 4-frequency average HTLs of the better- versus worse-hearing ear for each of the 48 children with ear-specific data available. 75% of the group had a difference of less than 12 dB between ears, and the remaining 25% had a difference of between 15 dB and 63 dB, indicating asymmetric hearing loss (Smyth & Patterson, 1985).

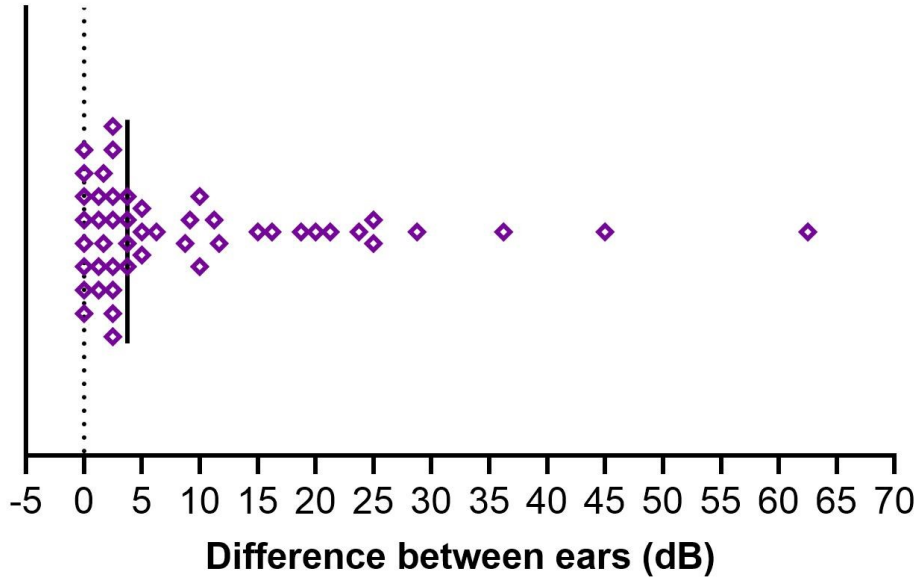


Figure 4.13: Difference between better- and worse-hearing ear 4-frequency mean HTLs

The difference between better- and worse-ear 4-frequency mean HTL averaged over 0.5, 1, 2 and 4 kHz is plotted as a diamond for each child. The group median difference is shown as a vertical black line.

For this group, eligibility screening forms were reviewed to identify whether the worse-hearing ears' HTLs fell within TAG166 (2009). Twelve children's worse-hearing ears could not be classified by TAG166 due to either missing

HTLs at 2 and/or 4 kHz, or because their hearing had only been tested in soundfield. Of the remaining 42 children, 13 (31%) had a worse-hearing ear that fell within TAG166 criterion for HTLs ≥90 dB HL at both 2 and 4 kHz and the remaining 29 (69%) had HTLs that fell outside TAG166 (2009) on both sides.

4.3.6. Summaries of group characteristics

Children newly eligible for CIs under TAG566 (NICE, 2019)

The 54 children newly eligible for CIs, 4.4% of the cohort, are those whose audiometric eligibility for cochlear implantation changed due to the revision of the NICE guidance. Nine of this group could not be classified by TAG166 (2009) because of missing HTLs at 2 and / or 4 kHz. The remaining 45 had been confirmed to have audiograms that placed them outside TAG166 (2009) but within TAG566 (2019). Together, these children had a median 4-frequency average HTL of the better-hearing ear within the severe range. Compared to the group outside the 2019 guidance, they were similar in terms of their age distribution and the proportion with additional needs, but family members were more likely to need a spoken or sign language interpreter. This group had a higher proportion of HTLs measured at 3 kHz than those outside TA566 (NICE, 2019) or those who could not be classified by it. Twenty-five percent of them had asymmetric hearing loss.

Children whose eligibility was unknown by TAG566 (NICE, 2019)

This group of 85 represented 7.0% of the HA caseloads. The number of children screened whose CI eligibility status had changed from being known to

being unknown (N=68) was greater than the number of children known to have been brought newly within criteria (N=54). This group had a median 4-frequency average HTL in the moderate range. Compared to the other NICE eligibility groups, children who could not be classified by the 2019 guidance were younger on average. They were the group most likely to have additional needs, with these affecting 78% of those aged 5 years or older.

Children previously eligible for CIs under TAG166 (NICE, 2009)

These 37 children comprised 3.0% of the cohort. Children using HAs who met 2009 guidance tended to be older than the wider cohort, comprising a small number of infants and with an age profile that gradually increased up to the 15- to 17-year-old category. Around half of the families needed sign-language interpreting, the highest of any NICE eligibility group. Recall, the screening form did not ask audiologists to specify which family member(s) needed the interpreters. However, optional free-text comments for this group often indicated that the BSL interpreters were needed for the children. This might indicate that some children within this group had gained insufficient benefit from HAs to develop spoken communication. This group also had the highest proportion of families who needed a spoken-language interpreter. Of those aged 5 years or older, 23% had additional needs.

4.4. DISCUSSION

This chapter assessed the impact of TAG566 (NICE 2019) on children's eligibility for CIs, by studying children without access to cochlear implantation. The results showed that TAG566 (NICE, 2019) widened access

to cochlear implantation and also increased the number of children whose eligibility for CIs was unknown.

4.4.1. Characteristics of paediatric hearing aid users

The first chapter objective was to document the demographic and audiological characteristics of whole caseloads of children using HAs. The findings of the present study will be discussed in relation to previously published data, including the CRIDE report (CRIDE, 2021) and the epidemiological study published by Fortnum et al. (2001).

4.4.1.1 Age distribution

The age distribution of children using hearing aids pooled across all sites in this study is similar to that found by Fortnum et al. (2001) for all children born between 1980 and 1995 in the UK with permanent hearing loss of 40 dB HL or greater in the better-hearing ear during 1998-99. Prevalence gradually increased from birth to around 9 years of age, beyond which it plateaued then declined for older teenagers. Fortnum et al. (2001) attributed the decline to some older teenagers having transitioned from paediatric to adult audiology services and / or left the education system, thereby not having been captured in their data collection. This roll-off was found for both the Huddersfield & Calderdale and Nottingham caseloads in the present study, but not for Bradford. This is likely to represent differences in local practice regarding the discharge of older teenagers to adult audiology caseloads (which were not included in screening) rather than an actual decrease in the prevalence of

hearing loss for older teenagers compared to children over 9 years and younger teenagers. It is not possible to determine the reasons for the greater proportion of children using hearing aids aged 6 to 8 years at Nottingham compared to the other hospitals in this study or compared to the Fortnum et al. UK-wide study. It is possible that the discrepancy reflects a difference in local management of children with temporary or single-sided hearing loss, who were excluded by (Fortnum et al., 2001) but included in the present study.

4.4.1.2. Family need for interpreting services

The proportion of families needing any interpreter varied significantly between sites, from 20% of the Bradford caseload to 2% at Huddersfield. This could reflect geographical differences in ethnic diversity and/or differences in record-keeping between sites that made it easier for audiologists to extract this information at some hospitals than others. The present study documented interpreters needed for Audiology appointments. In contrast, the CRIDE report collects information from deaf education services regarding the number of children who are known to have English as an additional spoken language at home, and information on the use of sign language versus spoken English in the child's educational setting. The CRIDE report (2021) reported that 88% of children used spoken English only at school, 2% used BSL only, 7% used spoken English together with signed support and 3% used another (unspecified) combination of signed and spoken communication. Hence 12% of children in the CRIDE report used some form of sign at school. In contrast, only 2% of the families screened in the present study used sign interpreting for audiology appointments. It is possible that this is due to BSL interpreters more

commonly being requested for older deaf children only, or that the parents of children used to communicating via spoken language with signed support do not feel it is necessary to request BSL interpreters for all hospital appointments.

In terms of spoken languages, the CRIDE report found that 14% of children with hearing loss in England were from homes where English was an additional spoken language. This is similar to the proportion of families (12%) who needed a spoken language interpreter for appointments from our cohort. However, it is important to note that not all families who speak English as an additional language will have been captured by the CUSP eligibility screening form, as it is unlikely that all of them need formal interpreting services. It is therefore likely that the CUSP cohort included a larger proportion of children from homes where English is a second language than is representative of England as a whole.

The most frequently requested interpreter language categories were South Asian and Eastern European. It is not possible to accurately determine children's ethnicity based on the interpreter language data. However, it is relevant to note that children of Pakistani or Bangladeshi ethnicity are more likely to have hearing loss than children from White or Black families (Butcher et al., 2018; Morton et al., 2002), as are Roma children (Latorre-Arteaga et al., 2017; Mašindová et al., 2015). Children's hearing loss can be under-diagnosed in Roma communities (Swanwick et al., 2021) and so the data in the present study may underestimate the true number of children with hearing loss who might be eligible for HAs in the geographical area served by these hospitals.

4.4.1.3. Additional needs

(Fortnum, Marshall, & Summerfield, 2002) found that 27.4% of the children with permanent hearing loss of moderate degree or greater had at least one other disability and believed this to be an underestimate due to missing data. They based this estimate on information from clinical and/or educational professionals and included any kind of physical, learning, communication or other difficulty. In contrast, the CRIDE report presents data on children who meet the government definition of special educational needs (SEN). This was estimated to be 23% of deaf or hard-of-hearing children in England (CRIDE, 2021). In the present study, data on additional needs were not collected for children aged under 5 years to meet ethical restrictions because it was not needed to establish their eligibility for the wider study. Screeners chose "Yes" or "No" to the presence of additional needs that influenced hearing test participation and then gave a free-text, general description of these in a non-specific way that would not allow any child to be identifiable. Children aged 5 years or older with missing data regarding additional needs in the present study were classed as not having additional needs, the same method used by (Fortnum, Marshall, & Summerfield, 2002). Using this approach, the 182 children with additional needs affecting their ability to do hearing tests comprised 20% of the CUSP cohort.

4.4.2. Prevalence of children affected by TAG566 (NICE, 2019)

This section will discuss the findings related to the second chapter objective, to calculate the prevalence of children using HAs who were newly eligible or of unknown eligibility for cochlear implantation following the implementation of

TAG566 (NICE, 2019). The results confirmed that TAG566 (NICE, 2019) widened access to cochlear implantation because the percentage of children using HAs who were audiometrically-eligible for CIs based on their air-conduction HTLs increased from 3.1% to 7.5%. This indicates that more children now meet the BCIG guidance for referral for CI eligibility assessment (British Cochlear Implant Group, 2019). The consequences of this are that staff at local HA services need to be aware of the new guidance, so that offers of CI eligibility assessment can be made to the newly eligible families.

There are limitations associated with this data that affect its ability to estimate how many additional children will now receive CIs as a consequence of TAG566 (NICE, 2019). First, the analysis is based solely on air-conduction thresholds and does not account for children with mixed hearing loss, because too few sites were able to extract tympanometry data due to resource constraints. Hence the study reveals the number of children with functional hearing loss within NICE criteria, but has not captured the proportion with temporary or permanent conductive overlays that might affect children's candidacy for CIs. Second, due to the COVID-19 pandemic, the period of time during which screening took place was longer than planned, taking until as late as the end of March 2021 in the case of Calderdale. While sites were requested to screen children with severe or profound hearing loss earlier on, this was not always possible due to limitations of record keeping systems. So some children who had become newly eligible for CIs might have already been referred for CI assessment prior to being screened and might have been missed.

Despite these limitations, to the author's knowledge at the time of writing, these data provide the best available evidence to indicate what proportion of

children using HAs might now be newly eligible for referral for CI eligibility assessments, which is helpful information for service providers.

4.4.3. Characteristics of children affected by TAG566 (NICE, 2019)

This section will discuss the findings related to the third chapter objective, to document the demographic and audiological characteristics of children using HAs who were: newly eligible for CIs; of unknown eligibility for CIs; and those who had been previously eligible for CIs but who remained without access to cochlear implantation.

4.4.3.1. Characteristics of children newly eligible for cochlear implantation

This group comprised children whose audiometric eligibility for CIs under TAG166 (2009) was either not known (N=9) or who fell outside TAG166 (2009) (N=45) and whose HTLs met TAG566 (2019) i.e., they became newly eligible for CIs as a result of TAG566 (2019). Their age distribution was similar to the wider cohort of HA users. However, in contrast to HA users as a whole, 11% of this group needed sign language interpreters for audiology appointments. This could mean that some of these children, who were not previously eligible for CIs, have been unable to develop spoken language via HAs. If this is the case, it may limit their access to CIs now that they are audiologically-eligible as it is possible that CI services might decide that it is too late for them to acquire spoken language skills (Hellman et al., 1991).

The higher proportion of this group who need spoken language interpreting compared to those outside TAG566 (2019) could be caused by a higher proportion of children from different ethnic groups experiencing severe or profound hearing loss than milder degrees of loss. This has been proposed as being the case for the Roma community (Swanwick et al., 2021) although the known under-ascertainment of hearing loss in this group, acknowledged by the authors, might disproportionately affect those with mild or moderate losses and so this cannot be established with certainty. The higher proportion of families needing spoken language interpreters in this group could also have arisen if there had been inequitable access to CIs for children outside TAG166 before the introduction of TAG566. In other words, if a greater proportion of children from families with good English proficiency who had HTLs close to but outside TAG566 (2019) progressed to cochlear implantation via applications for special funding compared to children from families with poor English language skills. This cannot be known from the current dataset. However, as proposed by Schuh & Bush, (2021), it is plausible that this might be the case because language barriers are related to poorer utilisation of healthcare services and care adherence (Timmins, 2002).

One quarter of this group had asymmetric hearing loss. Of those with ear-specific data available, a difference of ≥15 dB in better- versus worse-hearing ear 4-frequency HTLs was seen for 25%, and 31% had a worse-hearing ear that met the audiometric criterion of TAG166 (2009). It is not possible to know from this dataset for how long these children had experienced greater auditory deprivation on their worse-hearing side. Asymmetrical hearing loss has been identified as a barrier to the provision of optimal binaural hearing in children

(Gordon et al., 2021). It is known that asymmetrical auditory deprivation can lead to aural preference syndrome, (Anderson et al., 2022; Gordon et al., 2015) which is associated with poorer speech perception and spatial listening ability (Polonenko et al., 2018). This creates a risk that a quarter of those newly eligible for CIs, if they proceed to cochlear implantation, might have poorer spatial listening outcomes than their peers with symmetrical losses.

4.4.3.2. Characteristics of children whose eligibility for cochlear implantation is unknown

The requirement to measure up to ten HTLs to establish children's eligibility for CIs resulted in seven percent of the caseloads screened being of unknown eligibility for cochlear implantation. This group had the highest proportion of children with additional needs and was younger on average than the other NICE eligibility groups. The potential impact of any additional needs on a child's ability to benefit from CIs is considered by CI services within the ChIP framework during CI candidacy assessments (Hellman et al., 1991; Lazaridis et al., 2010). However, children with additional needs can be given access to cochlear implantation (Meinzen-Derr et al., 2010; Rawes et al., 2021). Optimising access to sound at as early an age as possible is crucial to help children develop the best possible language skills (Yoshinaga-Itano et al., 2020). It is therefore important for both HA and CI services to be aware that this group is now vulnerable to having less equitable access to cochlear implantation than under TAG166 (NICE, 2009). It is also important to establish whether outcomes for children in this group are significantly different to similar children using CIs, to guide how the uncertainty regarding their

suitability for CIs can be resolved. This will be discussed in more detail in Chapter 7.

4.4.3.3. Characteristics of children without access to cochlear implantation who had been previously eligible.

This chapter included the objective to characterise children who were using HAs despite being audiometrically-eligible for cochlear implantation. Such analysis might reveal characteristics among this group that had created a barrier to accessing implantation, and if so, help identify potential barriers to implantation for the children who had become newly eligible or whose eligibility remained unknown. Unlike some other countries, the NHS provides access to CIs for any person who meets the NICE guidance and is deemed an appropriate candidate by their clinical team, with no family purchase or medical insurance needed. However, despite this, 3% of the whole cohort of HA users screened had HTLs within TAG166 (NICE, 2009) and yet did not have access to cochlear implantation.

This group's characteristics suggested they may have comprised: infants with congenital profound losses; children with progressive losses recently found to have HTLs within NICE criteria and yet to be offered CIs; older children for whom cochlear implantation may not be appropriate either due to long duration of deafness and/or the presence of additional needs that may preclude cochlear implantation; and children whose families had declined CIs. The most marked difference between this group and the wider cohort was that they had the greatest need for both spoken and signed language interpreting services.

The most likely reason that half of the families in this group needed BSL interpreters was that HAs gave these children insufficient access to sound for them to develop spoken language proficiency. However, it is not possible to determine what proportion of the BSL interpreters were needed for parents rather than children, nor for which families the use of BSL was through choice. The number of children with HTLs meeting the 2009 guidance was small (N=37) and so their characteristics should be interpreted with caution. However, further research is warranted to better understand whether these children's characteristics reflect any past or present inequality in children's access to CIs in the UK.

4.4.4. Summary of potential limitations and biases

Limitations in the audiometric data included insufficient information to establish the prevalence or degree of conductive elements to children's hearing losses and no historical data to show how the losses had progressed over children's lifetimes. Further, only a small proportion of the population of England and Wales was included in the sampled caseloads. The areas included are not expected to be socio-economically representative of the wider population (Office for National Statistics, 2023), limiting the generalisability of the findings. Last, the COVID-19 pandemic extended the planned period of data collection. This meant that some potentially eligible teenagers might have become ineligible due to passing their 18th birthday before they were screened, and some children using HAs at the start of data collection could have progressed to implantation.

4.5 SUMMARY

This chapter assessed the impact of TAG566 (NICE 2019) on children's eligibility for CIs, by studying children without access to cochlear implantation. A group of children became newly eligible for CIs, and more children became of unknown eligibility. There were also children who had previously been eligible for CIs but who had not received them, and those who remained ineligible. Each group had distinct characteristics. The next chapter compares the outcomes of children from each of these groups to children using CIs, to inform CI candidacy decision making.

CHAPTER 5: COMPARATIVE CAREGIVER-REPORTED OUTCOMES OF CHILDREN WITH AND WITHOUT ACCESS TO COCHLEAR IMPLANTATION

5.1 INTRODUCTION

Comparing the outcomes of children using CIs with children using HAs who are close to CI eligibility is important because any discrepancy between the groups indicates some children are not receiving the optimal intervention. The scoping review in Chapter 2 identified five studies of parent-reported functional auditory performance of children with severe hearing loss using HAs and children using CIs.

The largest study was conducted in Australia by Wong et al. (2017). They found no significant difference in Parents' Evaluation of Aural/Oral Performance of Children (PEACH) scores for children with CIs compared to a group of 46 HA users with severe hearing loss (defined as three-frequency average HTLs of 60 to 80 dB HL). Similarly, in Germany Meister et al. (2015) found no significant difference between a group of 38 CI users and a group of 14 HA users with severe hearing loss (defined in line with the WHO; four-frequency average HTLs of 60 to 80 dB HL), nor between children with moderate hearing loss using HAs and children with CIs, assessed via the Functioning after Paediatric Cochlear Implantation (FAPCI) questionnaire. In India, Jallu et al. (2019) found significant improvements in Categories of Auditory Performance (CAP) scores over time for both HA users with severe hearing loss and CI users, however no comparison was made between the HA

and CI groups. Similarly, Trevisi et al. (2016) included four HA users with severe hearing loss and seven CI users in a study of children with CHARGE syndrome, reporting modest increases in Auditory Perceptive Performance (APP) scores over time for most children in both groups, but not making direct group comparisons. Only one study was based in the U.K., presenting case studies of five children before and after implantation (Tzifa & Hanvey, 2013). CAP scores increased by one point for four children and from 0 to 5 for the other, but statistical analysis was not performed. Clinical and educational practice varies between countries. It is important to establish comparative outcomes for children with CIs compared to children close to CI eligibility from the U.K. and to use NICE CI eligibility to define children's hearing. Chapter 2 identified that no studies had been published comparing tinnitus or listening fatigue outcomes for children with severe hearing loss using HAs to children using CIs. The primary goal of hearing devices is to improve listening and language development, but these outcomes are also influential in children's lives (Davis et al., 2021; Smith et al., 2019) and therefore important to understand, especially if they could plausibly be influenced by the provision of CIs versus HAs.

The purpose of this chapter was to fill these gaps in the evidence base, by comparing parent-reported functional listening, tinnitus, and listening fatigue outcomes for children using CIs to children using HAs with hearing close to or within CI eligibility, within the context of the NHS. Chapter 2 revealed that most comparative studies group together HA users with a wide range of hearing loss, rather than focussing on those close to CI eligibility. This study therefore aimed to recruit children using HAs with HTLs within or just outside

TAG566 (NICE, 2019). However, to make the sample representative of the clinical population, this project aimed not to exclude children on the basis of socio-economic characteristics, or the presence of any additional needs. A benefit of recruiting families via the screening forms described in Chapter 4 was that, unlike most studies, it was possible to document the characteristics of the respondents compared to the eligible population, giving an indication of the generalisability of the findings across the NHS.

In summary, the aim of this chapter was to compare the clinical effectiveness of HAs for children with hearing within or close to eligibility for CIs with the clinical effectiveness of CIs. This was to be achieved via the following objectives:

(i) Identify children using CIs and children using HA with HTLs within or close to NICE eligibility for cochlear implantation and recruit their caregivers to participate in a survey

(ii) Characterise the children represented in the survey, and their caregivers, demographically and audiologically

(iii) Compare children's caregiver-reported functional listening, tinnitus, and need for recovery from listening, controlling for potentially confounding variables

(iv) Compare caregiver satisfaction with children's current hearing devices and child acceptance of devices between the CI and HA groups

(v) Document the family need for interpreters and prevalence of additional needs of the survey sample and the whole eligible population

5.2 METHODS

5.2.1. Survey development

The survey was piloted and refined based on the feedback of members of the public and parents of children with hearing loss, to ensure that the questions were clear and that the time needed to complete the survey was acceptable. This advice including offering all families the choice to complete the survey either online or on paper (to be returned using a pre-paid return envelope). This was advised by both NHS professionals and parents of children with hearing loss, who suggested it would help families with limited or no access to the internet to participate. This had been raised as important before the pandemic, during which issues of digital poverty and the need for multiple family members to share devices for work, home-schooling, socialising and entertainment might have made the paper option even more valuable.

5.2.2. Recruitment sites

Each site that had agreed to undertake whole caseload screening for Chapter 4 also agreed to recruit to the survey. This included Bradford, Nottingham, Huddersfield and Calderdale. In addition, Sheffield Children's Hospital agreed to return eligibility screening forms for children known to have severe or profound hearing loss, and to recruit eligible children. Children with cochlear implants were recruited from the regional Yorkshire Auditory Implant Service (YAIS) where all children who had been implanted at Bradford were screened. Last, the Midlands Hearing Implant Programme based at Birmingham agreed

to perform screening and recruitment for children using HAs under assessment for CI, and of a sub-set of their children using CIs whose data was readily available via a research database.

5.2.3. Eligibility screening

The HA eligibility screening form has been described in Chapter 4, and aimed to characterize children in terms of age, sex, device use, family interpreter need, additional needs, and audiometric thresholds. The CI forms, like the HA forms, were Microsoft Word documents with pre-defined fields and drop-down options or calendars where appropriate. The CI eligibility screening form included the same characteristics fields as the HA form but omitted audiometric thresholds. Every HA and CI eligibility screening form returned by a site was reviewed. If information was missing, unclear, or appeared to be inaccurate or internally contradictory, the audiologist named on the form was asked for clarification.

5.2.4. Inclusion and exclusion criteria

For the hearing aid group, inclusion criteria were:

- Aged under 18 years at the time of eligibility screening
- Unaided air-conduction HTLs of ≥ 60 dB HL for at least two frequencies including 0.5, 1, 2, 3 and 4 kHz in both ears.
- Using at least one acoustic HA and no other type of device

For the cochlear implant group, inclusion criteria were:

- Aged under 18 years at the time of eligibility screening
- Using either: two CIs; one CI alone; or one CI with an acoustic HA in the contralateral ear

For both the HA and CI groups, the exclusion criterion was if the child used a bone conduction aid. The audiometric criterion for the HA group meant that all children who could be confirmed to meet either the 2009 or 2019 NICE guidance were eligible for the survey, plus children with up to 20 dB better hearing. The scoping review described in Chapter 2 had identified that there were few published studies of comparative outcomes for children with severe hearing loss using HAs to children using CIs that included children with additional needs. To address this gap, the presence of any kind of additional need was not an exclusion for families to participate.

5.2.5. Rationale for survey characterisation data and predictor variables

The opening section of the survey was designed to characterise children in more detail than was possible from eligibility screening form data, to enable comparison of the participants to populations described in other publications, to enable readers to compare this sample to their own populations of interest, and to document as many as possible of the characteristics that might need to be controlled for in comparisons between the HA and CI users. This section lists the characteristics and predictor variables collected, with explanations of the reasons for their inclusion. The questions used can be seen in Appendix B.

5.2.5.1. Children's age and sex

This information was already on the eligibility screening form, but was asked again within the survey as a cross-check of children's identity[2] and to ensure that the child's age was correct at the time of survey completion, in case a long time elapsed between screening and survey completion. It was planned to control for age in group comparisons as children's listening skills improve through childhood (Ching & Hill, 2007). Sex assigned at birth has been associated with the incidence of tinnitus in children (Nemholt et al., 2020).

5.2.5.2. Child's academic year education provision or employment status

The child's stage of education was collected to allow comparison to other populations and to identify any differences between the groups. It was plausible that these would differ, as Fortnum, Marshall, Bamford, et al., (2002) found children's educational placement varied as a function of hearing loss, including the proportions of children educated via aural/oral, Sign or sign-supported English, within mainstream, resource units or specialist schools.

5.2.5.3. Children's expressive communication mode and receptive communication modes at home and school

This was asked so that the proportions of children communicating via spoken language alone, spoken English supported by signs, or by sign-language alone could be compared between the groups. Communication mode used in school

[2] In one case I identified that a survey had been accidentally posted to the wrong child by the local clinical team because the age and gender on the survey did not match the screening form for that ID code. The local team double-checked and confirmed that the child the survey related to was not eligible for the study, and so a duplicate pack was sent to the correct family, and the first survey was excluded from analysis.

could then be compared to both the current CRIDE report (CRIDE, 2021) and data from a comprehensive study of deaf and hard-of-hearing children's outcomes conducted by (Stacey et al., 2006). Children learning via either auditory-oral or visual communication has been associated with oral / aural outcomes on the PEACH (Kuschke et al., 2022). Children's listening effort is affected by the classroom's auditory environment, clarity of teacher's voices, and access to lip-reading (Rudner et al., 2018). It was therefore plausible that need for recovery from listening might be less in a sign-based setting, and so any differences between the groups should be known when comparing this outcome.

5.2.5.4. Children's main spoken language at home

English being spoken as an additional language has been associated with poorer access to healthcare and healthcare outcomes in general (Timmins, 2002) and with poorer speech perception outcomes, greater delays in hearing intervention, and reduced access to hearing rehabilitation services for children (National Deaf Children's Society, 2021b; Turner & Lynas, 2000; Wu et al., 2015). Chapter 4 revealed that the proportion of families needing certain spoken language interpreters increased with increasing degree of children's hearing loss. Further, survey families from the HA and CI groups were not recruited from the same geographical areas, making it likely that the proportions of children from homes where English was an additional language would differ between groups. Notes from Audiology records regarding need for interpreters was likely an underestimate of families where English was not

the first language, so it was important to collect caregiver reports of the main spoken language.

5.2.5.5. Type of additional needs

Caregiver reports of any additional needs were collected as a more accurate measure than the eligibility screening data. Knowledge of the type of children's additional needs was planned to enable comparison with the populations in other studies. Eligibility screening forms contained information on additional needs affecting children's ability to take part in hearing tests, but this was expected to be an underestimate of the true prevalence of any additional needs. The survey provided a list of types of additional needs (autism, learning difficulties, visual impairment, physical disability; behaviour emotional or social difficulties, and speech language and communication needs) to allow classification into the additional needs groups defined by Cupples, Ching, Leigh, et al., (2018) to assess the impact of additional needs on the outcomes of children with hearing loss. Caregivers could specify other types of additional needs if required, and could select more than one additional need for each child. Classifying children as having either no or at least one additional disability was found to be a significant predictor of PEACH scores (Kuschke et al., 2022) and so was intended to be a predictor variable for modelling PEACH outcomes. The presence of some additional needs can also be a cause for a CI team to decline to offer cochlear implantation, and so it was plausible that the number and types of additional needs might vary between HA and CI groups.

5.2.5.6. Cause of children's hearing loss

Aetiology was not significantly associated with unaided HTLs in a study of 364 three-year-old children in Australia (Dahl et al., 2013) and so no difference between the HA and CI groups was anticipated. Further, the oral / aural outcomes of children using CIs have not been found to be associated with genetic variants including SLC26A4 and enlarged vestibular aqueduct syndrome (Chiong et al., 2018), nor with auditory neuropathy spectrum disorder (ANSD) (Bo et al., 2022; Dahl et al., 2013). However, some studies report that children's functional listening can be influenced by aetiology. Hearing loss due to cytomegalovirus infection was associated with better than average speech perception in noise for children using CIs (Varga et al., 2014). Caregivers were therefore asked to report these data, to allow for comparison between the groups and to aid the interpretation of the findings. The survey included pre-specified options from which respondents could choose more than one and choose the option that the cause was unknown to the hospital, or that the respondent could not remember the cause.

5.2.5.7. Age at identification of hearing loss and age at intervention with device

Early identification of hearing loss and intervention are crucial for children to achieve the best possible spoken language development and delays in either, resulting in longer-duration auditory deprivation, can influence children's outcomes (Ching et al., 2013; Yoshinaga-Itano et al., 1998, 2018). Caregivers were therefore asked for their child's ages at identification and intervention in

years and months, with the option to state that they could not remember, in recognition that this was a retrospective observational survey.

5.2.5.8. Daily hearing device use

The amount of a child's waking hours that a hearing device is used for is associated with their aural / oral outcomes, including children's scores on the PEACH (Kuschke et al., 2022). One of the items within the PEACH asks caregivers for daily device use as an ordinal variable as a proportion of the day (Ching & Hill, 2007).

5.2.5.9. Caregiver-responder and other main caregiver relationship to child

A CI team's perception of the quality of a child's family and school support is used when deciding whether to allow the child access to cochlear implantation (Hellman et al., 1991; Nikolopoulos et al., 2004). Further, a study from the USA found that children using HAs were more likely to be referred for CI eligibility assessment if they had married parents compare to non-married parents (Wiley & Meinzen-Derr, 2009). Therefore the relationships to the child of both the responder-caregiver (stipulated on the information sheet and in the survey introduction to be a main-caregiver to the child) and up to one other main caregiver were included. The term 'main caregiver' rather than 'mother' or 'father' was used to avoid false assumptions about family composition. The responder-caregiver could then choose how to describe their relationship to the child and that of any other main-caregiver. For each, pre-specified options included 'mum', 'dad, 'grandmother' 'grandfather' or 'other, please specify'.

5.2.5.10. Caregiver-responder and other main caregiver self-described hearing status and hearing device use

Attitudes toward CIs versus HAs can differ between parents who are deaf and those who are hearing (Hardonk et al., 2011), so caregivers' self-reported hearing status and hearing device use was collected via accessible question and response options that were approved by the PPI contributors.

5.2.5.11. Caregiver highest educational qualification

Parents' educational attainment is associated with children's functional listening skills (Cupples, Ching, Button, et al., 2018; Noblitt et al., 2018), language development (Dwyer et al., 2019), and daily hours of hearing device use (Marnane & Ching, 2015; Wiseman & Warner-Czyz, 2018). Caregiver highest educational qualification was therefore collected so that differences between the groups could be identified, and this could be controlled for in comparative analyses. Respondents were also given the option to respond *'prefer not to say',* in case this question seemed intrusive.

5.2.5.12. Previous or ongoing cochlear implant candidacy assessment

It was possible that some survey respondents would be carers of children using HAs who had undergone CI candidacy assessments but had not received CIs, or of children whose eligibility for cochlear implantation was ongoing at the time of survey completion. This information was collected, along with whether the responder-caregiver had ever considered CI eligibility assessment for their

child, to identify whether previous CI assessment was associated with caregiver satisfaction with the child's current hearing device.

5.2.6. Rationale for survey outcome measures

This section provides justifications for the outcome measures selected or developed for inclusion in the survey, along with details of the novel questions devised to assess tinnitus and need for recovery from listening.

5.2.6.1. Listening outcome measure selection

There are several caregiver-report measures available to assess children's oral / aural outcomes. It was important to select one that was appropriate to both the study aim, comparing hearing interventions, and the population being studied, which included children with a wide range of ages and developmental abilities. The tool therefore needed to be sensitive to the effects of hearing devices and appropriate for all children aged less than 18 years. Instruments often used in the CI literature include the FAPCI (Lin et al., 2007), and the Meaningful Auditory Integration Scale (MAIS) (Robbins et al., 1991) or the MAIS version for infants and toddlers (IT-MAIS) (Zimmerman-Phillips et al., 2000). However, the FAPCI is targeted at 2- to 5- year-olds, rather than the wider age range of children in this study. The MAIS and IT-MAIS were developed for children with profound deafness. There is evidence that children with other degrees of hearing loss might demonstrate ceiling effects on this test (Zheng et al., 2009). Alternative tools for children with a wide range of hearing losses include the Auditory Behaviour in Everyday Life (Purdy et al., 2002) and the

Parent version of the Children's Speech, Spatial and Qualities of Hearing questionnaire (Galvin & Noble, 2013). However, these were not appropriate for the full range of ages of children in the present study. In contrast, the Parents Evaluation of Aural/Oral Performance of Children (PEACH) (Ching & Hill, 2007) was developed to quantify the effectiveness of hearing devices for children over wide range of both age and degrees of hearing loss. PEACH scores correlate with average unaided HTLs and both normative data and the change score constituting a critical difference are documented (Ching & Hill, 2007). For these reasons, the PEACH was selected as the listening outcome measure for the CUSP survey.

5.2.6.2. Tinnitus outcome measure development

At the time of study development, there was no validated tool for assessing tinnitus in children and no consensus on its optimal assessment (Rosing et al., 2016). Subsequently, it was necessary to devise novel questions for the survey. The description of tinnitus was guided by the British Society of Audiology Paediatric Tinnitus Guidance (Benton et al., 2015) which advises audiologists to ask children if they *"hear noises in their ears or head"*. Because in this study parents would be answering questions independently, not in a face-to-face interview as was the context for the BSA guidance, this phrase was qualified by adding *"...that don't come from their hearing aids, cochlear implants, or surroundings (tinnitus)?"*. This wording was adapted from a study by Graham (1981). The first CUSP survey tinnitus question was intended to establish the presence or absence of tinnitus, so that any difference in prevalence between the HA and CI groups could be identified. To further

characterise children's experience, additional questions were devised that parents should be able to answer on behalf of their child. Smith et al., (2019) catalogued tinnitus-related problems experienced by children via a scoping review. They identified impacts across domains including physical health, cognitive health, hearing and listening, emotional health and quality of life. Of the impacts identified, the emotional impact of tinnitus causing annoyance, and its health effect on sleep, seemed the most likely for a parent to be aware of. Questions and response options were drafted that were honed with feedback from researchers and the PPI contributors. The final questions ensured parents were not forced into giving inappropriate or inaccurate responses by allowing options such as *'My child is not able to tell me' and 'I don't know, we haven't talked about it'* instead of forcing *'yes'* or *'no'* responses. The final questions and response options used were:

"Has your child ever told you that they hear noises in their ears or head that don't come from their hearing aids, cochlear implants, or surroundings?"

- *Yes*
- *No*
- *My child is not able to tell me*

Only if parents answered "Yes" were they asked to answer the subsequent questions:

"Does your child say that they find these noises annoying?"

- *Yes, my child has told me they find the noises annoying*
- *No, my child has told me that they do not find the noises annoying*
- *I don't know, we haven't talked about it*

"Does your child say that these noises disturb their sleep?"

- *"Yes, my child has told me that these noises disturb their sleep"*
- *"No, my child has told me that these noises do not disturb their sleep"*
- *"I don't know, we haven't talked about it"*

5.2.6.3. Need for recovery from listening outcome measure development

Fatigue is a complex, multi-dimensional concept, including physical, cognitive, emotional, relational, and physiological impacts, and being related to vitality and vigour (Bess et al., 2016; Stein et al., 2004). At the time of survey design, no validated tool for assessing listening-related fatigue had been developed for children or adults. Novel questions were therefore devised, based on the literature, PPI contributors, my clinical experience, and advice from researchers of fatigue and hearing loss.

For this proxy-report survey of children with a wide range of ages and developmental ability, it did not seem appropriate to expect caregivers to have accurate insight into all their children's cognitive, emotional, or relational experiences. Indeed, significant differences between parent-reported and child-reported listening fatigue have been documented (Werfel & Hendricks, 2016). The survey questions therefore focussed on need for recovery from listening, one aspect of listening fatigue identified in adults (Holman et al., 2019; Nachtegaal et al., 2009). Adults with hearing loss have higher need for recovery following work than adults without hearing loss, and it has been associated with short and long term health and psychological issues (Nachtegaal et al., 2009).

Need for recovery from listening can include wanting a rest from listening or experiencing headaches. When children remove their hearing devices it is visible to a parent. It was also likely that children would seek comfort from their caregiver for symptoms such as headaches, and that parents would be familiar with their child's descriptions of, or behaviour related to, tiredness. Questions were worded to avoid asking parents if their child e.g., 'felt' tired,

but rather to gauge whether children had demonstrated behaviours or volunteered information related to their need for recovery. Questions were intended to be appropriate across a spectrum of developmental ability. For example, to 'want a break' from listening could be communicated via signed or spoken language explanations, or by a non-verbal child habitually removing their devices following certain activities. Children sometimes remove their hearing devices due to broader behavioural or device rejection issues (Muñoz et al., 2016). It was therefore important for this question to discriminate between the child removing devices to provide a rest from listening and the child rejecting a device or objecting to being in a particular situation. Adults with listening fatigue often describe it as being experienced at the end of the day (Holman et al., 2019). The survey therefore asked how often the child wanted a break from their device following activities, rather than in general. As with the tinnitus questions, a response option was provided for caregivers who felt their child was not able to communicate their need for recovery, so respondents were not forced into inaccurate responses. Advice from PPI group members and researchers led to the inclusion of the response option 'Never' as well as the others. The experts who approved my questions included an author from the team who have since published the only validated tool to assess listening fatigue in children (Hornsby et al., 2022). The final questions and response options were:

"Following their usual activities, such as childcare / school / studying / work, how often does your child want a break from using their right/left hearing aid/cochlear implant?"

- *Never*
- *Hardly ever*
- *Sometimes*

- *Frequently*
- *They don't have a [HA/CI] for their right/left side*
- *My child is unable to tell me*

"How often does your child complain of headaches related to listening?"

- *Never*
- *Hardly ever*
- *Sometimes*
- *Frequently*
- *My child is unable to tell me*

"How often does your child complain of tiredness related to listening?"

- *Never*
- *Hardly ever*
- *Sometimes*
- *Frequently*
- *My child is unable to tell me*

Since survey design and distribution, both tiredness and headaches have been formally documented as some of the physical experiences of listening-related fatigue in children with hearing loss, and removing hearing devices as one of children's common coping strategies (Davis et al., 2021).

5.2.6.4. Child acceptance of and caregiver satisfaction with the child's hearing device(s)

The survey sought to measure parent satisfaction with the child's current hearing device. This was to provide insight into families' experiences of children's device use and a context for the potential impact of the thesis. It was plausible that caregivers of children using HAs would be less likely to consider an offer of CIs favourably if they were very satisfied with HAs, especially given the more invasive nature of CIs. For parents deciding whether to accept

CIs for their children, greater parental stress is associated with children having better outcomes with HAs, and lower stress is experienced by parents who know HAs do not give their child access to spoken communication (Burger et al., 2006). In other words, parents find choosing whether to go ahead with cochlear implantation more stressful when their child has more hearing to lose. Some children eligible for the CUSP survey had much better hearing than those studied by Burger et al., (2006) and it was not known whether CUSP study HA group parents would be willing to consider CIs, regardless of the study findings. They survey therefore asked: *"Are you satisfied that using [HAs/CIs] is in the best interests of your child at the moment?"* with response options: *'very satisfied', 'a little satisfied', 'a little dissatisfied'* and *'very dissatisfied'*. This question was not intended to accurately measure the acceptability of CIs to parents of children with HAs, but as an indicator of any difference in levels of satisfaction between groups that would warrant further investigation.

One important measure of the success of a hearing intervention is its effective use. Better listening and language development is associated with longer duration use per day (Park, Gagnon, et al., 2019; Wiseman et al., 2021). Documenting this was therefore important, both as a measure of children's device acceptance and as a potentially confounding variable in comparisons of listening outcomes. Sufficient daily device use in children has been variably defined as >8 hours per day (Easwar et al., 2018; Marnane & Ching, 2015), >10 hours per day (Tomblin et al., 2015), >75% of the day (Ching & Hill, 2007), and >80% of the child's waking hours (Park, Gagnon, et al., 2019). A question on device use is included within the PEACH, where parents select

whether the child has worn their device ‘Never’, ‘Seldom’, ‘Sometimes’, ‘Often’, or ‘Always’, defined as proportions of the day, namely 0%, 1-25%, 26-50%, 51-75% and 76-100% respectively. This was used to measure daily device use for the survey as it comes from a validated tool and included a category in line with the general consensus on acceptable daily use. Despite its importance, many children do not use their hearing devices for most of their waking hours (Wiseman et al., 2021; Wiseman & Warner-Czyz, 2018) and establishing sufficient use is a challenge that caregivers frequently seek advice about from professionals (Muñoz et al., 2016). Situational factors, children’s temperament and children’s device acceptance all influence daily device use (Moeller et al., 2009). So, to provide a more nuanced understanding of children’s acceptance of either HAs or CIs than daily device use alone, the survey asked: “How easy is it for you to get your child to use their left/right HA/CI?” with response options *‘very easy’, ‘quite easy’, ‘quite difficult’* and *‘very difficult’*. This question was approved by the PPI group.

5.2.7. Survey distribution and recruitment

Survey distribution began in February 2020. Initially, recruitment packs were printed for all eligible families, including those who were identified as needing any interpreter(s) for audiology appointments. Six surveys had been returned before the onset of COVID-19 pandemic restrictions, and the remainder were gathered during the pandemic, following a period when recruitment was halted. Once permission was granted to recommence recruitment, surveys could only be distributed in written English via the postal service. So, from the

onset of the pandemic, families who needed a spoken language interpreter were not approached.

Figure 5.1 shows the time-course of survey distribution. Coloured cells indicate that recruitment packs were posted to eligible families from a site during that month, blank cells indicate that no recruitment packs were sent. Before the end of the 3-year funded PhD period at the end of March 2021, every eligible child had a pack created and sent to hospital sites for distribution to families.

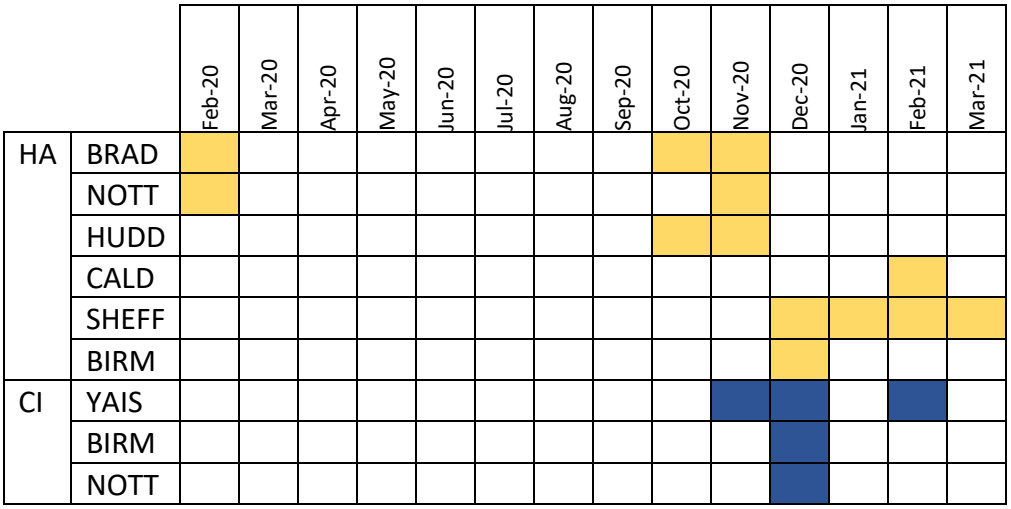


Figure 5.1: Time-course of survey distribution by site

BRAD = Bradford; NOTT = Nottingham; HUDD = Huddersfield; CALD = Calderdale; SHEFF = Sheffield; BIRM = Midlands Auditory Implant Service; YAIS = Yorkshire Auditory Implant Service

Figure 5.2 is a flow diagram illustrating the numbers of children screened, approached, and recruited from each site. Table 5.1 shows the numbers and proportions of families approached and recruited by recruitment site and Table 5.2 shows the number and proportions of participants returning their surveys online or on paper, by device use group.

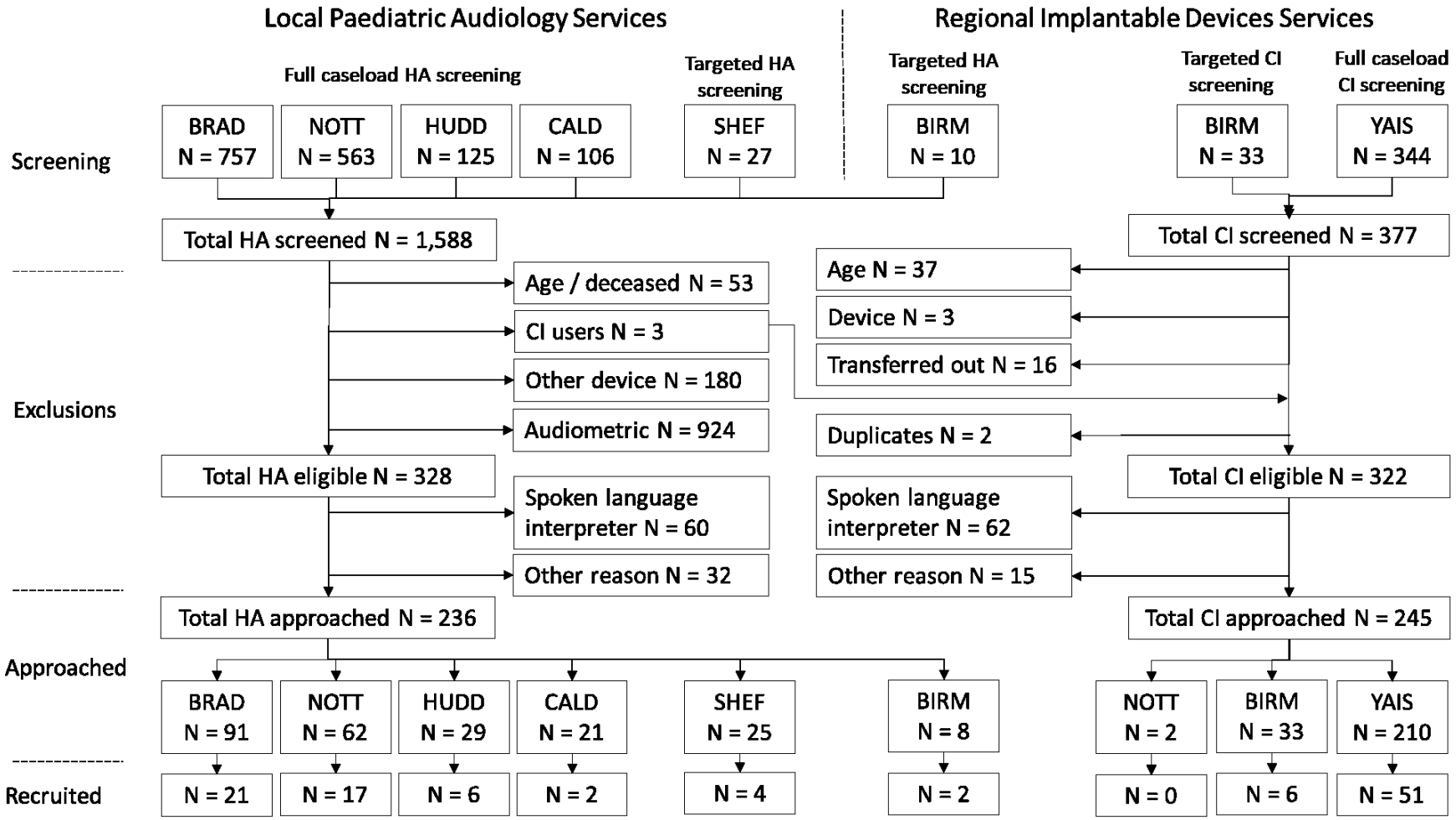


Figure 5.2: Flow diagram of survey participant identification and recruitment
BIRM = Birmingham; BRAD = Bradford; CALD = Calderdale; HUDD = Huddersfield; NOTT = Nottingham; SHEF = Sheffield; YAIS = Yorkshire Auditory Implant Service; HA = Hearing aid; CI = Cochlear implant;

Table 5.1 Numbers and proportions of families approached and recruited by site

Group	Site	Approached N	Participated N	Participated %
HA	BRAD	91	21	23
	NOTT	62	17	27
	HUDD	29	6	21
	CALD	21	2	10
	SHEF	25	4	16
	BIRM	8	2	25
	Total HA	**236**	**52**	**22**
CI	NOTT	2	0	—
	BIRM	33	6	18
	YAIS	210	51	24
	Total CI	**245**	**57**	**23**

BRAD = Bradford; NOTT = Nottingham; HUDD = Huddersfield; CALD = Calderdale; SHEF = Sheffield, BIRM = Birmingham; YAIS = Yorkshire Auditory Implant Service; CI = Cochlear implant; HA = Hearing aid.

The numbers of participants approached was based on the report of audiologists at local sites and could not be verified. Table 5.1 shows that participation rates between groups were similar overall, at 22% for families of children with HAs, and 23% for families of children with CIs.

Table 5.2: Survey completion method by device use group

Survey completion method	HA group (N = 52)	CI group (N = 57)
Paper	37 (71%)	39 (68%)
Online	15 (29%)	18 (32%)

HA = Hearing aid; CI = Cochlear implant

Table 5.2 shows that both the online and paper options were used for both groups in similar proportions, with the paper survey being used more frequently than the online version.

5.2.8. Statistical methods

This section describes the methods used to analyse selection biases, audiometric data, listening, tinnitus, need for recovery from listening, and satisfaction data. Analyses were performed using IBM SPSS Statistics version 28.0.1.1 (15).

5.2.8.1: Analyses of audiological data

For children using HAs, the unaided HTLs from their eligibility screening forms were analysed as previously described in Chapter 4, i.e., 4-frequency average HTLs of the better-hearing ear were calculated based on the two most-recent audiograms, using a value of 130 dB HL for HTLs beyond the upper limit of the audiometer. The 4-frequency average HTLs were used to classify children into the BSA's hearing loss categories. HTLs along with relevant additional comments were reviewed to categorise children into one of four NICE eligibility categories previously described.

5.2.8.2: Analysis of the Parents Evaluation of Aural/Oral Performance of Children scale

The PEACH comprises 13 items, 11 of which were used to calculate sub-scores of performance in quiet and background noise (Ching & Hill, 2007). The full PEACH questionnaire is shown in Appendix C. Questions from this were used in the CUSP survey. In line with the author's recommendations (Ching & Hill, 2007), the CUSP study survey asked caregivers to rate their children for each item as to whether they exhibited the described behaviour

0%, 1-25%, 26-50%, 51-75%, or 76-100% of the time, also defined as *'Never', 'Seldom', 'Sometimes', 'Often'* and *'Always'* respectively. Responses were assigned one of five integer values, from 0 for *'Never'* to 4 for *'Always'*. The only exception was item 2, where scoring was reversed to reflect the fact that experiencing discomfort to loud sounds is an adverse rather than advantageous outcome. Items 1 and 2, regarding device use and loudness discomfort, were intended to aid children's audiological management only (Ching & Hill, 2007). The remaining items were used to derive sub-scores for performance in quiet and background noise. However, due to a transcription error, one item from the noise sub-scale was omitted from both the paper and online versions of the CUSP survey. This meant that results would not be directly comparable to other PEACH data published, but that HA and CI group comparisons could be made, as the questions were presented to both groups in the same manner.

In the PEACH (2007), percentage scores were calculated for the quiet and noise subscales independently, then averaged to produce the final global score. This gave equal weighting to the quiet and noise sub-scales although the quiet sub-scale included one more item than the noise sub-scale. However, free-text feedback volunteered by CUSP survey participants indicated that item 12 on telephone use was difficult to answer. Questions on telephone use have become problematic in recent years because telephone technology has evolved. A telephone question was found to have poor validity in another caregiver-reported functional listening outcome tool (the Speech, Spatial and Qualities of hearing Questionnaire Parent version) when completed by caregivers independently (Killan et al., 2020). CUSP survey respondent felt the need to volunteer qualifications to their answers, or to choose more than one response

option, because their child's ability varied depending upon factors such as whether the call was audio-alone or audio-visual, whether children were listening via telephone handsets alone or using loudspeaker functions, etc. This suggested that item 12 was ambiguous and so it was excluded from calculations of the quiet sub-score for the present analysis. The item omitted in error from the CUSP survey was item 9, regarding the frequency with which a child initiated or participated in conversation in noisy environments. It was therefore not possible to include this item in the noise sub-scale score.

Table 5.3 gives descriptions of the behaviours addressed by each item. It also shows which items were used in the original PEACH to derive quiet and noise sub-scores and which were used to calculate quiet and noise sub-scores in the present study.

Table 5.3: PEACH items included in sub-scale scores for the original tool and CUSP study

Item	Aural/Oral behaviour description	PEACH Quiet	CUSP Quiet	PEACH Noise	CUSP Noise
1	Device use				
2	Loudness discomfort				
3	Respond to name in quiet	√	√		
4	Follow verbal instructions in quiet	√	√		
5	Respond to name in noise			√	√
6	Follow verbal instructions in noise			√	√
7	Follow story read aloud	√	√		
8	Initiate / participate in conversation in quiet	√	√		
9	Initiate / participate in conversation in noise			√	Omitted in error
10	Understand conversation in transport			√	√
11	Recognise voices	√	√		
12	Successfully use telephone	√	Asked but excluded from analysis		
13	Respond to sounds in environment			√	√

CUSP = Comparative oUtcomes for Severely and Profoundly deaf children study; PEACH = Parents Evalution of Aural/Oral Performance of Children scale

For the present study, the following formulae were used to derive scores:

Quiet sub-scale percentage score =
((Sum of scores for items 3, 4, 7, 8 and 11)/20) × 100
Noise sub-scale percentage score =
((Sum of scores for items 5, 6, 10 and 13)/16) × 100
Overall score =
(Noise sub-scale percentage score + Quiet sub-scale percentage score)/2

This method preserved an equal weighting for quiet and noise sub-scores, while not including the telephone question or the conversation in noise item that was omitted in error. This resulted in the PEACH not being used in an identical manner to that used in its validation or for its normative data. However, it was administered and analysed consistently to all parents of children in both groups, thus provided comparable scores for both the HA and CI groups within this study.

Missing responses to individual PEACH items were imputed using hot-deck imputation, as there was a risk that data were not missing completely at random. Hot-deck imputation allowed every participant to be included in analyses, reducing the risk of bias that could have been introduced by case-wise deletion or other imputation methods (Bennett, 2001). The following paragraph describes the method used to choose the values to impute.

Within cases, PEACH items are highly inter-correlated (Ching & Hill, 2007). Therefore, for each child with a missing item, the other children with full PEACH datasets who scored identically on the item most strongly correlated with the missing item were identified. Of these, those who were in the same device group, of the same gender, and who matched for the presence or absence of additional needs were chosen to form a matched group. The median

score this group were given for the missing item was imputed for the child with the missing item response (Bennett, 2001).

PEACH scores were compared between CI users and HA users in each of the NICE audiometric eligibility groups using multivariable linear regression modelling. As outlined in section 5.2.5, there were several variables that could plausibly be associated with children's PEACH scores, based on the literature. These needed to be controlled for in a comparison of children with HAs and CIs. However, with 109 participants this number of predictor variables was greater than is advised (Field, 2000). Therefore, an exploratory multiple regression model was created that included every potential predictor variable so that they could be examined to test for inter-correlation and significant associations with PEACH scores. A second, final model was then created, using only those variables with significant associations (Field, 2000). This section will describe the use of each potential predictor variable used in the exploratory model.

Device type:

Children were categorised into one of five groups. The first included all children using CIs. Children using HAs were classified into the four NICE eligibility groups: those known to be not eligible for CIs (outside TAG566); those known to be newly eligible for CIs (within TAG566 but outside TAG166); those whose eligibility for CIs was unknown; and those who had been audiometrically-eligible for CIs prior to the NICE guidance change, (those within TAG166). Dividing the children this way allowed a comparison of CIs versus HAs for children with different degrees of hearing loss, with

hearing loss categorised in a way that was most relevant to current NHS CI candidacy guidance, in line with the aim of the thesis.

Daily device use:

The PEACH item concerning device use asked caregivers to report use as a proportion of children's waking hours, and had no missing responses. As only a small number of children were reported to wear their devices for less than the maximum daily use category these data were used dichotomously, with children classified as either using their device(s) for 75% to 100% of their waking hours, or for less than this.

Additional needs:

Children's additional needs were based upon caregiver report within the survey, not on eligibility screening data. Children were classified into one of three groups, based upon the recommendations of Cupples, Ching, Leigh, et al., (2018) who found an association between language development and type of additional need in children with hearing loss. The first group had no additional needs, the second had additional needs described as 'Group B' by Cupples, Ching, Leigh, et al., (2018) including syndromes, vision, speech, and medical conditions but no conditions or syndromes entailing developmental delay. The last group had additional needs described as 'Group A' (Cupples, Ching, Leigh, et al., 2018). This included all children with developmental delay or autism who may or may not also have had 'Group B' 'conditions.

Responder-caregiver highest educational qualification:

Children were grouped dichotomously as having caregivers with degree-level education or higher, or highest educational qualification lower than degree level.

Sex:

PEACH scores were reported to be independent of whether children were male or female in a previous study (Bagatto & Scollie, 2013), however I included the variable within the exploratory model to ensure that it did not have a significant association with PEACH score for this group of children. Children were classified as being male or female. There were no differences between parent reports and sex assigned at birth recorded during eligibility screening.

Age of child at survey completion:

PEACH scores from children without hearing loss indicate that some improvement is seen in scores over around the first 36 months of life and normative data was available only for children up to age 50 months (Ching & Hill, 2007). As the CUSP group included children with hearing loss who might have delayed oral/aural development and older children and teenagers, age in completed years was used as a continuous variable in the exploratory model.

Receptive communication mode:

Due to the small numbers of children using only BSL as their receptive communication mode, children were classified dichotomously as using either only spoken language, or any form of receptive sign or gesture, including spoken language plus signed support, spoken language plus Makaton, or BSL only.

Main spoken language in the home:

Children were dichotomously classified based on caregiver reports as coming from either homes where English was the main spoken language, or from homes where a language other than English was the main spoken language.

Last, the quality of the final model was assessed by testing for the assumptions of independent errors, the distribution of standardised residuals, whether any cases exerted undue influence, ensuring non-zero variance, no multicollinearity of predictor variables (via tolerance and variance inflation factor values), homoscedasticity, normally distributed errors, and the assumption of independence (Field, 2000).

5.2.8.3: Analysis of the tinnitus questions

The children from HA and CI groups who were rated unable to tell their caregivers about tinnitus were described by age, sex, and additional needs, and the proportions of children from each group in this category were described in percentages. Excluding children unable to tell their caregivers about tinnitus, and those with missing data, the proportions of children who had reported tinnitus versus those who had not reported tinnitus were compared across the HA and CI groups using binary logistic regression modelling, controlling for age, sex, and the presence or absence of any additional needs. Age and additional needs were included as these can influence children's ability to explain symptoms they experience. For children who had reported tinnitus, the proportions from each device group who had reported it to affect their sleep and to be annoying were described in percentages and compared via Pearson's Chi-Squared tests where the data met the assumptions for this test, excluding children who had reported the presence of tinnitus but whose caregivers did not know if their child found it annoying or sleep-disturbing.

5.2.8.4: Analysis of the need for recovery from listening questions

The proportions of the HA and CI groups giving each different response option regarding wanting a break from both left and right devices, frequency of headaches, and frequency of tiredness from listening, were analysed via descriptive statistics. Children were then categorised into dichotomous groups based on whether they experienced tiredness or headaches either *"sometimes" / "frequently"*, or *"never" / "hardly ever"*. For group comparisons of the frequency of needing a break, children were classified by the ear that needed a break least frequently, or the only ear that was aided for children with one device. This was to gauge the frequency of children needing a break from listening via any devices at all and to avoid confusing needing a break from listening altogether with needing a break from a less-preferred device, such as a second, sequential CI. The proportions of children in these groups were compared between the HA and CI users via binary logistic regression modelling, controlling for school stage, school receptive language communication mode, sex, and the presence or absence of additional needs. HA users were classified as either being eligible for CIs (In TAG566) or not (outside or could not be classified via TAG566).

5.2.8.5: Analysis of child acceptance and caregiver satisfaction with hearing devices questions

The distributions of responses to questions regarding caregiver device satisfaction, ease of use and daily device use were compared via two-tailed Mann-Whitney U tests. Children's characteristics including NICE eligibility status, outcomes of any prior CI eligibility assessment referrals, whether

caregivers had ever thought about having a CI eligibility assessment for their child and the caregivers' own hearing and device use were described narratively in relation to parent satisfaction groups.

5.2.8.6: Characteristics of survey participants and eligible population

The characteristics of the survey respondents to those of all families screened who met the study eligibility criteria could be documented based on data from the eligibility screening forms. This included data presented in Chapter 4 for the HA users at Bradford, Huddersfield, Calderdale and Nottingham. It also included children identified by targeted eligibility screening of children with severe or profound hearing loss at Sheffield Children's Hospital, the caseload of CI users implanted YAIS, and the children either using CIs or using HAs and undergoing eligibility assessment for CIs, at the Midlands Hearing Implant Programme in Birmingham. Survey respondents were compared to the wider eligible population via descriptive statistics, in terms of the prevalence of additional needs and family need for interpreters.

5.3 RESULTS

5.3.1. Participant characteristics

Table 5.4 shows the characteristics of the children represented in the survey. The HA and CI groups were similar in many regards. However, the HA group included a larger proportion of infants, while the CI group included a larger

proportion of Key Stage 4 students. Greater proportions of the HA group attended special schools and had additional needs. As a result, age and the presence of additional needs were controlled for in comparisons of HA and CI group outcomes.

Table 5.4: Survey respondents' children's characteristics

	HA Group N = 52	CI Group N = 57
Gender		
Female	23 (46%)	28 (49%)
Male	28 (54%)	29 (51%)
Age in years		
Median (range)	9 (2 to 17)	10 (3 to 18)
Stage of education		
Infant / Pre-school (0 to 3 years old)	7 (13%)	0
Early Years (3 to 4 years old)	2 (4%)	3 (5%)
Early Years Reception (4 to 5 years old)	1 (2%)	2 (4%)
Key Stage 1 (5 to 7 years old, school years 1 & 2)	10 (19%)	11 (19%)
Key Stage 2 (7 to 11 years old, school years 3, 4, 5 & 6)	13 (25%)	16 (28%)
Key Stage 3 (11 to 14 years old, school years 7, 8 & 9)	9 (17%)	8 (14%)
Key Stage 4 (14 to 16 years old, school years 10 & 11)	6 (12%)	12 (21%)
A-levels at a school sixth-form (16 to 18 years old, school years 12 & 13)	2 (4%)	2 (4%)
Special School Post-16 (16 to 18 years old, school years 12 & 13)	1 (2%)	—
A-levels at a college (16 to 18 years old)	—	1 (2%)
Further Education (studying for a college course)	1 (2%)	2 (4%)
Higher Education (studying for a degree at college or university)	—	—
Training / Apprenticeship	—	—
In employment	—	—
Not in education, employment or training	—	—
Educational Setting		
College (Further education or A-levels)	1 (2%)	3 (5%)
Mainstream school, state-funded	21 (40%)	38 (67%)
Mainstream school, independent (private school, not state-funded)	3 (5%)	5 (9%)
Resource provision in a mainstream school	3 (5%)	8 (14%)
Special school for deaf pupils	5 (10%)	0 (0%)
Other special school, not specifically for deaf children	2 (4%)	1 (2%)
School aged and supported only at home i.e. home-schooled	4 (8%)	0 (0%)
Pre-school carer, e.g. nursery, nanny, or other day-care	5 (10%)	1 (2%)
Pre-school, at home	4 (8%)	1 (2%)
Missing	4 (8%)	0 (0%)

As discussed in Chapter 4, there were limitations in the method used to assess the presence of additional needs during eligibility screening. However, within the survey parents were asked to specify all additional needs their child had, providing a richer, more accurate picture of the children's characteristics. These are summarised in Table 5.5 along with other demographic data. The HA group included a greater proportion of children with speech, language, communication needs and autism, while the CI group had a larger proportion of children with visual impairment.

Table 5.5: Additional needs by device group

	HA Group N=52	CI Group N=57
Incidence of any additional needs		
No additional needs apart from hearing loss	23 (44%)	35 (61%)
Learning difficulty	9 (17%)	11 (19%)
Behaviour, emotional or social	8 (15%)	9 (16%)
Speech, language and communication	16 (31%)	12 (21%)
Autism spectrum disorder	5 (10%)	1 (2%)
Vision impairment	2 (4%)	5 (9%)
Physical disability	3 (6%)	2 (4%)
Other	10 (19%)	7 (12%)
Additional needs data missing	1 (2%)	—
Cumulative incidence of additional needs		
No additional needs apart from hearing loss	23 (44%)	35 (61%)
One extra additional need	10 (19%)	6 (11%)
Two extra types of additional needs	9 (17%)	7 (12%)
Three extra types of additional needs	7 (13%)	7 (12%)
Four extra types of additional needs	2 (4%)	2 (4%)
Additional needs data missing	1 (2%)	—

Additional needs specified under the 'Other' category that could not be classified into one of the pre-existing options included inflammatory bowel disease, global developmental delay, balance difficulties, attention

hyperactivity deficit disorder, Tourette's syndrome, sensory processing disorder, dyspraxia, hypermobility, dysexecutive dysfunction, and epilepsy. The number and proportions of children with different causes of hearing loss based on caregiver reports are shown in Table 5.6.

Table 5.6: Causes of children's hearing loss by device group

	HA Group N=52	CI Group N=57
Auditory neuropathy spectrum disorder	3 (6%)	9 (16%)
Cytomegalovirus	1 (2%)	4 (7%)
Other virus in utero	—	2 (4%)
Known genetic	5 (10%)	8 (14%)
Unknown genetic	10 (19%)	2 (4%)
Meningitis	1 (2%)	4 (7%)
Wide vestibular aqueduct syndrome	3 (6%)	3 (5%)
Other syndrome	2 (2%)	4 (7%)
Hospital says unknown	9 (17%)	17 (30%)
Hospital told me but I have forgotten	5 (10%)	2 (4%)
Prematurity	2 (4%)	2 (4%)
Other cause	3 (6%)	—
Missing	4 (8%)	—

Table 5.7 shows the characteristics of the responder-caregivers and other main caregivers by device group, including their relationships to the child, self-reported hearing status, their own hearing device use and highest level of education. No participating families had two mothers or two fathers. Overall, most surveys were completed by mothers, but a greater proportion were completed by fathers for the CI group than the HA group. Both groups had similar proportions of caregiver-responders regarding hearing status and hearing device use, but a greater proportion of the other main caregivers had hearing loss and used hearing devices within the HA group than the CI group.

Caregiver-responders from the CI group were the most likely to have an undergraduate degree or higher academic qualification, and a higher proportion of caregivers from the HA group did not wish to disclose their highest level of qualification. Compared to the CI group, a higher proportion of responder-caregivers from the HA group (all mothers) did not consider there to be another main caregiver for their child.

Table 5.7: Caregiver-responder and other main caregiver characteristics

Characteristic		Caregiver-Responder		Other main caregiver	
		HA group N = 52	CI group N = 57	HA Group N = 34	CI Group N=49
Relationship to child	Mum	51 (98%)	46 (81%)	1 (2%)	10 (18%)
	Dad	1 (2%)	10 (18%)	29 (56%)	37 (65%)
	Step-Dad	—	—	2 (4%)	—
	Legal Guardian	—	1 (2%)	—	1 (2%)
	Grandparent	—	—	2 (4%)	1 (2%)
Self-reported hearing	Good hearing	48 (92%)	55 (96%)	29 (85%)	47 (96%)
	Hard of hearing	3 (6%)	2 (4%)	2 (6%)	2 (4%)
	Deaf	1 (2%)	—	3 (9%)	—
Hearing device use	None	47 (90%)	57 (100%)	30 (88%)	48 (98%)
	Hearing aid	5 (10%)	—	3 (9%)	1 (2%)
	Cochlear implant	—	—	1 (3%)	—
Highest level of education	School	9 (17%)	9 (16%)	10 (29%)	11 (22%)
	Further Education	23 (44%)	18 (32%)	9 (26%)	23 (47%)
	Higher Education	16 (31%)	29 (51%)	11 (32%)	14 (29%)
	Unspecified foreign qualification	—	—	1 (3%)	—
	Prefer not to say	4 (8%)	1 (2%)	3 (9%)	1 (2%)
Other main caregiver	Child has another main caregiver	34 (65%)	49 (86%)	—	—
	No other main caregiver	18 (35%)	8 (14%)	—	—

CI = cochlear implant; HA = hearing aid; Further education = A-levels, college course, foundation degree or professional qualification; Higher education = undergraduate degree or higher academic qualification e.g. masters degree or PhD.

Table 5.8 showed that 94% of the HA respondents, and 100% of the CI respondents, were from families who did not require spoken language interpretation for audiology appointments. In the survey, parents were asked to report the main spoken language used in the home. This revealed a greater difference between the groups, with 15% of the HA families mainly using a spoken language other than English, in comparison to only one out of the fifty-seven families in the CI group. The interpreter categories for each group are shown in Table 5.8.

Table 5.8: Main spoken language used in children's homes by device group

Main spoken language used in home	HA Group	CI Group
English	44 (85%)	56 (98%)
Eastern European	—	1 (2%)
Other European	1 (2%)	—
Arabic	3 (6%)	—
South East Asian	4 (8%)	—
No spoken language, BSL only	—	—

Table 5.9 documents children's receptive and expressive communication modes at home and the communication mode used by their carers, teachers, or colleagues at home, day-care, pre-school, school, or college. Sign-supported English and spoken language together with Makaton were classified as 'spoken language together with signed support'. Two parents of children with CIs volunteered that their child used spoken language only during the day while using their hearing devices and British Sign Language or Sign-Supported English at bedtime when they had taken their speech processors off. They were classified as using spoken language together with signed support. If parents

volunteered that children used lip-reading to aid their understanding of speech, with no mention of any use of sign or gesture, communication mode was classified as 'spoken language only'.

Table 5.9: Children's receptive and expressive communication modes by device group.

	HA Group N=52	CI Group N=57
"How do you communicate with your child at home?"		
Spoken language only	35 (67%)	35 (61%)
Spoken language together with signed support	12 (23%)	22 (39%)
Spoken language together with pictures	1 (2%)	—
British Sign Language only	3 (6%)	—
Missing	1 (2%)	—
"How does your child communicate with you at home?"		
Spoken language only	40 (77%)	41 (72%)
Spoken language together with signed support	7 (13%)	13 (23%)
British Sign Language only	3 (6%)	1 (2%)
British Sign Language, gestures, and Makaton	—	1 (2%)
Vocalisations, body language, and facial expressions	2 (4%)	—
Missing	—	—
"How do your child's carers / teachers / colleagues communicate with your child?"		
Spoken language only	36 (69%)	44 (77%)
Spoken language together with signed support	8 (15%)	11 (19%)
British Sign Language only	3 (6%)	2 (4%)
Not applicable, pre-school aged and no carer outside the family	2 (4%)	—
Missing	3 (6%)	—

HA = Hearing aid; CI = Cochlear implant

Parents were asked to specify their child's age at first confirmation of hearing loss, first HA fitting, and first CI fitting (for the CI group) in years and months. Many parents could not recall the ages this accurately, volunteering only the

age in completed years rather than completed years and months. Therefore, all age at diagnosis and intervention data were analysed in completed years of life only. Mann-Whitney U tests were applied to compare ages at diagnosis and first HA fitting between the groups. This is because the data were non-parametric, skewed toward younger ages, and with a highly significant test statistic on the Kolmogorov-Smirnov test ($p<0.01$ in both cases). Mann-Whitney U tests indicated that the HA group were significantly older at first detection of hearing loss than the CI group and significantly older at first HA fitting ($p<0.05$ in both cases).

Children's audiological characteristics are summarised by device group in Table 5.10. Half of the participants in the HA group had HTLs outside the 2019 NICE guidance and the other half were distributed between the other three NICE eligibility groups.

Table 5.10: Children's audiological characteristics

	HA Group N = 52	CI Group N = 57
NICE CI audiometric eligibility group		
Outside TAG566 (2019)	26 (50%)	—
Outside TAG166 (2009) and within TAG566 (2019)	11 (21%)	—
Outside TAG166 (2009), could not determine if within TAG566 (2019)	9 (17%)	—
Within TAG166 (2009)	6 (12%)	—
Better ear unaided hearing thresholds: 4 frequency average		
Mean (standard deviation) (dB HL)	68 (18.09)	—
Range (dB HL)	30 to 125	—
BSA hearing loss classifications (better-hearing ear)		
Mild (21 – 40 dB HL)	2 (4%)	—
Moderate (41 – 70 dB HL)	30 (58%)	—
Severe (71 – 95 dB HL)	16 (31%)	—
Profound (>95 dB HL)	4 (8%)	—
Device fitting		
Unilateral HA	1 (2%)	—
Bilateral HA	51 (98%)	—
Bimodal (HA + CI)	—	—
Bilateral CI	—	57 (100%)
Ages at diagnosis and intervention		
Median age at first confirmation of any hearing loss in years (range)	<1 (0 to 10)	<1 (0 to 2)
N children with missing data re confirmation of hearing loss age	3 (6%)	—
Median age at first HA fitting in years (range)	1 (0 to 10)	<1 (0 to 3)
N children with missing data re first HA fitting	5 (10%)	5 (9%)
Median age at first CI fitting in years (range)	—	1 (0 to 9)
N children with missing data re age at first CI	—	1

CI = Cochlear implant; HA = Hearing aid; NICE = National Institute for Health and Care Excellence; TAG = Technology Appraisal Guidance; BSA = British Society of Audiology

Tables 5.4 to 5.10 provide detailed characterisation of the HA and CI groups. The level of detail resulted in small numbers of individuals falling into many categories and so, apart from age at identification and intervention, comparisons were made descriptively rather than statistically.

In summary, HA and CI group participants were broadly similar in terms of children's gender and age range; the proportion using spoken only versus any signed communication; bilateral HA or CI use; and responder-caregiver hearing status and hearing device use. Differences between the groups included more children using HAs being under 4 years of age and more children with CIs being aged 14 to 16; and a smaller proportion of children with CIs having additional needs. Greater proportions of HA group responder caregivers were female, did not have another main caregiver, and had lower highest educational qualification or with-held information about their education.

Table 5.11 summarises key characteristics for the CI group and the HA users categorised by NICE eligibility group. Respondents who selected "prefer not to say" regarding their highest level of educational qualification were classed as having below degree-level education for this table and subsequent analyses.

Table 5.11: Key characteristics by Device and NICE eligibility group

Characteristics	HA				CI
	Previously eligible	Newly eligible	Eligibility unknown	Ineligible	
Group size	N=6	N=11	N=9	N=26	N=57
Median Age (years)	13	10	7	9	11
IQR Age (years)	7.5 – 17.0	4.5 – 13.5	4.0 – 11.0	6.0 – 12.5	7.0 – 14.0
Mean 4-freq HTLs better-hearing ear (dB HL)	101	78	69	56	—
Range 4-freq HTLs better-hearing ear (dB HL)	75 - 125	70 - 90	56 - 84	30 - 70	—
Male	2 (33%)	5 (45%)	6 (67%)	13 (50%)	28 (49%)
Female	4 (67%)	6 (55%)	3 (33%)	13 (50%)	29 (51%)
Caregiver has degree-level education	3 (50%)	2 (18%)	4 (44%)	7 (27%)	29 (51%)
English is an additional language in the home	2 (33%)	—	1 (11%)	5 (19%)	1 (2%)
Child has at least one additional need	5 (83%)	3 (37%)	5 (56%)	15 (58%)	23 (40%)

HA = Hearing aid; CI = Cochlear implant; TAG = National Institute for Health and Care Excellence Technology Appraisal Guidance; IQR = Interquartile range; 4-freq HTLs = Hearing threshold levels averaged over 0.5, 1, 2, and 4 kHz.

5.3.2. Comparative caregiver-reported listening outcomes

This section presents the PEACH score results, with individual HA group data points plotted as a function of 4-frequency HTLs relative to CI group outcomes, followed by the results of the exploratory and final multivariable linear regression models.

5.3.2.1. Individual and device group scores

Overall the HA users had a mean total PEACH score of 61.6% (standard deviation = 22.81) and the CI group mean was 77.9% (standard deviation = 22.81). Table 5.12 shows the results of the CI group and HA users grouped by NICE CI eligibility.

Table 5.12: PEACH scores of the CI users and HA users by NICE cochlear implant eligibility group.

Group	Total PEACH score	
	Mean	Standard deviation
CI (N=57)	77.9	13.95
Ineligible (N=26)	69.7	18.90
Eligibility unknown (N=9)	55.5	18.62
Newly eligible (N=11)	64.3	16.90
Previously eligible (N=6)	30.5	28.49

TAG = National Institute for Health and Care Excellence Technology Appraisal Guidance; PEACH = Parents Evaluation of Aural/Oral Performance of Children scale

Individual data points for the HA group are plotted as a function of 4-frequency average HTLs of the better-hearing ear in Figure 5.3 below, alongside dashed lines indicating the distribution of the CI group's scores. Children's PEACH scores varied from 100% to 0% and a wide range of scores can be seen within each group. Table 5.11, showed the difference in 4-frequency average HTLs between the HA groups. Those falling outside TAG566 (2019) had the lowest HTLs, followed by those who could not be classified via TAG566 (2019), then those who were newly eligible for CIs following the guidance change (within TAG566 (2019) but outside TAG166 (2009)). Unsurprisingly, those who fell within TAG166 (2009) had the highest 4-frequency HTLs on average. A trend can be seen for PEACH scores to decrease with increasing 4-frequency average HTL. However there is a good deal of scatter in the data. Given the differences in group characteristics previously noted in Table 5.11, this underscored the importance of controlling for other plausibly influential variables. The following sections will address this by presenting the outcomes of multivariable linear regression modelling.

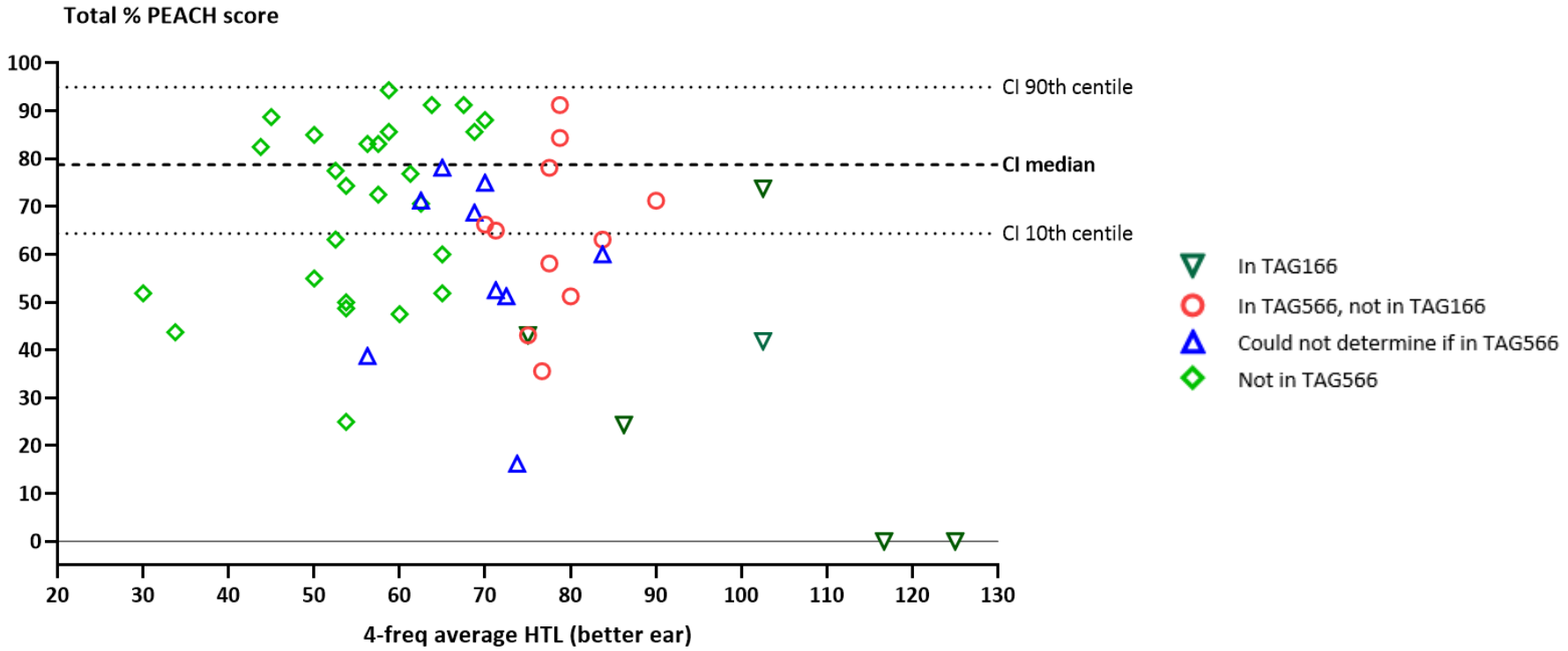


Figure 5.3: PEACH scores as a function of four-frequency unaided HTLs of the better-hearing ear.

HA users' individual scores are plotted as symbols. Dashed horizontal lines show the median, 10th and 90th centile scores of the CI group.

5.3.2.2. Exploratory multiple linear regression model

The outcomes of the exploratory linear regression model are shown in Table 5.13. The unstandardised beta coefficients represent the change in the PEACH score associated with each predictor variable, relative to the reference value for that variable and controlling for the others. For example, compared to being a CI user, being a HA user from the group who had been audiometrically eligible for CIs under the original NICE guidance TAG166 (2009) was associated with having a worse PEACH score by 38%. There was an acceptably low level of multicollinearity between all the potential predictor variables, with variance inflation factor values below 10 and tolerance values were above 0.1 (Field, 2000).

There were significant associations between PEACH score and device group, additional needs group, and responder-caregiver educational attainment. None of the other potential predictor variables were significantly associated with the PEACH score for this population. The exploratory model included 12 beta coefficients. Field (2000) advises that a multiple linear regression model should have at least 15 participants per beta coefficient. The inclusion of all the potential predictor variables resulted in less than 10 participants per coefficient, therefore in line with Field's recommendations, a final model was created that included only the predictors with significant associations with PEACH scores.

Table 5.13: Coefficients, significance and collinearity statistics of the exploratory PEACH multiple linear regression model

Variable	Unstandardised Beta coefficients	Significance	95% Confidence interval for β		Collinearity statistics	
			Lower bound	Upper bound	Tolerance	VIF
Device group versus CI users:						
Previously eligible	**-38.195**	**0.000**	-52.596	-23.794	0.772	1.295
Newly eligible	**-12.619**	**0.018**	-23.005	-2.232	0.851	1.175
Eligibility unknown	**-17.320**	**0.003**	-28.490	-6.150	0.881	1.135
Not eligible	-1.035	0.807	-9.439	7.369	0.649	1.540
Additional needs versus none:						
Group B additional needs	**-12.167**	**0.003**	-20.066	-4.268	0.716	1.396
Group A additional needs	**-12.131**	**0.003**	-20.167	-4.095	0.751	1.331
Dichotic variables:						
Caregiver degree-level education	**7.389**	**0.027**	0.853	13.926	0.804	1.243
Male	-2.067	0.498	-8.095	3.961	0.919	1.088
Age in years	-0.001	0.998	-0.702	0.700	0.834	1.199
Spoken language only	1.328	0.748	-6.860	9.517	0.703	1.423
English main home language	6.924	0.266	-5.372	19.219	0.727	1.375
Highest daily use category	6.318	0.231	-4.095	16.731	0.847	1.181

VIF = variance inflation factor. Hypothetical reference case was: Child using CIs with no additional needs. Significance values of $p<0.05$ are shown in bold. Group A additional needs = children with any developmental delay or autism with or without other additional needs; Group B additional needs = children with any visual, speech, or medical condition not involving developmental delay (Cupples, Ching, Leigh, et al., 2018); TAG166 = National Institute for Health and Care Excellence Technology Appraisal Guidance.

5.3.2.3. Final linear regression model

To maximise the number of participants per beta coefficient, children were classified into two rather than three additional needs groups in the final model, because the exploratory model revealed that the sub-categories proposed by (Cupples, Ching, Leigh, et al., 2018) were associated with an almost identical decrease in PEACH score of 12% compared to children without additional needs. Table 5.14 shows a summary of the strength of the final model and Table 5.15 provides beta coefficients, significance values, 95% confidence intervals of beta coefficients and collinearity statistics for each predictor variable.

Table 5.14 Final PEACH regression model summary

R	R Square	Adjusted R Square	Std. error of the estimate	R Square Change	F Change	Significance of F change	Durbin-Watson
0.697	0.485	0.455	14.974	0.485	16.022	<0.01	1.931

Table 5.15: Coefficients, significance of predictor variables and collinearity statistics of the final PEACH multiple linear regression model

Variable	Unstandardised Beta coefficients	Significance	95% Confidence interval for β		Collinearity statistics	
			Lower bound	Upper bound	Tolerance	VIF
Constant	79.358	**0.000**	73.254	85.462	—	—
Device group versus CI users						
Previously eligible	**-41.951**	**0.000**	-54.974	-28.929	0.917	1.090
Newly eligible	**-12.917**	**0.013**	-22.999	-2.834	0.877	1.140
Eligibility unknown	**-18.690**	**0.001**	-29.382	-7.999	0.935	1.070
Ineligible	-3.750	0.304	-10.948	3.448	0.860	1.163
Other predictor variables						
Additional needs present	**-12.571**	**0.000**	-18.804	-6.339	0.837	1.195
Caregiver degree-level education	**7.181**	**0.026**	0.876	13.487	0.840	1.191

VIF = variance inflation factor. Hypothetical reference case was: Child using CIs with no additional needs. Significance values of $p<0.05$ are shown in bold; TAG = National Institute for Health and Care Excellence Technology Appraisal Guidance

5.3.2.4. Assessing the quality of the final model

The quality final model met the assumptions of multivariable linear regression modelling listed in the methods section (Field, 2000). Please see Appendix D for details. The model was both accurate for the sample and generalisable to the population.

5.3.2.5. Outcome of the final PEACH model

All groups of children using HAs scored significantly worse on the PEACH questionnaire than children using CIs except for children using HA who could be confirmed to fall outside current NICE audiometric guidance TAG566 (NICE, 2019). Controlling for caregiver education and the presence of any additional needs, children using HAs whose hearing thresholds met the previous NICE audiometric guidance (NICE, 2009) had scores that were poorer by an average of 42%; those newly eligible (falling outside TAG166 but within TAG566) scored lower than their peers with CIs by 13%; and children using HAs whose missing audiometric data meant their audiometric eligibility for CIs was unknown scored 19% worse on average. The presence of additional needs was associated with poorer PEACH scores by an average of 12.5% and having a responder-caregiver with a degree-level education or higher was associated with the child having a better PEACH score by 7% on average.

5.3.3. Comparative caregiver-reported tinnitus outcomes

The first tinnitus question asked: *"Has your child ever told you that they hear noises in their ears or head, that don't come from their hearing aids, cochlear implants, or surroundings (tinnitus)?"* Response options included *"Yes", "No",* and *"My child is not able to tell me*". Ten parents chose the last option, representing seven girls and three boys. One was from the CI group (2%) and the other nine used HAs (17%). The children who were not able to tell their parents about tinnitus included all but one of those aged under 4 years. Three had no additional needs; they were all aged 3 years or less. The remaining seven had additional needs and were aged 2 to 9 years (median 2 years). The children from the HA group who were unable to tell their parents about tinnitus had four-frequency average HTLs ranging from 43.75 to 125 dB HL. A further three parents did not answer this question, representing two children with CIs and one with HAs. Table 5.16 summarises the presence of tinnitus data from all caregiver-respondents.

Table 5.16: Tinnitus prevalence data from all caregivers

Question: *"Has your child ever told you that they hear noises in their ears or head, that don't come from their [HAs/CIs] or surroundings (tinnitus)?"*		
Responses:	**HA Group N = 52**	**CI Group N = 57**
"No"	34 (65%)	41 (72%)
"Yes"	8 (15%)	13 (23%)
"My child is not able to tell me"	9 (17%)	1 (2%)
Missing data	1 (2%)	2 (4%)

Excluding children with missing data, and children who were unable to tell their parents about tinnitus, eight out of 42 children with HAs (19%) and 13

out of 54 children with CIs (24%) had told their parents they had experienced tinnitus. These made up 21.9% of those with data who were able to report. Data from the 96 children with data who were able to tell their caregivers about tinnitus were analysed via binary logistic regression modelling to compare the prevalence of reported tinnitus between the HA and CI groups. The model compared the proportion of "Yes" and "No" responses between the groups, controlling for age, sex, and the presence of any additional needs. There was no significant association between the being in the HA or CI group and having reported tinnitus ($p>0.05$).

Data representing these 96 children's reports to caregivers about tinnitus annoyance and tinnitus disturbing their sleep are shown in Figures 5.4 and 5.5 respectively.

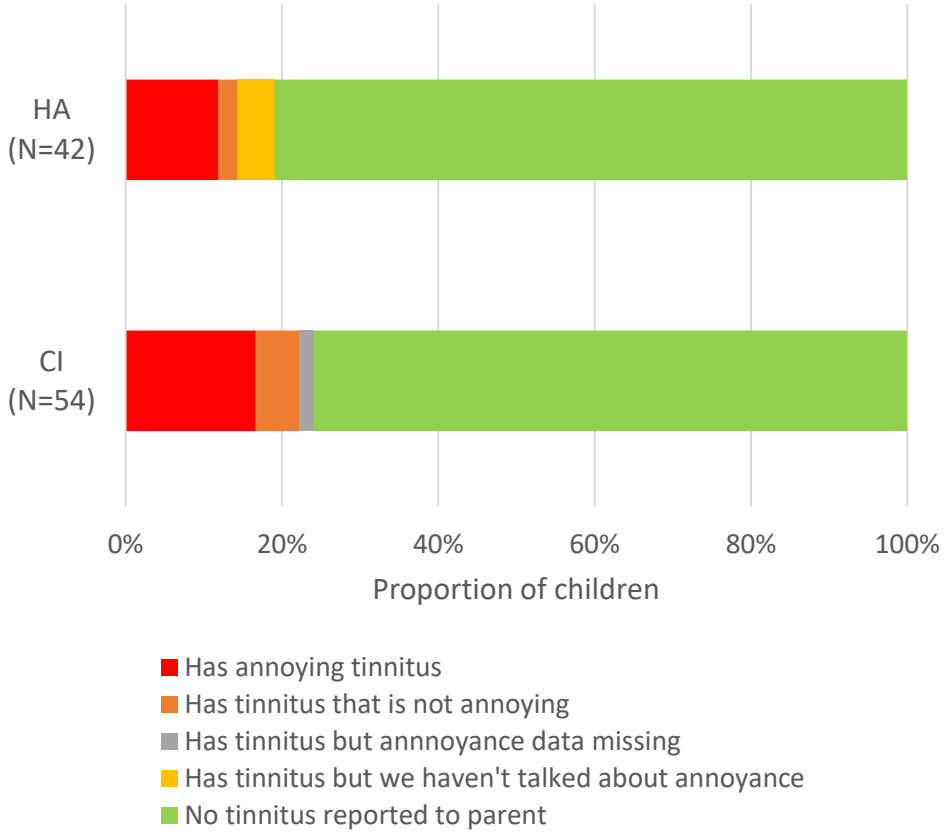


Figure 5.4: Proportion of children who had reported their tinnitus to be annoying by device group

HA = Hearing aid group; CI = Cochlear implant group

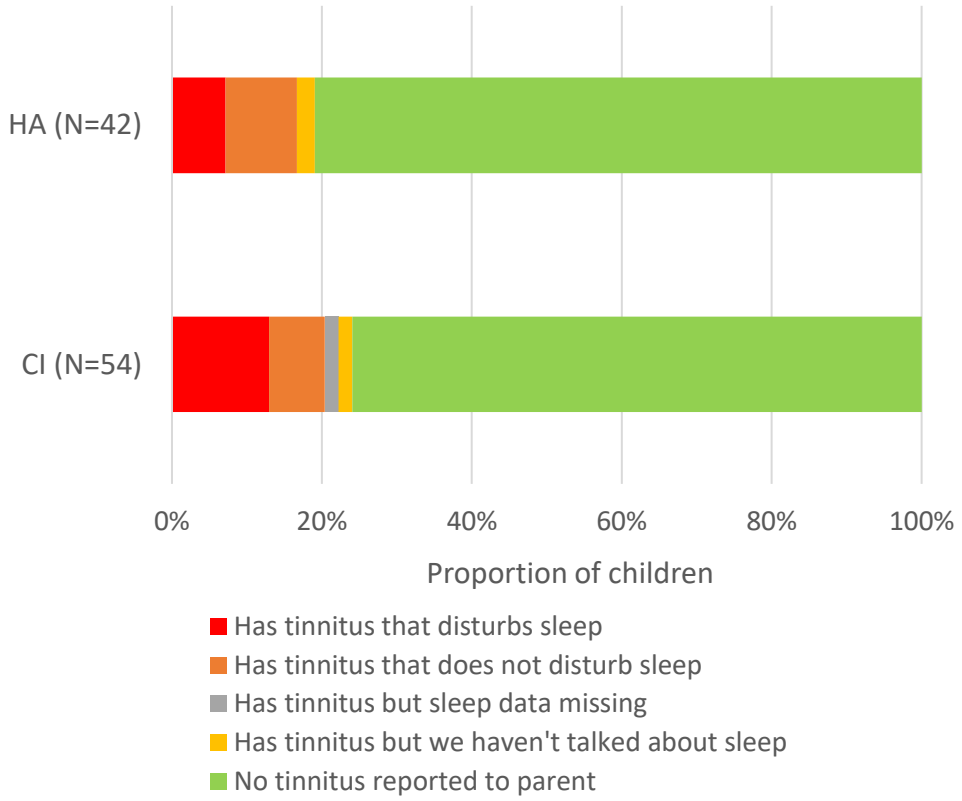


Figure 5.5: Proportion of children who had reported that tinnitus disturbed their sleep by device group

HA = Hearing aid group; CI = Cochlear implant group.

Excluding children who could not tell their parents about tinnitus, those with missing data, and those who had tinnitus but had not talked to their caregivers about whether it was annoying or disturbed their sleep, the proportions of children from the HA and CI groups who reported having experienced annoying tinnitus or tinnitus that disturbed their sleep were compared via Pearson's chi-squared tests. There was no significant difference in the proportion of children from the HA or CI groups who experienced annoying tinnitus ($p>0.05$). Too few children using HAs reported having tinnitus that disturbed their sleep to meet the assumption of a Chi-squared test (that at least five children are expected to fall within each possible category) and so it was

not possible to make a valid statistical comparison for this outcome. Similarly, it was not possible to include NICE eligibility groups in these analyses, because the numbers of children with annoying or sleep-disturbing tinnitus in the different categories were too small to meet the assumptions of the test (Field, 2000). Excluding cases where the parents responded “I don’t know, we haven’t talked about it”, of the children who had reported having tinnitus it was annoying for 83% and 75% of the HA and CI groups respectively.

5.3.4. Comparative caregiver-reported need for recovery outcomes

Responses regarding the need for recovery questions were obtained for most children. From the CI group, one parent did not respond to any of the need for recovery questions. One child from each group used only one device. Other than these, every parent of a child with CIs gave a response to the questions on how frequently their child wanted a break from right or left device use, and parents of four children using HAs said their child would not be able to tell them. These data are summarised in Table 5.17.

Table 5.17: Children's need for a break from hearing devices

Question: *"Following their usual activities, how often does your child want a break from using their device?"*				
N of children with device in that ear	**Right HA N = 51**	**Left HA N = 52**	**Right CI N = 57**	**Left CI N = 56**
Responses:				
Never wants a break	24 (47%)	24 (46%)	32 (56%)	32 (57%)
Hardly ever wants a break	11 (22%)	10 (19%)	8 (14%)	8 (14%)
Sometimes wants a break	8 (16%)	10 (19%)	14 (25%)	12 (21%)
Frequently wants a break	4 (8%)	4 (8%)	2 (4%)	2 (4%)
My child is unable to tell me	4 (8%)	4 (8%)	—	—
Missing data	—	—	—	—

HA = Hearing aid; CI = Cochlear implant

Excluding children who were not able to report to their parents regarding wanting a break from device use, 14 out of 48 HA users (29%) sometimes or frequently wanted a break from either HA, compared to 16 out of 57 CI users (28%). Figure 5.6 plots these data as cumulative bar charts.

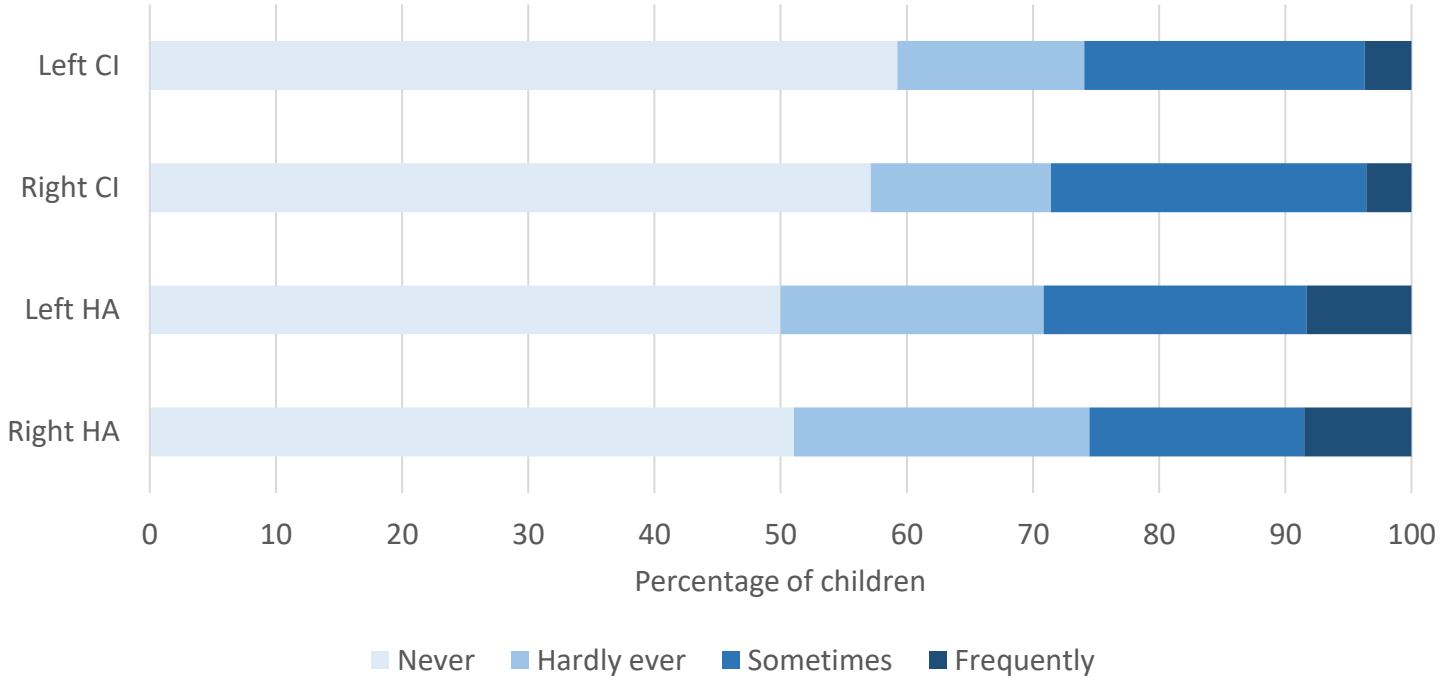


Figure 5.6: Proportions of children falling within each frequency of wanting a break category by ear and device group

HA = Hearing aid; CI = Cochlear implant; Left = Left ear; Right = Right ear

Seven children with bilateral devices were reported to have differing frequency of wanting a break from their left versus their right device. The remaining 97 children were reported as either having the same frequency of wanting a break from their devices in both ears or were only using one device.

Pearson's Chi-Square test of independence was run to see if there was a significant difference in the proportions of the CI and HA groups who needed a break from all device use either sometimes or frequently versus never or hardly ever. There was no significant difference, with 25% of both groups responding that they sometimes or frequently needed a break (X^2 (1, 97) = .00, p = 1). Binary logistic regression modelling found no significant difference between the device use groups, and no significant associations between needing a break from listening and gender, educational stage, or the presence of additional needs.

Table 5.18: Headaches related to listening by device group.

Question: *"How often does your child complain of headaches related to listening?"*		
Responses:	**HA Group N = 52**	**CI Group N = 57**
Never	26 (50%)	30 (53%)
Hardly ever	11 (21%)	8 (14%)
Sometimes	6 (12%)	14 (25%)
Frequently	1 (2%)	2 (4%)
"My child is unable to tell me"	8 (15%)	2 (4%)
Missing data	—	1 (2%)

Excluding children with missing data and those who were unable to tell their caregivers about headaches, seven out of 44 (16%) of the children with HAs had reported that they sometimes or frequently experienced headaches related to listening, compared to 16 out of 54 (30%) of the CI group. A Pearson's Chi-

Square test revealed that this difference was not statistically significant ($X^2(1, 98) = 2.54$, p=.111). Binary logistic regression modelling found no significant difference between the device use groups, and no significant associations between headaches due to listening and gender, educational stage, or the presence of additional needs. Figure 5.7 plots the proportions of responses falling within each headache frequency category, excluding children without a rating.

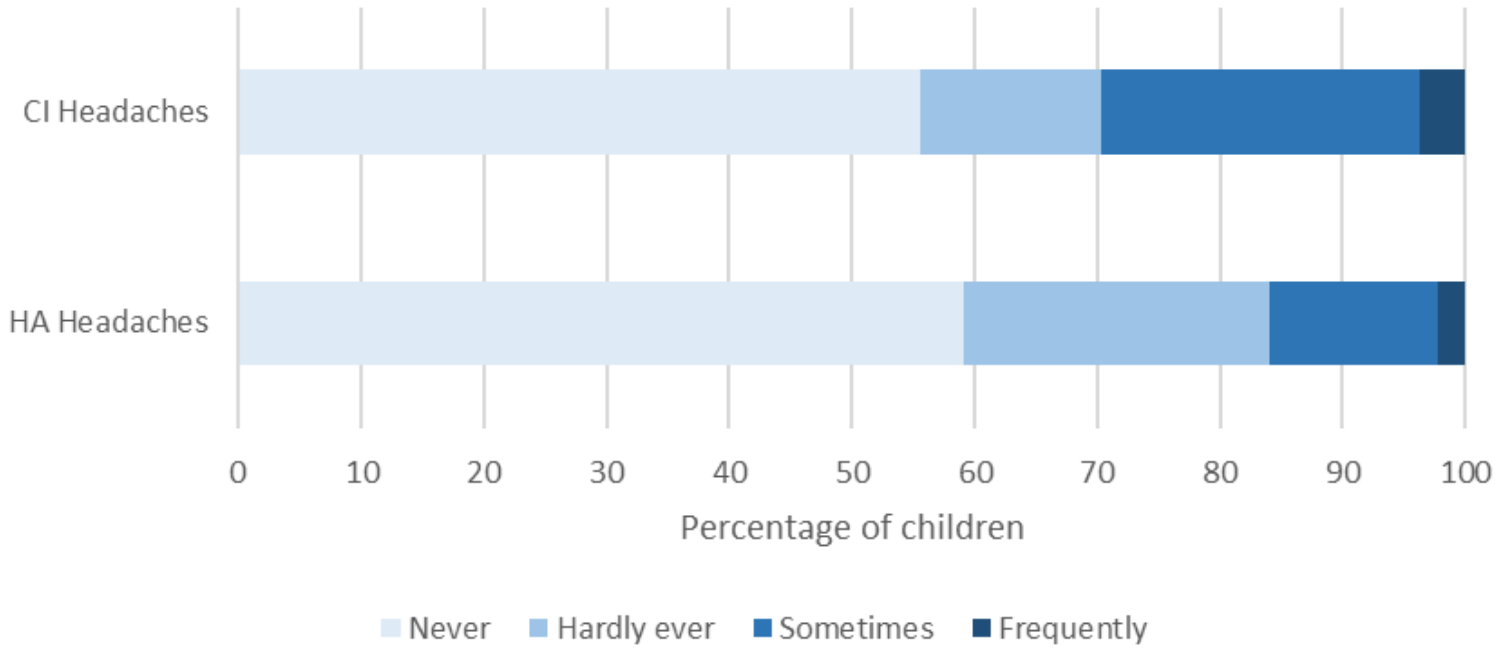


Figure 5.7: The proportion of children falling within each headaches from listening category by device group

HA = Hearing aid; CI = Cochlear implant

Table 5.19: Responses to the question on tiredness related to listening by device group.

Question: *"How often does your child complain of tiredness related to listening?"*		
Response:	**HA Group N = 52**	**CI Group N = 57**
Never	19 (36%)	18 (32%)
Hardly ever	9 (17%)	11 (19%)
Sometimes	15 (29%)	16 (28%)
Frequently	2 (4%)	8 (14%)
"My child is unable to tell me"	7 (13%)	3 (5%)
Missing data	—	1 (2%)

Table 5.19 shows responses to the tiredness questions. Excluding missing data and the children who were unable to tell their parents about tiredness related to listening, 17 out of 45 (38%) of the HA group had reported tiredness related to listening to their caregivers sometimes or frequently, compared to 25 out of 53 (45%) of the CI group. A Pearson's Chi-square test indicated that this difference was not statistically significant (X^2 (1, 98) = .563, p =.453). Binary logistic regression modelling found no significant difference between the device use groups, and no significant associations between tiredness due to listening and gender, educational stage, or the presence of additional needs. Figure 5.8 plots the proportions of responses falling within each tiredness from listening frequency category, excluding children without a rating.

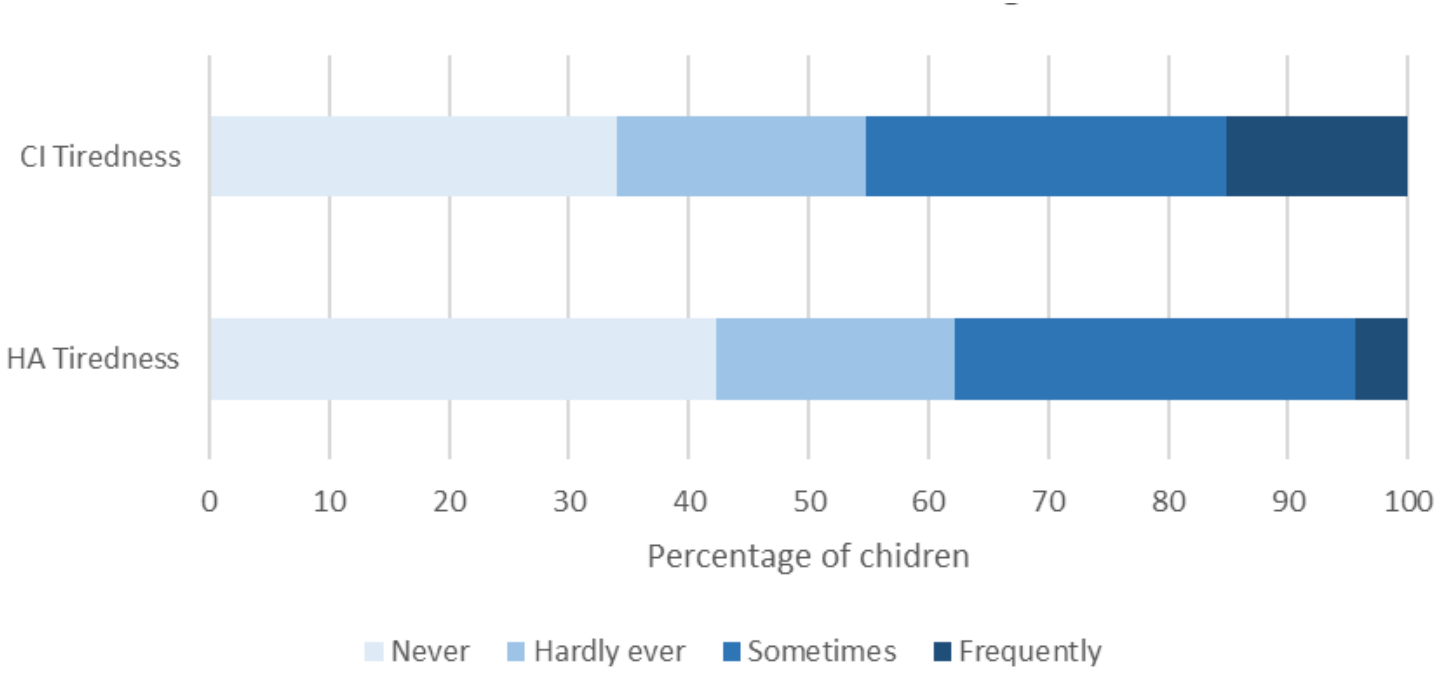


Figure 5.8: The proportion of children falling within each tiredness from listening category by device group

HA = Hearing aid; CI = Cochlear implant

5.3.5. Comparative child device acceptance and caregiver device satisfaction outcomes

This section compares caregiver satisfaction with their child's current device type, children's daily device use, and children's acceptance of their devices between the HA and CI groups.

5.3.5.1. Caregiver satisfaction for the hearing aid and cochlear implant groups

Caregiver satisfaction with the child's current device type was assessed with the question *"Are you satisfied that using [hearing aids / cochlear implants] is in the best interests of your child at the moment?"* Responses were missing for one child from each group, leaving 51 HA users and 56 CI users. Figure 5.9 shows the distributions of respondents' answers. All caregivers of children using CIs reported they were very satisfied that CI use was in their child's best interests. Most caregivers of HA users shared this high level of satisfaction, however responses for the HA group were distributed across all options.

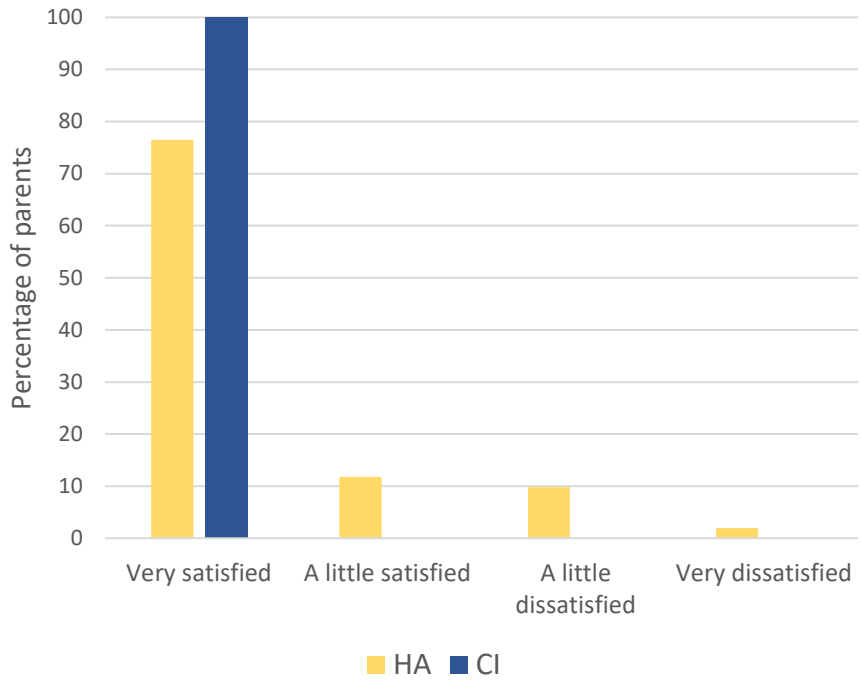


Figure 5.9: Distributions of caregiver satisfaction with children's current hearing devices.

HA = Hearing aid; CI = Cochlear implant

A Mann-Whitney U test was carried out to test for differences between the groups. This revealed significantly lower device satisfaction among caregivers of children using HAs compared to caregivers of children using CIs ($U = 1092$; $z = 2.09$; $p < .05$). The characteristics of HA families falling within each caregiver satisfaction group will be described narratively in the following section, including children's eligibility for CI under NICE, history of prior CI eligibility assessments, and caregivers' own hearing status and device use.

The caregiver with the lowest device satisfaction was the parent of a child within TAG566 (2019) who had recently completed eligibility assessment for cochlear implantation. The family had accepted and were awaiting CI surgery. The five caregivers who were 'A little dissatisfied' that using HAs was in their child's best interests included one family in the middle of CI eligibility

assessment, one family who had never been referred for or considered CI eligibility assessment, and three families whose children had undergone CI eligibility assessments but been declined surgery by their CI team. Three of the children were previously eligible for CIs, one was newly eligible, and the other child's hearing fell outside TAG566 (2019). All of the responder-caregivers and other main caregivers had good hearing and did not use hearing devices themselves.

Six caregivers reported being only 'A little satisfied' that HA use was in their child's best interests. Four of their children had HTLs outside TAG566 (2019) and two could not be classified by TAG566 (2019). One family was in the middle of the CI eligibility process and the others had not considered it.

The remaining 39 caregivers of children with HAs (75% of the HA group) reported being 'Very satisfied' that HA use was in their child's best interests at the moment. They looked after children from every NICE CI eligibility group. The high satisfaction HA group included three children with HTLs within TAG166, all of whom had been offered cochlear implantation and whose families had declined surgery. The group also included nine children who were newly eligible for CIs (having HTLs outside TAG166 but within TAG566). Of these, only one had previously undergone CI eligibility assessment and they had been declined surgery by the CI team. Seven HA users whose CI eligibility via TAG566 could not be determined fell into this group, none of whom had been referred for CI assessment, and none of whom had caregivers who had ever considered this for their child. The remaining 20 children (51% of the 'Very satisfied' group) had HTLs that placed them outside NICE eligibility for cochlear implantation and none of these been assessed for CIs. All of the

families with one or more caregivers who were deaf or hard of hearing reported being ‘Very satisfied’ with their child’s HA use.

5.3.5.2. Child acceptance of hearing devices for the hearing aid and cochlear implant groups

Child acceptance of hearing devices was assessed using the question *“How easy is it for you to get your child to use their [hearing aid / cochlear implant]?”.* This question was asked for right and left ears separately. Figure 5.10 shows the distributions of the groups’ responses. Very similar distributions can be seen between groups. Group comparison was made via two-tailed Mann-Whitney U tests. These confirmed no statistically significant difference in ease of use between HA and CI users. Results were ($U = 1398.5$, $z = -0.18$, $p > .05$) and ($U = 1421$, $z = 0.05$, $p > .05$) for right and left ears respectively.

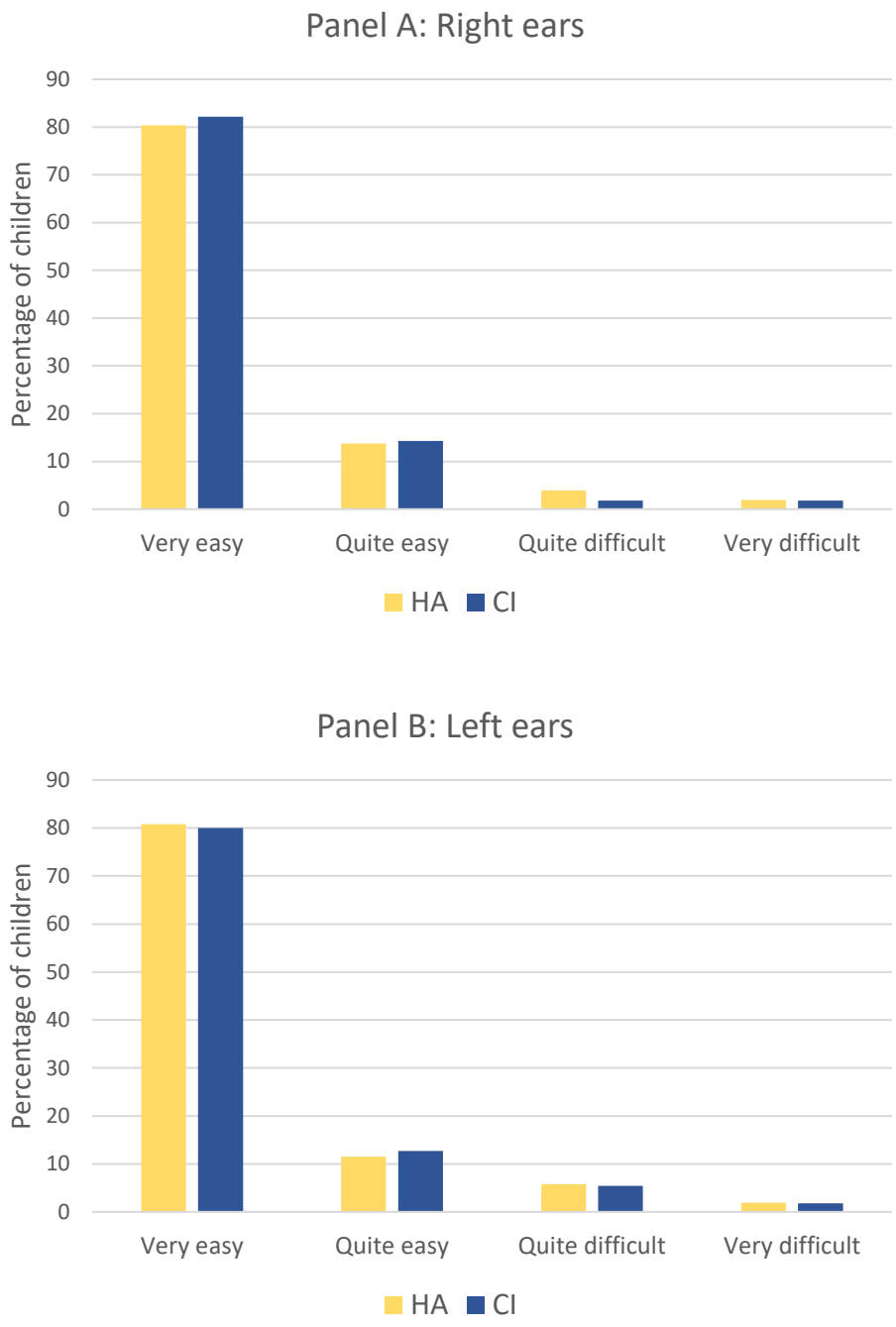


Figure 5.10: Distributions of ease of use categories by device group
HA = Hearing aid; CI = Cochlear implant.

5.3.6.1. Daily device use for the hearing aid and cochlear implant groups

Daily device use was reported for all participants (N=109). Figure 5.11 shows the distribution of children's parent-reported daily device use, in proportions of

the day, with the questions taken from the PEACH questionnaire. A greater proportion of the CI group were reported to wear their devices 'Always' (76 to 100% of the day). However device use was high for both groups, with only a small proportion of HA users being reported to use their devices only 'Seldom' (1-25% of the day) or 'Sometimes' (26-50% of the day).

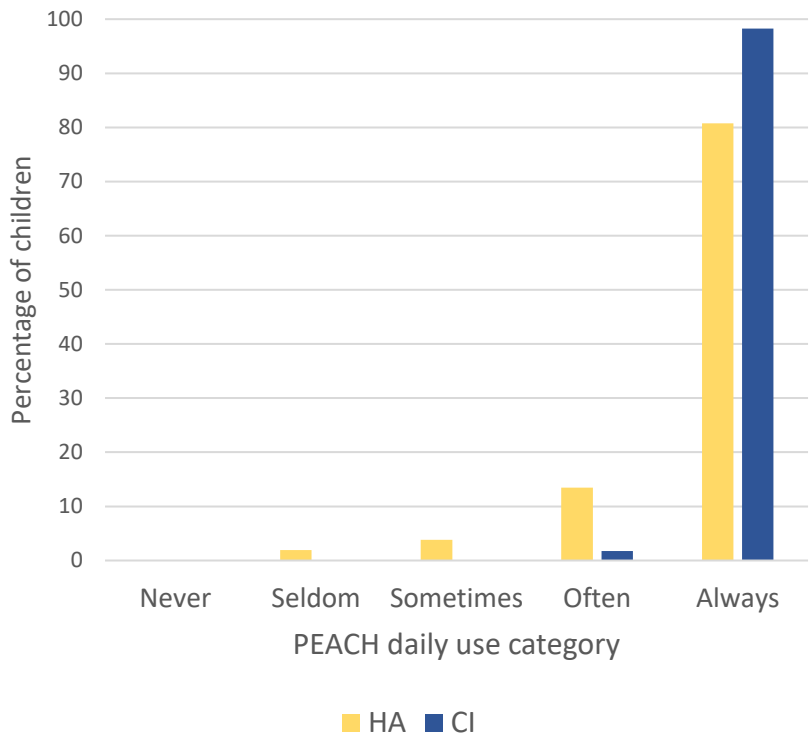


Figure 5.11: Children's daily device use by device group.

HA = Hearing aid; CI = Cochlear implant; PEACH = Parents Evaluation of Aural / Oral Performance questionnaire. 'Never' = 0%; 'Seldom' = 1-25%; 'Sometimes' = 26-50%; 'Often' = 51-75%; 'Always' = 76-100% of the time.

A trend for lower device use among HA users is seen, however a Mann-Whitney U test did not find a statistically significant difference between the groups ($U = 1221.5$; $z = 1.58$; $p > 0.05$).

5.3.6. Representativeness of the sample to the eligible population

Table 5.20 shows the number and proportions of eligible versus recruited families by device use group who were known to need different categories of spoken and/or signed interpreter(s) for audiology appointments.

A few families needing a spoken language interpreter were recruited because their recruitment pack had been printed before the onset of the COVID-19 pandemic and their local services deemed it appropriate for them to be approached. Needing a BSL interpreter was not an exclusion for approach.

Unsurprisingly, the survey population differed from the whole eligible population. Families who did not need an interpreter for hospital appointments were over-represented in both groups. They constituted the whole sample for the CI group, despite comprising less than 80% of the eligible population. It is likely that this was due to the lack of opportunity for participation via a hospital interpreter.

Table 5.20: Interpreter needs of eligible families versus survey respondents

	HA Group		CI Group	
	N Eligible N = 328	Participants N = 52	Eligible N = 322	Participants N = 57
Interpreter(s) needed				
None	251 (77%)	49 (94%)	255 (79%)	57 (100%)
BSL only	17 (5%)	1 (2%)	7 (2%)	—
South Asian and BSL	3 (1%)	—	4 (1%)	—
Eastern European and BSL	10 (3%)	—	2 (<1%)	—
South Asian only	11 (3%)	—	42 (13%)	—
Eastern European only	28 (9%)	—	3 (1%)	—
Other spoken only	8 (2%)	2 (4%)	8 (2%)	—
Missing	—	—	1 (<1%)	—
Any South Asian	14 (4%)	—	46 (14%)	—
Any Eastern European	38 (12%)	—	5 (2%)	—

BSL = British Sign Language

Table 5.21 shows the numbers and percentage of children aged 5 years or older with or without additional needs affecting their ability to participate in hearing tests, comparing all eligible children to the participants. Excluding children under 5 years of age means that the numbers are smaller than those in Table 5.20. Data regarding additional needs were taken from the eligibility screening forms. There was more similarity between the survey participants and eligible population in this regard than with the need for interpreters.

Table 5.21: Additional needs of eligible children aged 5 years or older versus survey respondents

Additional needs category	HA Group		CI Group	
	Eligible N = 284	Participated N = 45	Eligible N = 290	Participated N = 54
No additional needs	219 (77%)	34 (76%)	242 (83%)	51 (94%)
Has additional needs	53 (19%)	8 (18%)	42 (14%)	3 (6%)
Could not extract data from notes	9 (3%)	2 (4%)	4 (1%)	—
Missing	3 (1%)	1 (2%)	2 (1%)	—

HA = Hearing aid; CI = Cochlear implant

5.4 DISCUSSION

This chapter compared the clinical effectiveness of HAs for children with hearing within or close to eligibility for CIs. The following sections discuss the findings as they relate to the chapter objectives.

5.4.1. Recruitment of caregivers of children close to or within NICE TAG566 (2019) and of children using cochlear implants

The first chapter objective was to identify children using CIs and children using HAs with HTLs that placed them within, or close to, NICE eligibility for cochlear implantation and recruit their caregivers to participate in a survey. This was achieved by: developing a survey and information sheets with the input of parents, professionals and researchers; screening 1,965 patient records; approaching 481 eligible families; tracking each with a pseudonomised study identification code; and providing the choice to respond to the survey on paper or online. The caregivers of 109 children were recruited. The 52 children using HAs had unaided thresholds of no more than 20 dB better than the CI candidacy criterion in TAG566 (NICE, 2019), with hearing losses ranging from mild to profound. A similar number of caregivers of children with CIs participated also (N=57).

5.4.2. Participant characteristics

The second chapter objective was to characterise caregivers and their children demographically and audiologically. This was achieved by combining data from the eligibility screening forms with parent-reports of their own and their

children's characteristics within the survey. This meant the population was well characterised, enabling both comparison to other populations and for confounding variables to be controlled for in the comparative analyses that will be discussed in Section 5.4.3.

It has been reported that children of single-parent families were less likely to access CI eligibility assessments than children of married couples in the U.S.A. (Wiley & Meinzen-Derr, 2009). In the present study, a greater proportion of children using HAs were cared for by mothers who did not consider there to be any other significant caregiver for their child. A greater proportion of children using HAs overall were from families where English was not the primary language, and a lower proportion of responder-caregivers from the HA group had a degree-level education or higher. It cannot be known to what extent these group differences arose because these characteristics were barriers to cochlear implantation, because it was necessary to recruit the HA and CI users from different geographical areas. All but one child used either two HAs or two CIs, meaning that the results of this chapter address one of the gaps identified in Chapter 2, as few previously published studies included children using bilateral CIs (Killan et al., 2022). The implications of these comparative analyses will be discussed in the following sections.

5.4.3. Comparative outcomes for children with and without access to cochlear implantation

The next chapter objective was to compare children's caregiver-reported functional listening, tinnitus, and need for recovery from listening, controlling for potentially confounding variables. This was achieved by using the PEACH

questionnaire and novel questions regarding children's reports of tinnitus and need for recovery from listening in the survey.

5.4.3.1. PEACH scores

The key finding of this chapter was that on average, controlling for additional needs and caregiver educational attainment, all groups of children using HAs scored significantly poorer on the PEACH than children using CIs, except for those who could be confirmed to fall outside the audiometric criteria of current NICE guidance (NICE, 2019). This suggests that the change in NICE guidance has achieved its aim of making cochlear implantation accessible to more children who are likely to hear better with CIs than with HAs, while not indicating cochlear implantation for children who are likely to have equivalent aural / oral outcomes via HAs.

However, HA users whose PEACH scores were poorer than CI users included the group whose audiometric eligibility for CIs was unknown due to missing audiometric thresholds. This group could not be confirmed to be audiologically eligible or ineligible for cochlear implantation by TAG566 (NICE, 2019). They also did not meet the BCIG's current guidance for referral for CI eligibility assessment (British Cochlear Implant Group, 2019).

The difference between PEACH scores of CI users compared to children using HAs in this study is in contrast to the group comparison studies of children's functional listening identified in the scoping review, namely Wong et al. (2017) and Meister et al. (2015) who found equivalent performance for children with severe hearing loss using HAs to children using CIs. Both of these studies defined 'severe' hearing loss as having average HTLs between 60

to 80 dB HL, Wong et al. (2017) using a 3-frequency average across 0.5, 1 and 2 kHz and Meister et al. (2015) a 4-frequency average across 0.5, 1, 2, and 4 kHz. It is unclear whether Wong et al. (2017) included any of the eight children in the wider study with losses >80 dB HL in the sub-set of 46 HA users with 'severe' hearing loss whose PEACH scores were compared to the CI users. Only two children with 4-frequency mean HTLs above 80 dB HL were identified by Meister et al., (2015) and these were excluded from comparative analysis.

In contrast, every group of HA users in the present study apart from those confirmed to fall outside TAG566 (2019) included some children with 4-frequency mean HTLs exceeding 80 dB HL. So, it is possible that there were larger proportions of children in this UK sample with HTLs of >80 dB HL without access to CIs compared to children in Australia and Germany, and that this lowered their group's average PEACH scores. This argument is strengthened by considering the publication providing normative data for the PEACH, published a decade earlier than the study by Wong et al. by the same Australian research group (Ching & Hill, 2007). That study included more children with severe and profound losses who had not received CIs and did identify a decrease in HA users' PEACH scores with increasing degree of hearing loss.

The multiple linear regression model identified two characteristics beside NICE eligibility that were significantly associated with parent-reported aural / oral outcomes. In agreement with previous research, higher caregiver education had a significant, positive association with PEACH scores (Noblitt et al., 2018) and a significant negative association was found with the presence

of additional needs (Kuschke et al., 2022). However, it was not expected that the model would account for all the variability in the PEACH scores, and indeed it accounted for less than half of the variance. This was partly due to limitations in the methods used to collect predictor variable data on the impact of additional needs, home language, and daily device use for example. These limitations are discussed below.

The exploratory model allowed for differential effects of different types of additional needs by classifying children into three groups based on the literature (Cupples, Ching, Leigh, et al., 2018). However, these groups were both associated with a 12% lower PEACH score compared to children without additional needs. It is possible that different classifications of degree and type of additional needs could have led to more of the variance in the data being accounted for.

Caregiver reports of daily device use are likely to overestimate true use (Walker, McCreery, et al., 2015). If it had been possible to collect this data based on objectively recorded data-logging, perhaps a more accurate and wider range of daily use would have been found. This might have increased the ability of the model to detect an association between daily use and PEACH scores as reported elsewhere (Kuschke et al., 2022). It is also possible that the children in this sample had higher than typical daily device use, creating ceiling effects.

There are also factors known to influence children's functional auditory performance that could not be included in the model because it was not possible to collect data on them. This included the type of loss and degree of auditory deprivation. Bravo-Torres et al., (2020) found that conductive hearing

loss had a less detrimental impact on PEACH scores than sensorineural loss. Type of loss was not known for the survey participants as it was not feasible to extract the necessary parameters from children's records.

Age at intervention has been found to be associated with PEACH scores (Goh et al., 2018). However, despite a statistically significant difference between the groups, this could not be used in the model as it was not possible to distinguish between congenital, progressive or acquired hearing losses. In studies of children recruited from neonatal hearing screening programmes, ages at hearing loss identification and first HA fitting are sometimes used to quantify duration of auditory deprivation, because it is known that all children had a congenital hearing loss. This can be done by including age at intervention as a variable in modelling factors associated with outcomes and/or by using the difference between chronological age and age at first HA fitting to calculate a "hearing age" (e.g., Uhler et al., 2021). In contrast, recruitment for this survey was based on identifying children of any age from lists of those actively managed with hearing devices and included information from only their two most-recent hearing tests. For some children, later age at diagnosis could represent a disadvantage, due to longer duration auditory deprivation. For others it could be a measure of advantage, due to having had early-life experience of better hearing before a loss was acquired or progressed. Inclusion of variables related to auditory deprivation and type of hearing loss may have resulted in a model that accounted for more of the variance in the data.

It is important to note that the PEACH reflects only caregiver reports of children's aural/oral skills and cannot be sensitive to every difference in

listening and language skills between HA and CI users. Further research is needed to compare the outcomes of children with CIs to children using HAs who are close to CI eligibility across a wider range of outcomes such as those discussed in Chapter 2, including word and phoneme perception in quiet and noise, supra-segmental speech perception, and spatial listening.

5.4.3.2. Tinnitus

A systematic review of childhood tinnitus found that prevalence estimates ranged from 23.5% to 62.2% for children with hearing loss (Rosing et al., 2016), including one study of children with at least one CI in which tinnitus prevalence of 38% was reported (Chadha et al., 2009). The prevalence of 21.9% found in the present study is therefore lower than expected and is likely an underestimate of the true prevalence among respondents' children. One important difference is that this survey asked caregivers if their child had told them they had tinnitus, whereas the studies in Rosing's systematic review were based on direct child-reports (Rosing et al., 2016). Children often do not tell their parents when they experience tinnitus (Mills & Cherry, 1984; Raj-Koziak et al., 2011). It is therefore likely that additional children in the survey cohort did experience tinnitus but their caregivers did not know.

The likelihood that these data underestimate tinnitus prevalence in the sample is strengthened by examining the proportions of children with tinnitus who had told their caregivers it was annoying. This was 83% and 75% of children with tinnitus from the HA and CI groups respectively. These proportions can be compared to other studies of tinnitus in children with hearing loss based on child-reports, although caution must be taken as tinnitus severity, or the degree

to which it was ‘severe’ or ‘bothersome’, is defined heterogeneously between studies. Seven studies of children with hearing loss were included in Rosing and colleagues systematic review (2016). The lowest proportion of children with tinnitus who found it to be ‘severe’ (11%) was from a study that asked children if they were ‘worried all the time’ by their tinnitus (Graham, 1981) and the highest proportion (33.3%) was from a study asking children if they were ‘occasionally concerned’ by their tinnitus (Viani, 1989). Mills & Cherry (1984) found that 23.8% of children with tinnitus were ‘bothered’ by it, a term that is arguably closest to the ‘annoyed’ description used in this survey. Hence it is possible that, in this study, children whose tinnitus was annoying to them were more likely to have told their parents about it than children who experienced tinnitus but did not find it annoying.

Despite the limitations of this caregiver-report study design in giving an accurate tinnitus prevalence estimate, the questions were administered in the same way to both the HA and CI groups. No significant difference was found between groups in the proportions of children experiencing any tinnitus, nor in the proportions with annoying tinnitus. This could suggest that providing CIs to children similar to the survey HA group is unlikely to put them at an increased risk of experiencing tinnitus. However, further research is needed to confirm this. There were too few children with caregiver-reported tinnitus to statistically compare tinnitus affecting children’s sleep between CI and HA users, or to examine the prevalence of tinnitus by NICE eligibility group. (Chadha et al., 2009) found that for children with CIs who experienced tinnitus, this was most experienced at night when the speech processor had been removed. Children rarely use their HAs or CIs while falling to sleep, and

so it remains plausible that in some cases CI surgery changes a child's night-time tinnitus and that this affects their sleep. Prospective, pre-post studies of children's self-reported tinnitus before and after cochlear implantation, and cross-sectional studies of parent- and child-reports of tinnitus for larger groups of CI and HA users with severe hearing loss could give stronger evidence.

In contrast to the findings of Nemholt et al. (2020), sex assigned at birth was not significantly associated with the incidence of tinnitus for this group. Nemholt et al. (2020) studied 501 children, using child-reports of tinnitus. It is possible that the present study's sample size and methodology was under-powered to detect whether there was a difference in tinnitus prevalence between boys and girls.

5.4.3.3. Need for recovery from listening

There was no significant difference between the HA and CI users in the frequency of needing a break from devices, or frequency of tiredness and headaches due to listening reported by caregivers. Nor were school stage, gender, the presence of additional needs, or receptive communication mode found to have significant associations with these outcomes. To the author's knowledge, at the time of writing no comparable data have been published on the prevalence of these experiences among children with hearing loss. These results can therefore be used as a benchmark for future research.

Parents underestimate the fatigue experienced by their children with hearing loss when measured via non-hearing-specific outcome measures (Hornsby et al., 2017; Werfel & Hendricks, 2016). They have also been found to underestimate the prevalence of headaches in the general adolescent population

(Nakamura et al., 2012). It is likely that the same was true in this study. However questions were asked in an identical manner to both the CI and HA groups, so while the results are not expected to be an accurate reflection of children's own subjective experiences, the group comparison is valid. The results tentatively suggest that provision of CIs to the wider range of newly-eligible children might not worsen their need for recovery from listening. However, more research is needed based on child-reports, using the now-available validated tool to assess listening fatigue (Hornsby et al., 2022).

5.4.3.4. Child acceptance of and parental satisfaction with hearing aids or cochlear implants

Overall, caregiver satisfaction with HAs and CIs was high, with all parents of children using CIs reporting that they were very satisfied that CI use was in their child's best interests. However, on average, the caregivers in the HA group were less satisfied than the caregivers of children with CIs.

There was not a straightforward relationship between NICE eligibility group and HA satisfaction. Several children previously eligible for CIs under TAG166 had caregivers who were very satisfied that HA use was in their child's best interests. This agrees with evidence that decision making around cochlear implantation can be influenced by parents' preferences and beliefs about communication mode and the importance of academic success as well as the advice of clinical teams (Li et al., 2004). However, most of the children whose caregivers were dissatisfied with HAs were audiometrically-eligible for CIs. What seemed to differentiate these caregivers was whether the child's HA use had been chosen by the family or imposed upon them by CI professionals.

This could arise if parents felt unable to have ownership of their child's management when their preferences could not be reconciled with the CI team's decision (Kiger et al., 2021). Because of the nature of the CI candidacy selection process, only the HA group included families who had no choice regarding their child's device. In contrast, all families of CI users at one time had been offered the choice as to whether to stay using HAs or accept the professionals' offer of CIs. However, this interpretation is speculative as the number of families with children falling within TAG166 (NICE, 2009) were small.

Overall, high levels of child acceptance of devices were seen for HA and CI users. The distributions of caregivers' reports of how easy it was to get their child to use their hearing devices was near-identical between the HA and CI groups. While there appeared to be a trend toward lower daily device use among the HA users, this was not statistically significant. Only one caregiver of the 57 children using CIs reported that their child used their CIs for less than 75% of the day. This is very similar to the caregiver reported CI use found by Ching et al., (2013). However, reported daily usage was higher than would be expected from a representative clinical population if objectively measured via data logging (Wiseman et al., 2021). It is possible that, in addition to caregiver overestimates of true daily use (Walker, McCreery, et al., 2015), the survey respondents might not have been representative of the wider clinical population.

5.4.4. Characteristics of the survey participants versus the eligible population

Families who needed spoken language interpreters for audiology appointments were under-represented in both the HA and CI groups. This was likely due to sample selection bias introduced by not approaching families who needed an interpreter. There are many other factors known to influence children's hearing-related outcomes that were unknown for the whole population screened, making it impossible to know fully how representative the sample was of the wider population. These factors include maternal education and the richness of the home language environment (Gilkerson et al., 2018; Marnane & Ching, 2015; Noblitt et al., 2018).

Despite these limitations, Table 5.11 shows that the HA children represented in the CUSP survey approximately mirror the age and need for interpreter characteristics of the whole caseloads described in Chapter 4 when classified by NICE eligibility group. For example, the survey participant children using HAs whose HTLs met TAG166 (NICE, 2009) were on average older and had the greatest proportion of homes where English was an additional language. Children represented in the survey whose eligibility for CIs could not be determined by TAG566 (NICE, 2019) were the youngest group of survey participants on average. Direct comparisons are not appropriate because one set of data was from screening forms and the other from care-giver reports, but this is an encouraging sign that the recruited participants were reasonably representative of the population.

HA and CI group characteristics in tables 5.4 to 5.10 could have been statistically compared using tests such as Pearson's Chi-square or Z-tests for comparisons of proportions of these independent groups falling within

different categories. However, the HA and CI survey families were not recruited from the same geographical areas. Government data showed that these areas differed socio-economically. For example, overall the areas included in HA recruitment had a higher proportion of births to non-UK born mothers and shorter life expectancy than the areas from which the CI group were recruited (Office for National Statistics, 2020, 2021). Further, participants constituted only 22% and 23% of their eligible populations for the HA and CI groups respectively. Hence statistical comparisons of the groups' characteristics could not be used as generalisable evidence to compare the characteristics of children who do or do not receive CIs in England. So, rather than directly comparing group characteristics within tables 5.4 to 5.10, these characteristics were included in statistical comparisons of group outcomes as potentially confounding variables where indicated by the literature.

5.4.5. Summary of potential limitations and biases

Families speaking English as an additional language were under-represented in this chapter due to the unplanned exclusion of families who needed spoken language interpreters. The lack of information on conductive hearing loss among the cohort meant this could not be controlled for in analysis of the PEACH. There was also a possible bias in the characteristics of survey respondents compared to the wider population. Caregivers sampled were both willing and able to complete a lengthy survey during the pandemic, possibly while also home-schooling and working under difficult circumstances. It is possible that these families might not be fully representative of the wider cohort in terms of relevant characteristics such as their ability to establish

consistent device use or to have accessed and participated in hearing rehabilitation services during the child's development. Results are also limited by a reliance on caregiver reports that could not be verified, for example caregiver-reported daily device use could not be cross-checked with objectively measured daily use recorded by the devices because of a lack of access to medical records. Last, all parents of children using CIs had the opportunity to choose which device their child would use, while this was not the case for most parents of children using HAs. It is therefore possible that a conscious or subconscious desire to seek to justify this choice might bias their responses toward reporting more favourable outcomes with CIs. However such bias cannot be assumed, especially as no significant difference was found for the HA group outside NICE audiometric guidance and the CI group.

5.5. SUMMARY

This chapter compared the clinical effectiveness of HAs for children with hearing within or close to eligibility for CIs with the clinical effectiveness of CIs. Children outside current NICE guidance TAG566 (NICE, 2019) had equivalent parent-reported aural/oral outcomes to children using CIs. Children using HAs who were previously audiometrically eligible for CIs, newly eligible, or whose eligibility could not be determined, had poorer aural/oral outcomes than children using CIs. There was no significant difference in tinnitus or need for recovery between HA and CI users on the basis of caregiver reports. On average, caregivers of children using CIs had greater satisfaction with hearing device type than caregivers of children using HAs whose children had been declined access to CIs.

CHAPTER 6: EDUCATIONAL OUTCOMES FOR CHILDREN USING HEARING DEVICES FROM THE BORN IN BRADFORD COHORT STUDY

6.1. INTRODUCTION

The scoping review in Chapter 2 found only two papers addressing educational outcomes for children with severe hearing loss using HAs compared to children using CIs (Fitzpatrick et al., 2012; Gantz et al., 2016). Both studies addressed literacy. Fitzpatrick et al., (2012) compared word reading, pseudo-word reading, spelling, and silent reading comprehension in 20 children with HAs, to 21 children with CIs, aged 6 to 18 years. Gantz et al., (2016) compared performance on the Woodcock Reading Mastery Test (Woodcock, 1998) before and 2 years after cochlear implantation with an EAS device, for four adolescents. There is therefore a gap in the evidence base regarding comparative educational outcomes for children under the age of 6 with severe hearing loss using HAs compared to children using CIs. Neither of the studies above were based in the UK healthcare or education systems and neither reported on other educational outcomes such as mathematics. The children studied by Fitzpatrick et al. (2012) had not received newborn hearing screening and consequently had later ages at diagnosis and first intervention than the children identified in the CUSP study, reflecting differences in care a decade ago compared to the present time. Also, it is plausible that differences in clinical and/or educational support in Canada prior to 2012 compared to present-day UK practice limits the generalisability of the findings.

In the UK, every child in state-funded schools has been assessed against the Early Years Foundation Stage Profile (EYFSP) which includes a broad range

of learning goals for literacy, numbers, and personal and social development (Cotzias & Whitehorn, 2013). EYFSP scores have been found to associate with subsequent diagnosis of autism (Wright et al., 2019) but their potential relationship to children's hearing has not been explored. This may be because few datasets in the UK have integrated healthcare and educational outcomes for sufficiently large groups of children.

One example of hearing and education data integration in the UK is the Avon Longitudinal Study of Parents and Children. For this study, audiological test results were gathered not via routine hospital records but at additional research visits that participants could choose to attend. Analyses identified associations between otitis media with effusion, visual impairment, and home language environment (Hall et al., 2014; Hill et al., 2019). The children studied were predominantly of White British ethnicity and the educational outcomes included English, Maths and Science scores at Key Stage 2 (aged 11) and attainment of 5 or more GCSEs at grades A* to C at age 16 (Hill et al., 2019) and IQ at age 8 (Hill et al., 2019).

Another longitudinal study of children in the UK is the Born in Bradford (BiB) cohort study (Wright et al., 2013). This study collected routinely assessed early years educational outcomes for children, along with parental consent for linkage of study data with all routinely collected medical data. Analyses of the BiB dataset has identified the impact of uncorrected visual impairment on early years literacy (Bruce et al., 2016). As a member of staff at Bradford Royal Infirmary and a member of the local care team during my PhD, I had access to routine audiological data to link with BiB data. This created an exciting opportunity to link the CUSP study eligibility screening data to BiB research

data, to explore the impact of hearing loss on children's early educational outcomes. Based on the number of participants, it was expected that there would be too few children with severe or profound hearing loss to address the gap in the evidence base identified by the scoping review in Chapter 2. However, the ethnic composition and educational outcome measures of BiB were different to that of the Avon study. Collaboration offered the potential to derive novel information on how hearing loss impacted the educational outcomes of children during their first year of formal education in a UK context.

This chapter aimed to measure the impact of hearing loss on the early years educational outcomes of children screened for the CUSP study and enrolled in the BiB cohort.

This aim would be achieved via the following objectives:

(i) Establish a study group by linking CUSP eligibility screening data with BiB data to identify children in both studies with documented EYFSP scores.

(ii) Characterise the study group, audiologically and by characteristics known to be significantly associated with EYFSP scores from the literature.

(iii) Create a control group of children matched to the study group for potentially confounding characteristics.

(iv) Compare the study group's EYFSP scores to nation-wide benchmark data.

(v) Measure the impact of hearing loss on children's EYFSP scores via statistical comparison of the study and control groups.

6.2. METHODS

This was a retrospective observational study of educational outcomes for children who had used hearing devices at the time of CUSP study eligibility screening and who were also enrolled in the BiB cohort, compared to a group of matched controls.

6.2.1. Data sources

Mothers had been recruited between 2007 and 2010 at Bradford Royal Infirmary, while pregnant, at a routine oral glucose tolerance test. (Raynor, 2008). The design of the BiB study enabled linkage of the routine clinical audiology data collected during CUSP study eligibility screening with an extensive dataset including maternal health records, socio-economic and research data, and data from the Local Authority education records. A data dictionary outlining the parameters available could be viewed via the BiB website (BiB website, accessed in 2020). Audiological data were extracted from AuditBase, the electronic record system used at Bradford Royal Infirmary. BiB participants had already consented for BiB research data to be linked to the child's routine clinical data, and so the HRA granted ethical approval for me to link audiology data with BiB dictionary data for children who had been screened for the CUSP study without obtaining further parental consent.

6.2.2. Data linkage

All patients under paediatric audiology at Bradford Royal Infirmary issued with at least one hearing aid or bone conduction aid as of November 2019 had been screened for eligibility for the CUSP study, as described in section 4.2 (N=757). All children who had received at least one CI at the Yorkshire Auditory Implant Service (YAIS) had also been screened as described in section 5.2.3, and filters within the YAIS electronic audiology records system AuditBase were used to identify children with CIs who were both born within the BiB recruiting period and had a Bradford postcode (N=18). Following the HRA approved protocol, these children's hospital numbers were shared with the BiB data custodian who determined that 77 children using any kind of hearing device (HA, bone conduction aid or CI) at the time of CUSP study eligibility screening were also enrolled in the BiB cohort study. Figure 6.1 is a flow diagram illustrating the identification of these children. Children who completed their Reception year (the academic year during which children turn five years of age) in 2012 had not been assessed with the same EYFSP framework and therefore had missing data. One further child was excluded as their non-normal HTLs were noted in their medical records as being believed non-organic (having no physiological origin). In total, 64 children with confirmed hearing loss had both used hearing devices at the time of CUSP eligibility screening and had early years educational outcome data available.

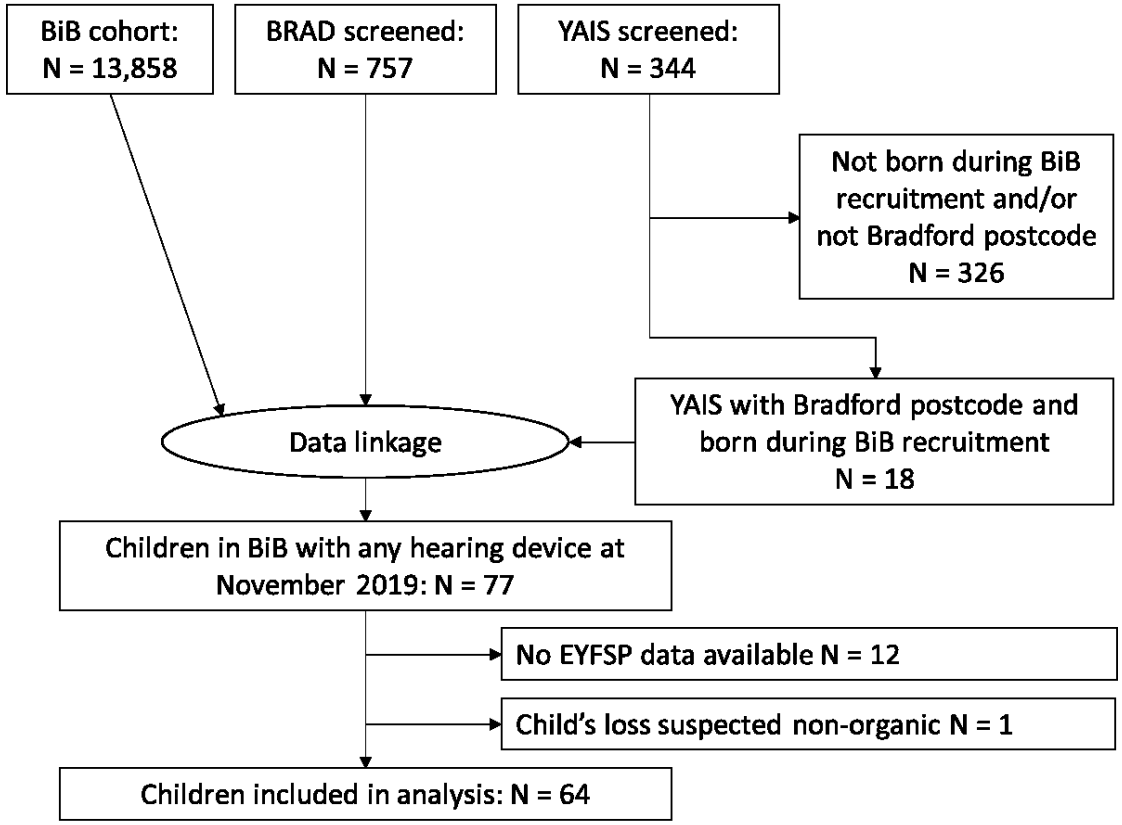


Figure 6.1: Flow diagram of the identification of children using hearing devices and within the Born in Bradford cohort.

BiB = Born in Bradford; BRAD = Bradford Royal Infirmary paediatric hearing aid caseload; YAIS = Yorkshire Auditory Implant Service paediatric cochlear implant caseload; EYFSP = Early Years Foundation Stage Profile.

6.2.3. Audiological data extraction

The clinical audiology data available from the CUSP eligibility screening forms has been described in Chapter 4 for children using HAs, and in Chapter 5 for children using CIs. Children managed with either HAs or bone conduction aids were on the same waiting list system at Bradford Royal Infirmary, hence all children using any hearing device had been assigned a pseudonomised CUSP study identification code. However, for children using bone-conduction aids extraction of HTLs was not required so these children had missing audiometric data. Furthermore, once the linked data were received from the BiB team, it was discovered that the EYFSP assessments had taken place up to several years prior to the audiograms on the CUSP eligibility screening forms. Subsequently, it was necessary to revisit each of the eligible children's audiology records and extract data from the appointments that took place closest in time to both the EYFSP assessments, including the children who might not have had their HTLs extracted for Chapter 4 as they used bone conduction aids. Unaided thresholds were not extracted for ears implanted with a CI at the time of the EYFPS assessment.

As described in Chapter 3, data were first extracted by a specialist ENT registrar who was able to be on site to access the data during the early stages of the COVID-19 pandemic. A data extraction form was designed with space for any queries regarding data interpretation to be noted. Once direct access to the hospital systems was once again permitted, the cases where the registrar had been unsure (for example over how to classify a hearing device based only on its trade name, how to report HTLs where the child had not responded at a

certain stimulus level) were rechecked and a selection of other cases were reviewed at random to check for the accurate transcription of data.

Children were classified as meeting, falling outside, or not being classifiable by TAG566 (NICE, 2019) and their better- and worse-hearing ears were classified by the BSA hearing loss descriptor categories (British Society of Audiology, 2018) based on a four-frequency average including 0.5, 1, 2 and 4 kHz, or as many of these frequencies as were available. Children were classed as having hearing within normal limits if the four-frequency average was <21 dB HL in both ears. If one ear fell within this range but the other was ≥21 dB HL, the child was classed as having unilateral hearing loss. If both ears had a four-frequency average ≥21 dB HL, the child was classed as having bilateral hearing loss.

6.2.4. Outcome measure: Early Years Foundation Stage Profile total scores and Good Level of Development

This chapter relied on retrospective analysis of previously collected data, so the EYFSP was chosen as it was already available within the BiB dataset. This assessment is applied to every child in a state-funded early years education setting in England during the final term of the 'Reception' year. Across 17 learning items, teachers classified children as 'Emerging', 'Expected' or 'Exceeding'. A 'good level of development' (GLD) is defined as gaining 'expected' or 'exceeding' in each of the 12 items within the domains 'communication and language', 'physical development', 'personal, social and emotional development', 'literacy', and 'mathematics' (Cotzias & Whitehorn, 2013). Other domains include 'understanding of the world', and 'expressive

arts and design' (Cotzias & Whitehorn, 2013). Table 6.1 lists each of the 17 items within these domains.

Table 6.1: Domains and items of the Early Years Foundation Stage Profile

EYFSP Domains	EYFSP item codes	EYFSP item names
Domains used for establishing GLD		
Communication and language		
	ELG 01	Listening and attention
	ELG 02	Understanding
	ELG 03	Speaking
Physical development		
	ELG 04	Moving and handling
	ELG 05	Health and self-care
Personal, social and emotional development		
	ELG 06	Self-confidence and self-awareness
	ELG 07	Managing feelings and behaviour
	ELG 08	Making relationships
Literacy		
	ELG 09	Reading
	ELG 10	Writing
Mathematics		
	ELG 11	Numbers
	ELG 12	Shape, space and measures
Other Domains		
Understanding of the world		
	ELG 13	People and communities
	ELG 14	The world
	ELG 15	Technology
Expressive arts and design		
	ELG 16	Exploring and using media and materials
	ELG 17	Being imaginative

Key to Table 6.1: EYFSP = Early Years Foundation Stage Profile; ELG = Early learning goal; GLD = Good level of development.

6.2.5. Selection of control variables from the BiB dataset

The EYFSP was revised, and the new version piloted, during 2012. It is the revised version, used nation-wide from the Spring of 2013 onward, that was

used for the present study, as most BiB participants were assessed via this version. Cotzias & Whitehorn (2013) published a report for the Department of Education on the outcomes of the 2012 pilot scheme, based on data gathered from approximately 500 schools across 19 local authorities. The report identified significant associations between certain characteristics and total EYFSP point scores. Characteristics with statistically significant positive associations to total EYFSP scores included: being a girl, being born during the autumn term, and being of 'Indian' ethnicity (compared to 'White British'). Characteristics with statistically significant negative associations included: being born in the Summer term; being in receipt of free school meals; English being an additional language; having special educational needs; and being a member of 'Gypsy/Romany', 'Irish Traveller', 'Bangladeshi', 'Pakistani', 'Black Other', 'White Other', 'Black Caribbean', 'Other', or 'Black African' ethnic groups (compared to 'White British'). The authors noted that this model did not control for every other likely influential characteristic, noting the omission of parental education (Cotzias & Whitehorn, 2013), nor did the authors report any analyses of inter-correlation of predictor variables. Later analyses of EYFSP data within the BiB dataset revealed further significant associations, including beneficial effects of longer gestation (Pettinger et al., 2019) and a negative impact of the presence of autism (Wright et al., 2019). All of these potential control variables were available and therefore included in analysis apart from autism diagnosis.

6.2.6. Creating the control group

A control group was created by matching each child in the hearing loss study group with a child from the wider BiB cohort who had EYFSP data but no CUSP eligibility screening form. First, children were matched for characteristics known to be significantly associated with EYFSP scores from the local authority education records, namely ethnic origin category, academic term of birth, whether the child received free school meals, whether the child spoke English as an additional spoken language, and gender (Cotzias & Whitehorn, 2013). If a child had no additional needs identified on their CUSP eligibility screening form, they were matched to cases recorded as 'Not SEN' (Special Educational Needs Support). If a child had additional needs noted on their eligibility screening form, they were matched to cases recorded as 'ECHP' (Education and Health Care Plan) or 'SEN Support'.

Each child within these 64 groups of matched cases were then reviewed to find those that also matched for gestational age and maternal education based on the BiB Baseline database. Where possible, children were matched to those with the same gestational age in completed weeks and maternal education. Where this was not possible, matches were sought with gestational age within 1 week and the same maternal education category. Where this was not possible, children were matched to those with the closest available maternal education category. Where a study group child's maternal education was missing, a match whose maternal education was also missing was chosen. Where this was not possible, maternal education was not matched for.

After this matching process, when more than one potential matched case was available, one control child was selected at random.

6.2.7. Statistical methods

Total EYFSP scores were calculated in the same manner as Cotzias & Whitehorn, (2013), by assigning scores of 1 for 'Emerging', 2 to 'Expected', and 3 to 'Exceeding' for each of the 17 items and summing the total, producing a score between 17 and 51. Whether each child met the GLD criterion was assessed by calculating if they achieved 'Expected' or 'Exceeding' for all 12 relevant items.

The proportion of children meeting the GLD criterion, and the proportion achieving 'Emerging', Expected', or 'Exceeding' for each of the 17 items were analysed via descriptive statistics for comparison to national benchmark data. The difference between total EYFSP scores achieved by the study and control groups were analysed via dependent two-tailed t-tests for matched pairs. Differences in total EYFSP scores for study and control groups of children with either unilateral, mild, or moderate degree of hearing loss were compared via Mann-Whitney U tests. The proportions of children in the study and control groups attaining the GLD were compared via Pearson's Chi-Square test. Children with severe or profound losses and CI users were excluded from sub-group analysis due to their small numbers.

6.3. RESULTS

This section achieves the chapter objectives of describing the study group's demographic and audiological characteristics, documenting the outcome of

case-control matching, comparing the study group's EYFSP results to nation-wide benchmark data, and exploring the impact of hearing loss on EYFSP results by comparing data for the study and control groups.

6.3.1. Participant characteristics

This section describes the study group's demographic and audiological characteristics.

6.3.1.1. The study group's demographic characteristics

Table 6.2 summarises the study group's demographic characteristics using data from the BiB data dictionaries. Additional data from the CUSP forms are shown regarding additional needs or need for interpreter noted in their audiology records.

Table 6.2: Children's demographic characteristics

	Study Group N = 64
Sex assigned at birth	
Number Male (%)	27 (42)
Gestational age	
Median gestational age in completed weeks (range)*	39 (28 to 42)
Number in each academic term of birth group (%)	
Autumn	25 (39)
Spring	17 (27)
Summer	22 (34)
Family socio-economic status at child's birth	
Median National IMD¶ 2010 decile score (interquartile range)	2 (1.00 to 3.75)
Median Bradford IMD¶ 2010 decile score (interquartile range)	4 (2.00 to 6.00)
Child received free school meals	17 (26)
Number of children in each ethnicity group (%)	
Bangladeshi	2 (3)
Black Caribbean / Black Other	1 (2)
Mixed	1 (2)
Pakistani	37 (58)
White British	23 (36)
Special needs	
Number of children receiving any SEN Support (%)	55 (86)
Additional needs noted on CUSP screening form (%)	9 (14)
Maternal education∫	
Number with < 5 GCSE equivalent (%)	9 (14)
Number with 5 GCSE equivalent (%)	29 (45)
Number with A-level equivalent (%)	8 (13)
Number with Higher than A-level (%)	8 (13)
Number with Foreign unknown (%)	1 (2)
Number with "Other" (%)	2 (3)
Number "Don't know" (%)	2 (3)
Home language	
Child spoke English as an additional language†	32 (51)
Interpreter needed for audiology appointments from CUSP screening form	11 (17)

IMD = Index of Multiple Deprivation; SEN Support = Special Educational Needs support provided through school; GCSE = General Certificate of Secondary Education; *Gestational age was missing for one child. ¶IMD decile scores were missing for 6 children; ∫Maternal education was missing for 5 children; †English language data were missing for one child.

Of the 11 children whose families needed interpreters for audiology appointments, eight needed a South Asian language interpreter alone, one needed both South Asian and BSL interpreters, one needed a BSL interpreter

alone, and one was noted to need an interpreter but no language was recorded. The Index of Multiple Deprivation (IMD) scores show the relative socio-economic status of families on a scale from 1 to 10 inclusive. Table 6.2 shows that at least 75% of the study group came from the poorest 40% of families in the country. Comparison of the national and Bradford IMD data reveals the relative poverty of Bradford compared to the nation as a whole, and the median IMD score of the study children was below that of the Bradford average.

6.3.1.2. Audiological characteristics

Two children's unaided HTLs were measured in a soundfield only. In these cases, it was assumed that hearing loss was symmetric, and the same soundfield thresholds were attributed to both ears for analysis. For the other 62 children, ears fitted with acoustic hearing aids, bone conduction aids, or no listening device had ear-specific unaided threshold data. Children were identified because they used hearing device(s) under the care of either paediatric audiology or YAIS at the time of eligibility screening during autumn 2019. However, their historical audiology records revealed that many were not aided at the times of their EYFSP assessments during 2013 to 2016. The study group included children with sensorineural, conductive and mixed hearing losses, as well as children for whom the type of loss could not be determined. Table 6.3 summarises the types of losses by right and left ears. Overall, type of loss could be confirmed for 69% of right ears and 67% of left ears. For the remaining cases, there was either ambiguity regarding whether the losses were mixed or not, or children could not be classified.

Table 6.3 Type of hearing loss at EYFSP assessment

Type of hearing loss	Right ears N=64	Left ears N=64
Confirmed sensorineural loss	21 (33%)	24 (38%)
Confirmed conductive loss	9 (14%)	11 (17%)
Confirmed mixed loss	4 (6%)	4 (6%)
Either sensorineural or mixed loss	3 (5%)	3 (5%)
Either conductive or mixed loss	7 (11%)	6 (9%)
Normal hearing	10 (16%)	4 (6%)
Could not be classified	10 (16%)	12 (19%)

Table 6.4 presents children's audiological characteristics including degree of hearing loss, NICE CI eligibility category and hearing device use.

Table 6.4: Audiological characteristics

	Study Group N = 64
Better-ear hearing loss	
Normal (<21 dB HL)	15 (23%)
Mild (21 to 40 dB HL)	23 (36%)
Moderate (41 to 70 dB HL)	20 (31%)
Severe (71 to 95 dB HL)	1 (2%)
Profound (> 95 dB HL & CI users)	5 (8%)
Worse-ear hearing loss	
Normal (<21 dB HL)	0
Mild (21 to 40 dB HL)	19 (30%)
Moderate (41 to 70 dB HL)	32 (50%)
Severe (71 to 95 dB HL)	8 (13%)
Profound (> 95 dB HL & CI users)	5 (8%)
Audiometric eligibility for CI by NICE Guidance	
Within TAG566 (2019)	6 (9%) (N=4 with CI)
Outside TAG566 (2019)	54 (84%)
Could not classify by TAG566 (2019)	4 (6%)
Hearing loss symmetry	
Hearing within normal limits bilaterally	0
Unilateral hearing loss	15 (%)
Bilateral hearing loss	49 (%)
Devices	
Bone conduction aid	2 (3%)
Acoustic HA(s)	27 (42%)
Bilateral CI	3 (5%)
Unilateral CI and contralateral HA	1 (2%)
Not aided	31 (48%)
Device use by hearing loss symmetry	
Unilateral hearing loss and unaided	12 (19%)
Unilateral hearing loss and aided	3 (5%)
Bilateral hearing loss and unaided	19 (30%)
Bilateral hearing loss and aided	30 (47%)

CI = cochlear implant; HA = hearing aid; NICE = National Institute for Health and Care Excellence; TAG566 = Technology Appraisal Guidance 566. Degree of hearing loss categories derived from British Society of Audiology descriptors.

6.3.2. Results of case control matching

All study group children could be matched to other BiB cohort children with post-2012 EYFSP data who did not have a CUSP screening form and shared all the characteristics known to influence EYFSP scores from within the local authority education record database. The number of potential matches ranged from four to over 500. The next stage involved identifying which children also matched for gestational age and maternal education. Gestational age was missing for one study group child. In this case the median value of the whole BiB cohort (39 weeks) was assumed. There were more missing data for maternal education than other variables. For ten children it was not possible to match for maternal education because it was either missing or marked as 'Other' in the records of the study group child. For three children, no exact maternal education match was available who also matched for the other parameters (N=3).

Overall, at least one complete match was found for 48 children (75%), defined as matching exactly for ethnic origin; academic term of birth; English as an additional language; free school meals; additional needs; gender; gestational age to the same week, and maternal education. The remaining 16 study group children (25%) were partially matched to control cases, defined as having gestational age within one week of target and / or maternal education matching not being possible. The number of potential matches per study child varied from one to 44. This was reduced to one control child per study child by randomly selecting form the available matched controls, creating a group of 64 control cases.

Table 6.5: Study and Control group demographic characteristics

	Study Group N = 64	Control Group N = 64
Sex assigned at birth		
Number Male (%)	27 (42)	27 (42)
Gestational age*		
Median gestational age (weeks)	39	39
IQR gestational age (weeks)	38 to 40	38 to 40
Range gestational age (weeks)	28 to 42	28 to 42
Academic term of birth (%)		
Autumn	25 (39)	25 (39)
Spring	17 (27)	17 (27)
Summer	22 (34)	22 (34)
Family socio-economic status N (IQR)		
Median National IMD¶ 2010 decile	2.0 (1.00 to 3.75)	2.0 (1.00 to 2.75)
Median Bradford IMD¶ 2010 decile	4.0 (2.00 to 6.00)	3.5 (2.00 to 5.00)
Child received free school meals	17 (26)	17 (26)
Ethnicity group N (%)		
Bangladeshi	2 (3)	2 (3)
Black Caribbean / Black Other	1 (2)	1 (2)
Mixed	1 (2)	1 (2)
Pakistani	37 (58)	37 (58)
White British	23 (36)	23 (36)
Special needs N (%)		
Child receiving any SEN Support (%)	55 (86)	9 (14)
Additional needs noted on CUSP screening form (%)	9 (14)	—
Maternal education		
Number with < 5 GCSE equivalent (%)	9 (14)	10 (16)
Number with 5 GCSE equivalent (%)	29 (45)	32 (50)
Number with A-level equivalent (%)	8 (13)	9 (14)
Number with Higher than A-level (%)	8 (13)	7 (11)
Number with Foreign unknown (%)	1 (2)	0
Number with “Other” (%)	2 (3)	0
Number “Don’t know” (%)	2 (3)	0
Missing (%)	5 (8)	6 (9)
Home language		
Child EAL†	32 (51)	33 (52)
Interpreter at audiology appointments	11 (17)	—

IMD = Index of Multiple Deprivation; SEN Support = Special Educational Needs support provided through school; GCSE = General Certificate of Secondary Education; IQR = Interquartile range; EAL = English as an additional language. *Gestational age was missing for one child from the study group. ¶IMD decile scores were missing for 6 children from the study group and 6 children from the control group; †English language data were missing for one child from the study group.

Table 6.5 contrasts the demographic characteristics of the study and control groups. Despite some missing data from the study group, the overall gestational ages of the groups did not differ in terms of the median, range, or inter-quartile range between the study group and matched controls. Small differences can be seen in characteristics that were not controlled for, such as the index of multiple deprivation deciles. This indicated that the control group came from slightly more deprived families than the study group. The control group included fewer children whose maternal education was classed as "Foreign unknown", "Other" or "Don't know". The most frequent maternal education category was "5 GCSE equivalent" for both groups.

6.3.3. Study group and nation-wide EYFSP data

In the present study, 17 out of 65 (26%) of the study group children achieved a GLD on the EYFSP, compared to 41% nation-wide (Cotzias & Whitehorn, 2013). This section will also compare the distribution of total EYFSP scores, and the proportions of children scored as 'Emerging', 'Expected', or 'Exceeding' for each of the 17 items.

Figure 6.2 is a reproduction of Chart 2.4 from the Department for Education report on the EYFSP pilot (Cotzias & Whitehorn, 2013)[3]. It is plotted here beside Figure 6.3 which shows the equivalent distribution of point scores for

[3] The document (Cotzias & Whitehorn, 2013) states "You may re-use this information (excluding logos) free of charge in any format or medium, under the terms of the Open Government Licence" (p79). Reproduction of the figures within this thesis falls within these terms, which are available at: https://www.nationalarchives.gov.uk/doc/open-government-licence/version/3/

children with hearing loss in the present study. Comparison of Figures 6.2 and 6.3 reveals the differences in group mean total point scores, which was 32 points nation-wide in 2012, and 24.7 points for the study group. There was a 10-fold difference in the proportion of children being given the lowest possible score of 17 (not meeting the expected level of development for any item). This was 2.4% of children in the national pilot dataset and 25% of the study group. In contrast to Cotzias & Whitehorn, (2013), the study group had a bimodal total score distribution. The modal score was the lowest available and a second peak was seen at the modal score of the national pilot data, namely 34 points. This score can be obtained through various combinations of 'Emerging', 'Expected', and 'Exceeding' grades across the 17 items but is also the score gained by scoring 'Expected' in all items.

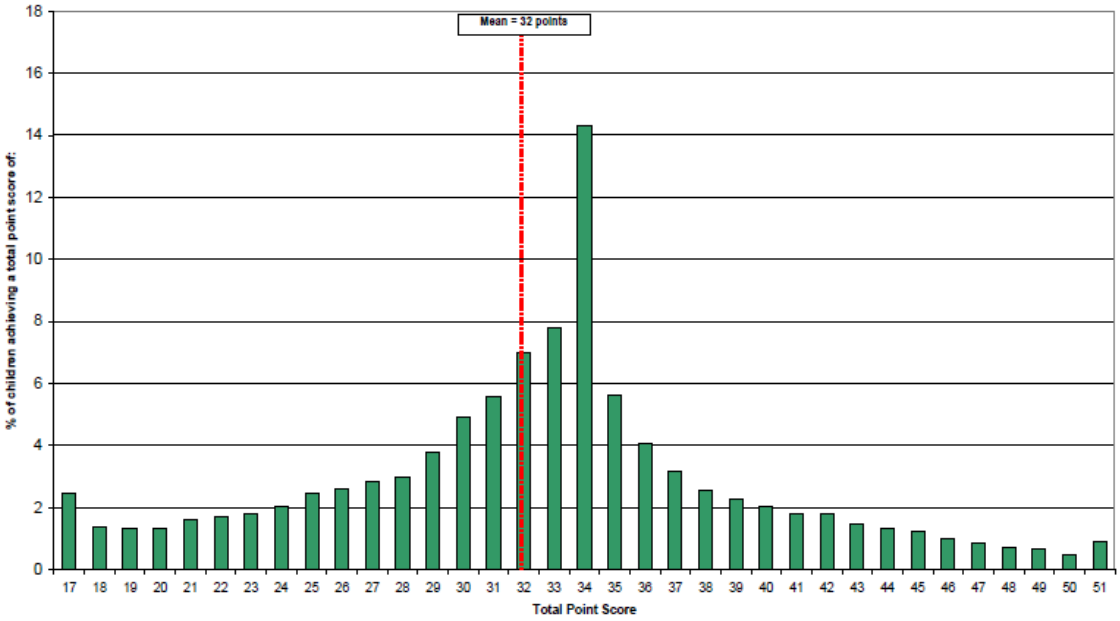


Figure 6.2: Distribution of total EYFSP point scores Department of Education Report on EYFSP pilot data for all children (Cotzias & Whitehorn, 2013).
Mean score (32 points) indicated by the vertical red dashed line

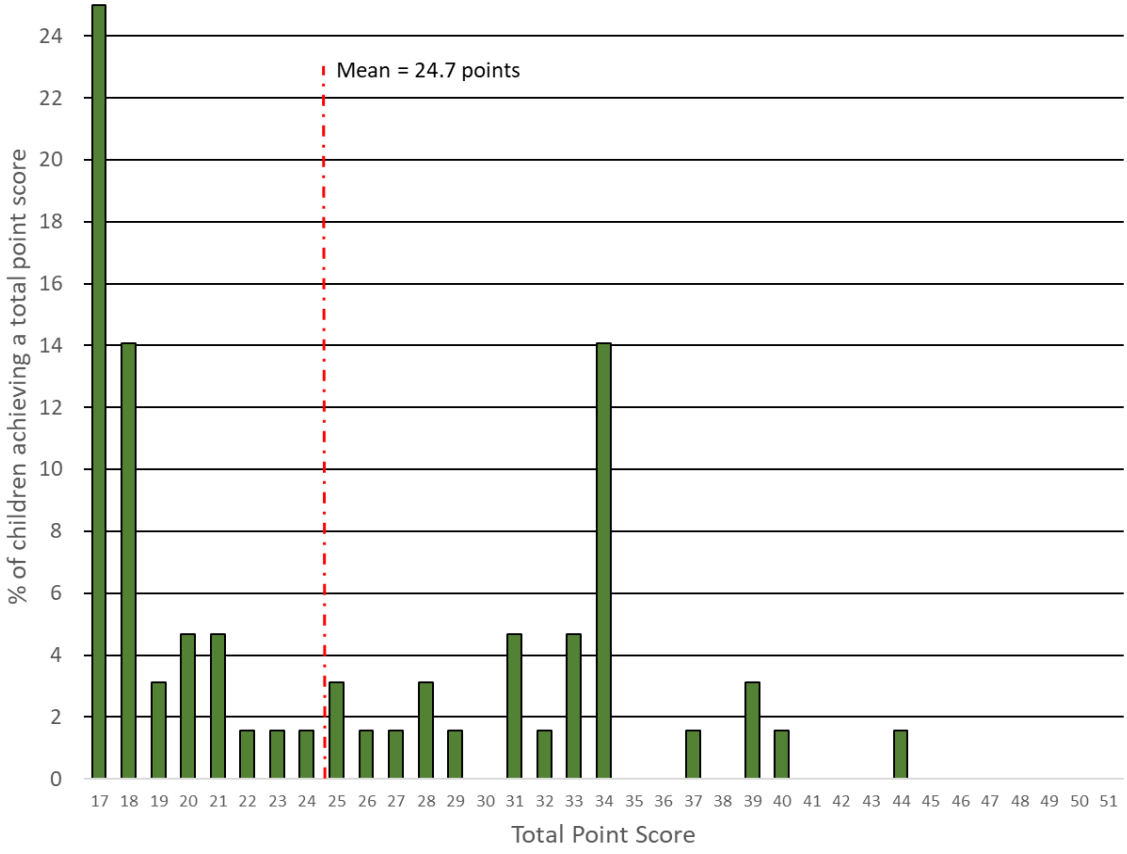
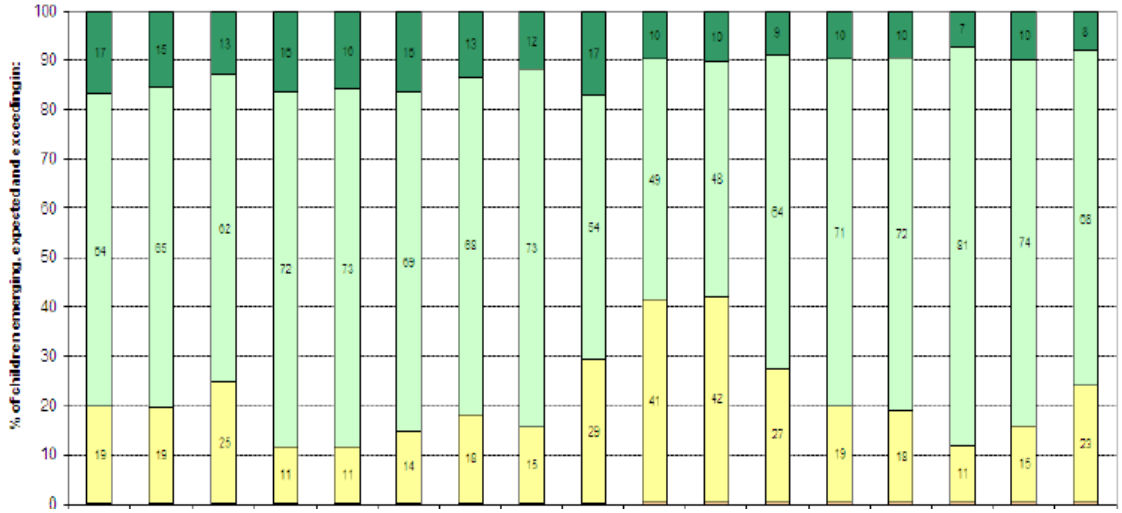


Figure 6.3: Distribution of total EYFSP point scores for children in the present study.
Mean score (24.7 points) indicated by the vertical red dashed line

Figure 6.4 shows Chart 2.7 from Cotzias & Whitehorn, (2013) (Panel A) alongside equivalent data from the study group of the present study (Panel B). This figure reveals greater proportions of children in the present study being scored as ‘Emerging’ across all 17 items compared to the nation-wide data.

Panel A: Data from Cotzias & Whitehorn, (2013)



Panel B: Data from the study group in the present study

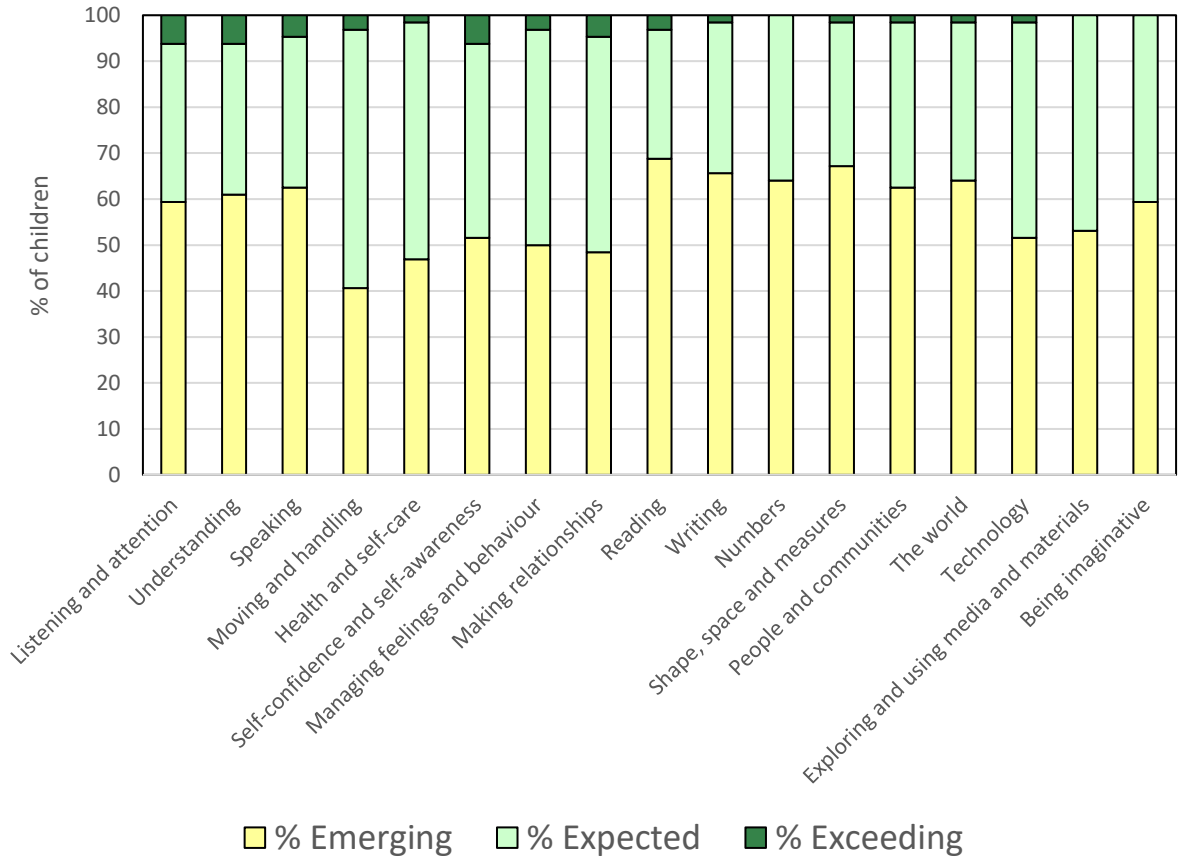


Figure 6.4: Proportions of children assessed as 'Emerging', 'Expected', and 'Exceeding' across all 17 EYFSP profile items for nation-wide pilot data and the present study.

There are several reasons why it is problematic to use the data within Cotzias & Whitehorn's report (2013) as a comparison group to estimate the impact of hearing loss on the study group's outcomes. First, the pilot dataset was collected in 2012 whereas the study group were assessed in 2013 to 2016 inclusive. It was possible that children's outcomes had changed over time as teachers adapted to the new assessment system. While year of assessment was not used as a matching variable, the process did result in the study and control groups both including children assessed across the four assessment years. The proportions of study and control group participants assessed during each year are shown in Table 6.6.

Table 6.6: Numbers of study and control group participants undergoing EYFSP assessments per year

Year of assessment	Study Group (N=64)	Control Group (N=64)
2013	22	21
2014	17	15
2015	12	23
2016	13	5

Second, the pilot data were weighted to represent the national demographics of children across England. However, the report does not state what proportion of children nationally had each of the relevant characteristics including gender, ethnic group, or being in receipt of free school meals. This precludes a direct comparison as these characteristics are likely to differ between the whole country and the sub-set of children in the present study. Further, in terms of special needs, Cotzias & Whitehorn, (2013) only classified children as either being in receipt of any special educational needs support from their school or

not. Hearing loss can be sufficient cause for SEN support to be provided in the absence of any other additional needs. This means the national pilot study data cannot distinguish between children with normal hearing, children with hearing loss alone, and children with hearing loss and additional needs. Table 6.5 shows that 86% of the study group received SEN support but only 14% had notes of disabilities affecting their ability to do hearing tests on their eligibility screening forms. Whilst it was established in Chapter 5 that the eligibility screening form data are an under-representation of all additional needs, it remains plausible that many of the children were receiving SEN Support due to their hearing loss alone. So, while this section has achieved the chapter objective of comparing the EYFSP scores to nationwide benchmark data, providing the wider context for the performance of the study group, alternative methods must be used to parse out the effects of hearing loss on children's outcomes.

6.3.4. Relationships between degree of hearing loss, aided status, and total EYFSP scores

Figure 6.5 plots total EYFSP scores against the 4-frequency mean HTLs of children's better-hearing ears for those without CIs. Children with CIs are shown on an adjacent axis. Figure 6.5 reveals interesting trends in the data. First, the overall range of total EYFSP scores varies by hearing loss classification group for the children without CIs. Children with unilateral or mild losses had the widest score distributions, with scores both above and below the national average. In contrast, no children with moderate hearing loss or greater, including those with CIs, scored above the national median score of 34. Second, some children across the whole range of 4-frequency average HTLs scored either 18 or 17, meaning that they had failed to reach the 'Expected' level of development in either all, or all but one of the 17 learning goals.

Last, a relationship can be seen between degree of hearing loss and the likelihood of a child being aided at the time of their EYFSP assessment. All but three children with unilateral losses were unaided and all children with average better-ear HTLs of 45 dB HL or greater were aided. More even proportions of aided and unaided children were seen for mean HTLs between 30 and 45 dB HL. This means that regression modelling to find any association between degree of hearing loss and EYFSP scores would be confounded by aided status and vice versa. Further, the number of study group participants was too small to meet the assumptions of linear regression modelling as described in Chapter 5, due to the large number of other variables known to influence EYFSP scores that would need to be controlled for.

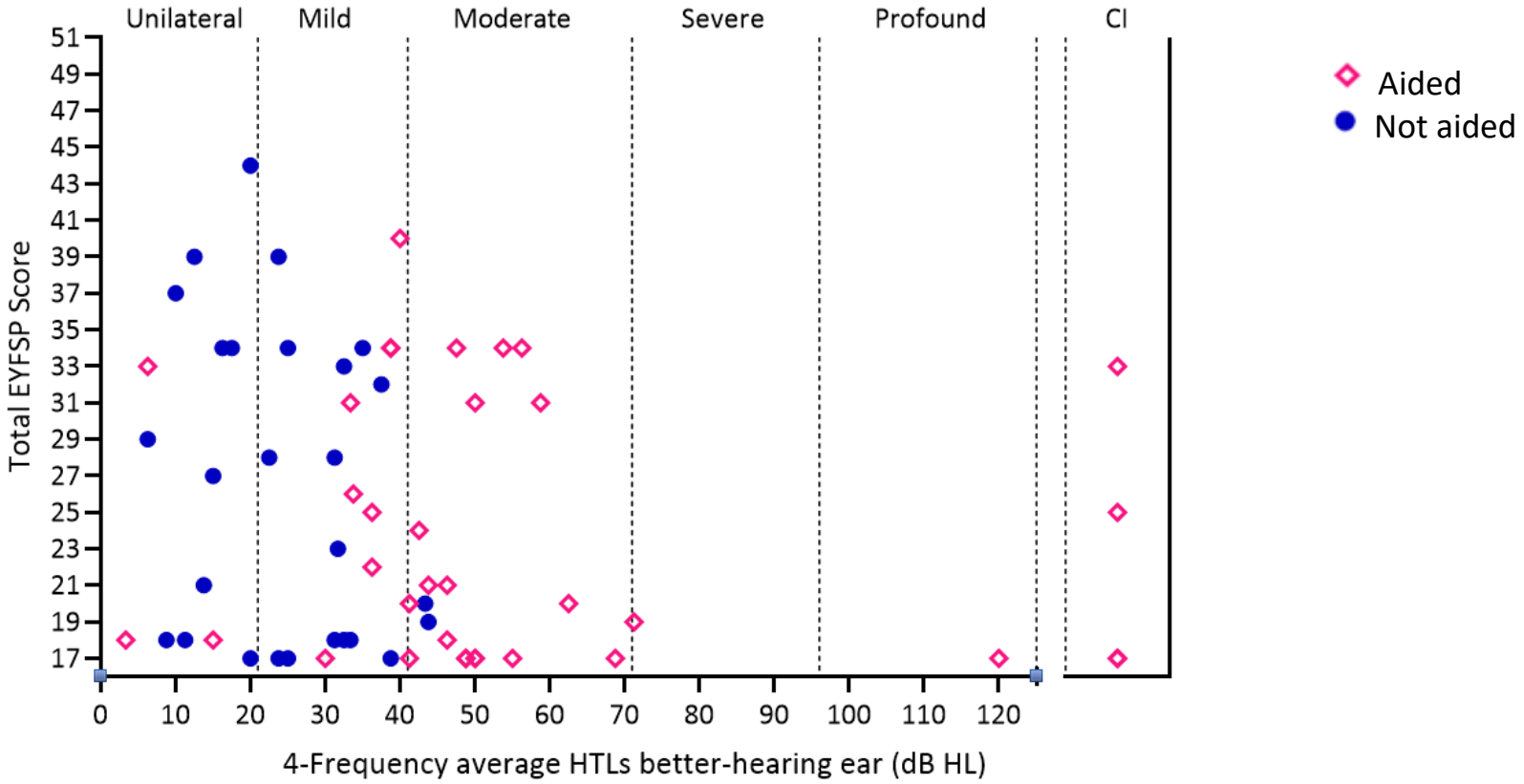


Figure 6.5: Total EYFSP scores by degree of hearing loss
EYFSP = Early Years Foundation Stage Profile; HTLs = Hearing threshold levels; CI = cochlear implant. Degree of hearing loss category by the British Society of Audiology classification system, or CI use, are shown by vertical dashed lines and headings. Open pink diamonds represent individual children who were aided at their EYFSP assessment and dark blue circles show individual children who were not aided. If more than one child had the same 4-frequency average HTL and EYFSP score, or used a CI with the same score, their symbols are superimposed.

Despite these limitations, the study group contained sizeable groups of children with unilateral, mild, or moderate hearing loss, revealing the potential to investigate the combined effect of hearing loss being managed via standard NHS hearing device provision on early educational outcomes, controlling for potentially confounding variables. The next section therefore achieves the objective of measuring the effect of hearing loss on children's early educational outcomes by comparing the study and control groups.

6.3.5: Total EYFSP scores for the study and control groups

The study and control groups will be compared in terms of both their total EYFSP scores and the proportion gaining a GLD. Comparisons will first be made for the study and control groups as a whole and then for the sub-groups with unilateral, mild, or moderate hearing loss. Figure 6.6 shows the distribution of total EYFSP scores for the study group compared to the group of matched controls.

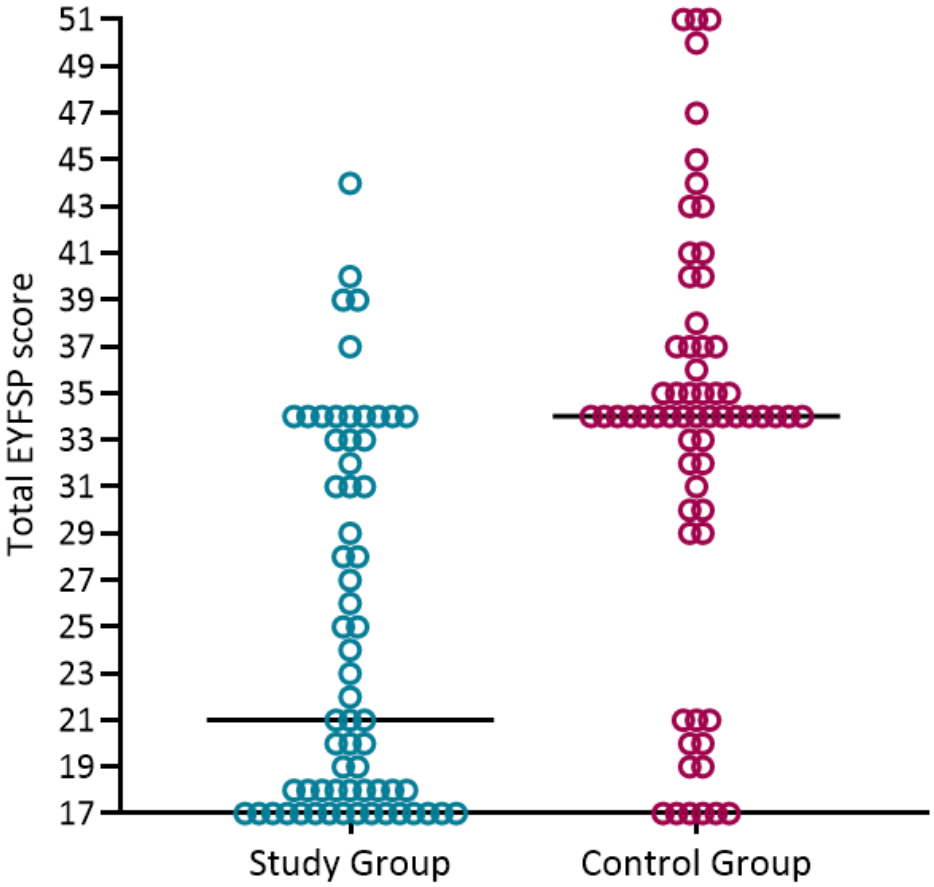


Figure 6.6 Distributions of study and control group total EYFSP scores
EYFSP = Early Years Foundation Stage Profile. Individual children's scores are represented by open circles, and the median group scores by solid horizontal lines. Each circle represents one unique case.

The study group had a mean EYFSP score of 24.7 (standard deviation = 7.88) and the control group mean score was 33.1 (standard deviation = 8.94). A dependent t-test was run to determine whether there was a significant difference between the matched pairs of study and control group participants. The difference in mean scores was 8.41 (95% confidence interval of the difference = 5.69 to 11.12). This difference was statistically significant ($t(63) = -6.18$, $p < .001$).

Next, the effect of degree of hearing loss was explored by comparing total EYFSP scores for the sub-groups of children with unilateral, mild, or moderate hearing loss. For sub-groups, the statistical significance of the differences between median scores was tested for via two-tailed Mann-Whitney U tests due to smaller sample sizes coupled with non-normal score distributions. Results are shown in Table 6.7. Statistically significant results are shown in bold.

Table 6.7: Total EYFSP score distributions for the study and control groups by hearing loss category

Hearing loss group	Study Group median score	Control Group median score	U test statistic	z score	Statistical significance
Unilateral N = 15 Study N = 15 Control	27.0	34.0	72.0	-1.66	$p = .097$
Mild N = 23 Study N = 23 Control	**26.0**	**34.0**	**147.0**	**-2.57**	$\mathbf{p = .010}$
Moderate N = 20 Study N = 20 Control	**22.3**	**34.5**	**77.5**	**-3.30**	$\mathbf{p < .001}$

There was a statistically significant difference between group median scores for the mild and moderate hearing loss groups compared to control groups, but the difference did not reach statistical significance for children with unilateral hearing loss.

6.3.6. Proportions of children from the study and control groups attaining a Good Level of Development

The GLD was attained by 17 (27%) of the study group and 46 (72%) of the control group. Pearson's chi-square test of independence was performed to examine the relation between children being in the study or control group and attaining a GLD. The relation between these variables was significant, ($X^2(1)$, N = 128, 26.29, $p < .001$).

The proportions of children within the unilateral, mild, or moderate hearing loss groups who attained a GLD were also compared to their matched controls via Pearson's Chi-Square tests of independence. Results are shown in Table 6.8. Statistically significant results are shown in bold. Each comparison met the test assumption of at least five children being expected to fall into each category. There was a significant relationship between attaining a GLD and being in the study or control groups for every hearing loss group.

Table 6.8: Proportions of children attaining a Good Level of Development from the study and control groups by hearing loss category

Hearing loss group	Study Group % with GLD	Control Group % with GLD	Pearson Chi-square value	Statistical significance
Unilateral N = 15 Study N = 15 Control	**33.3**	**86.7**	**8.89**	***p* = .003**
Mild N = 23 Study N = 23 Control	**30.4**	**65.2**	**5.58**	***p* = .018**
Moderate N = 20 Study N = 20 Control	**20.0**	**70.0**	**10.10**	***p* = .001**

6.4. DISCUSSION

This chapter measured the impact of hearing loss on the early years educational outcomes of children screened for the CUSP study who were enrolled in the BiB cohort. Hearing loss was associated with a lower median total score equivalent to dropping an attainment category across 13 of the 17 learning items, and a lower probability of gaining a good level of development, controlling for potentially confounding variables.

6.4.1. Linking data to establish a study group

A study group of 64 children were identified by linking CUSP eligibility screening data, supplemented with older audiology records, to BiB cohort data. This group was characterised audiologically and by characteristics known from the literature to be significantly associated with EYFSP scores.

6.4.2. Study group characteristics

A strength of the overall BiB study design, using routine educational and clinical data rather than relying on research visits, is that this chapter represents a group of children at risk of being under represented in the literature. The children in this chapter were similar to the overall BiB cohort in that around one quarter were in receipt of free school meals, and the largest ethnic group was children of Pakistani heritage (Wright et al., 2019). This is in contrast to the Avon Longitudinal Study publication on the impact of recurrent conductive hearing loss on children's IQ, in which 94% of children were White, and non-

English speakers were excluded due to the nature of the assessment tools (Hall et al., 2014). It was not known how many children in the present study did not speak fluent English, but English was an additional language in the home for half of them. The proportion of mothers with higher than GCSE qualification was similar between the present study and that of Hall et al., (2014). In terms of socio-economic status, Table 6.2 showed both the relative poverty of Bradford compared to the rest of the nation, and also that at least half of the study group were from the most deprived fifth of the country's population. While Bradford is not socio-economically representative of the country as a whole, getting representative samples of children from deprived families in hearing research has been noted to be a challenge (Tomblin et al., 2020). The challenge is important to overcome, since lower socioeconomic status is associated with poorer oral language and educational outcomes (Bradley & Corwyn, 2002) and a less-rich home language environment (Piot et al., 2022), while better quality cognitive stimulation at home can moderate the effect of conductive hearing loss on children's development (Hall et al., 2014). This study therefore fills an important gap in the literature by including children from relatively deprived families including those of Pakistani ethnicity.

The study group included children with conductive, mixed, and sensorineural hearing losses. There were too few children with severe or profound hearing loss, or using CIs, to allow statistical comparison of the outcomes of these groups. There was a trend for more children to be fitted with hearing devices with increasing degree of hearing loss of the better-hearing ear. This meant that measuring the impact of being aided versus not being aided was not possible with the present dataset. However, this retrospective study still provides unique

insight into the impact of hearing loss on early years foundation stage profile outcomes of children receiving standard NHS care for their hearing loss.

6.4.3. Case control matching

Because of the size of the BiB cohort, it was possible to create a control group of cases who were matched for most of the variables known to influence EYFSP outcomes. Maternal education data was not available for a minority of participants, limiting the degree to which this could be controlled for. Following data linkage, hospital numbers were only available for the children who had been screened for the CUSP study, not the rest of the BiB cohort. Therefore, it was not possible to check if any control group participants had a history of otological problems or documented hearing loss from their hospital records. For example, some children in the control group likely experienced otitis media with effusion during their Reception year (Hall et al., 2014), meaning that the study and control group comparisons may represent an underestimate of the effect of hearing loss on EFYSP scores. It was not possible to match for variables that were not available within the BiB dataset, such as the presence of autism and children's non-verbal intelligence. However, despite these limitations, the overall size of the BiB cohort enabled the creation of a control group that were well matched for characteristics previously established to influence EYFSP scores including ethnicity, gestational age, academic term of birth, speaking English as an additional language, being in receipt of free school meals, and the presence of additional needs (Cotzias and Whitehorn, 2013).

One limitation is that the data on additional needs for the study group, based on eligibility screening results, was known from Chapter 5 to be an underestimate of the prevalence of any additional needs and so some additional children from the study group might have been better matched to controls with SEN support. It was not possible to gauge the type and severity of additional needs experienced by control group children with SEN support, and so these could have been more or less likely to influence EYFSP scores than the additional needs experienced by their study group matches.

Last, while not formally matched for, data for the study and control groups were collected over the same time period, 2013 to 2016. This means the estimate of the impact of hearing loss on children's scores was not confounded by changes in the total EYFSP scores or proportion of children gaining a GLD over this time period due to changes in teaching practice (Noble, 2016).

6.4.4. EYFSP outcomes compared to national data

In the present study, 26% of children in the study group achieved a GLD in contrast to 41% of the cohort assessed in the 2012 pilot study (Cotzias & Whitehorn, 2013). This low pass rate is even more striking when compared to government data from the data collection period of this study. Nationally, the proportion of children achieving a GLD was 52% in 2013, progressively increasing to 69% by 2016 (Noble, 2016). The random selection method used in creating the control group led to both the control and study groups including children with assessments distributed across 2013 to 2016.

The control group pass rate was 72%. Nationally, the gender gap noted by Cotzias & Whitehorn, (2013) persisted throughout the duration of data

collection, with girls scoring higher than boys across all 17 items (Noble, 2016). In 2016, 77% of girls achieved a GLD compared to 62% of boys (Noble, 2016). The greater proportion of girls compared to boys in the present study may therefore go some way to explain why a greater proportion of children in the control group achieved a GLD compared to the national average. These trends highlight the importance of using matched controls to estimate the impact of hearing loss on children's outcomes. This will be discussed in the next section.

6.4.5. The impact of hearing loss on children's EYFSP scores

The median score of the control group was 34, reflecting the national modal score throughout 2013 to 2016. In contrast, the study group (children who were using hearing devices in 2019) had a median score of 21. This is equivalent to the difference between a child meeting the 'Expected' level on all 17 items, and a child only meeting the 'Expected' level on 4 items and being classed as 'Emerging' for the remaining 13.

In terms of the GLD, not only did the study group have a significantly lower pass rate overall, but so did each of the sub-groups of children with unilateral, mild, or moderate hearing loss. This suggests that standard NHS audiology and educational practice combined during 2013 to 2016 was not sufficient for these children to access the curriculum and develop as they could have done in the absence of hearing loss.

Hearing devices had been provided to all children with 4-frequency average HTLs greater than 45 dB HL. However, provision of devices was less consistent for those with HTLs ≤45 dB HL. The nature of study group

eligibility in this retrospective study meant that it is known that all of these children would later be fitted with some kind of hearing device. The results of this chapter show that the largely unaided children with unilateral and mild losses had fallen behind their hearing peers prior to first device fitting. This suggests that hearing device intervention could be considered much sooner following the identification of hearing loss, even for those with milder losses, as evidence is emerging for their efficacy in these groups (Tomblin et al., 2020). While it was not possible to determine the exact type of loss for all participants, the study group included children with conductive, mixed and sensorineural losses. It is likely that the need to intervene earlier applies across these groups, and that any appropriate device or surgical options that improve access to speech sounds would be expected help close the gap to the developmental progress demonstrated by children in the control group.

It is also concerning that the children with moderate losses, who were mostly aided, had also fallen behind their hearing peers, despite HAs being known to moderate the effects of hearing loss in this group (Tomblin et al., 2020). This could suggest that earlier provision of devices was needed, and/or that more work is needed to ensure that they have access to the appropriate educational support services. These could include frequency modulation remote microphone systems, soundfield systems, preferential seating arrangements, communication strategies and extra educational support (British Academy of Audiology Service Quality Committee, 2022; National Deaf Children's Society, 2015). Further research is needed to establish whether fitting practices for children with unilateral and mild losses have changed over time, and to

examine the educational support that is available for children with hearing losses of all types and degrees.

6.4.6. Summary of potential limitations and biases

One limitation of this study was that the factor(s) contributing to children being classed as needing SEN support or ECHP support were not known. This limited the extent to which additional needs could be controlled for between the study and control groups. In term of bias, the population of Bradford is known to be of lower socio-economic status on average than the nation as whole, as seen in Table 6.5. This limits the generalisability of this chapter's findings to the wider population. However, since it is plausible that children from the most deprived backgrounds are more likely to have a strong association between EYFSP scores and later academic attainment (Atkinson et al., 2022; Treadaway, 2019) this does not diminish the value of this work.

6.5. SUMMARY

This chapter measured the impact of hearing loss on the early years' educational outcomes of children screened for the CUSP study and enrolled in the Born in Bradford cohort study. Unilateral or bilateral hearing loss, managed via routine NHS and educational care, was associated with significantly poorer educational outcomes by the end of children's first year of school for children in the Born in Bradford cohort completing their Reception year in 2013 to 2016. The implications of this, along with the results of the earlier chapters, will be discussed in Chapter 7.

CHAPTER 7: GENERAL DISCUSSION AND RECOMMENDATIONS FOR FUTURE RESEARCH

This thesis aimed to add to the evidence base to guide clinicians when choosing whether to offer HAs or CIs to children with hearing loss within or close to the 2019 NICE guidance (TAG566). Chapter 2 catalogued previously published studies of comparative outcomes for children with severe hearing loss using HAs to children using CIs. Chapter 3 described the plans originally devised to fill the gaps in the evidence base, along with adaptions made due to the COVID-19 pandemic. Chapter 4 characterised complete caseloads of children without access to cochlear implantation within the NHS. Chapter 5 compared caregiver reported functional listening, tinnitus, and need for recovery from listening outcomes for children using CIs or children close to or within CI candidacy using HAs, controlling for potentially confounding variables. Chapter 6 measured the impact of hearing loss on the early years educational outcomes of children in the Born in Bradford cohort study. This chapter will synthesise and discuss the findings of the thesis in terms of their implications for children's hearing care and for future research.

7.1. PROPERTIES AND VALIDITY OF KEY OUTCOME MEASURE

The key finding toward the main thesis aim was that all sub-groups of children with HAs scored significantly lower on the PEACH questionnaire than the group using CIs, except for those confirmed to fall outside the audiometric criterion of TAG566 (NICE, 2019). In behavioural tests of speech perception, such as the one initially planned as the primary outcome measure for this

study, the acoustic environment and test stimuli can be tightly controlled. This is beneficial when comparing performance between groups in a standardised way. However, it is not possible to replicate real-world listening scenarios in the clinic. The conceptual validity of the PEACH lies in its measurement of children's real-life behaviours in real-life settings. As a parent-report tool, its use allowed the inclusion of children with additional difficulties, making the participants more representative of the population than behavioural studies that need to exclude younger children and those with developmental delay. Further, PEACH scores are known to correlate with functional hearing outcomes such as degree of hearing loss (Bagatto et al., 2011; Marnane et al., 2015) and standardised language measures obtained in behavioural assessments of children (Ching et al., 2019). In summary, the key outcome measure of this thesis permitted a meaningful comparison of children's performance in situations where hearing devices are most needed, including a clinically representative population.

7.2. IMPLICATIONS OF FINDINGS FOR NHS AND EDUCATION SERVICES

Chapter 4 revealed that children's characteristics varied as a function of their eligibility for CIs under TAG566 (2019). Hence certain characteristics could present barriers to children accessing cochlear implantation within the NHS. First, missing audiometric data was associated with having unknown eligibility. The implications of this for children's management will be discussed, and potential solutions proposed, informed by the findings of both Chapters 4 and 5. Other characteristics included families' need for interpreters

and children's additional needs. These will be discussed in terms of their clinical implications. The COVID-19 pandemic has had far-reaching consequences for the delivery of healthcare services across the NHS and evidence for the consequences of this related to findings from Chapter 4 will be discussed. The asymmetrical hearing losses of one quarter of the children newly eligible for CIs identified in Chapter 4 will be discussed in terms of spatial listening rehabilitation needs. Last, the implications of Chapter 6 will be discussed with reference to current guidance on the management of children with any degree of hearing loss, and the importance of minimising the effect of this on their educational outcomes.

7.2.1. Missing audiometric thresholds as a barrier to accessing cochlear implantation and options for changing clinical practice

This study identified that TAG566 (NICE, 2019) increased the number of children for whom audiometric eligibility for CIs was unknown. The increased uncertainty was due to the greater number of HTLs that must be measured to establish eligibility definitively under TAG566 (NICE, 2019) compared with TAG166 (NICE, 2009). The children whose audiometric eligibility was unknown did not meet the current BCIG guidance for referral for CI candidacy assessment, because this is the same as TAG566 (NICE, 2019). Chapter 5 confirmed that, controlling for the presence of additional needs and other confounding variables, the caregiver-reported functional listening outcomes of children whose CI eligibility was unknown were significantly poorer than those of children using CIs. This finding indicates that they may represent a group that could benefit from cochlear implantation.

As described in Chapter 1, the audiometric criterion from TAG566 (NICE, 2019) is used as both the trigger for HA departments to refer children for CI eligibility assessment at CI services, and also within CI services to establish whether a child is eligible for cochlear implantation via the NHS without a special case for funding being made to commissioners. This section will focus on the implications of the findings of Chapter 4 on the first stage, referral for CI eligibility assessments, and will present two alternative approaches to ensuring that all children who are eligible for a CI eligibility assessment can receive one.

Option 1: Increase the number of HTLs measured at HA appointments

One approach to ensuring that as many potentially eligible children as possible are referred for CI eligibility assessment would be to advise HA services to measure a full set of HTLs, including 3 kHz, for all children using HAs where there is any possibility that they might meet NICE guidance for CIs. In this scenario, the BCIG referral guidance would remain as it is now, the same as the NICE audiometric eligibility criterion. A potential advantage of this approach is that children's audiometric CI eligibility would continue to be monitored at the family's local department, integrated with their existing routine care. This would limit the travel and childcare costs incurred by families.

However, because it is not always possible to obtain a full set of HTLs during one visit for younger children or those with learning difficulties (Wood, 1993), it might still be necessary for local HA services to see children on more occasions than they presently do, increasing families' frequency of travel, time

off work, childcare for siblings, and time off school. Another potential disadvantage of this approach is the extra time that would need to be spent on audiometric assessments at each HA review. This would either require longer appointments or take away from time that would otherwise be spent on HA adjustments, functional outcome assessment and the conversations around child and parent observations about the child's hearing ability, choice of communication mode, and individual management plans required by the British Academy of Audiology Quality Standards in Paediatric Audiology (British Academy of Audiology Service Quality Committee, 2022). Chapter 4 established that only one quarter of children using HAs had HTLs measured at 3 kHz when their two most-recent audiograms were considered together, suggesting that the measurement of all ten relevant HTLs was not considered a clinical priority by most audiologists, at least at the time of screening. Adopting this change in practice would require re-training and new clinical protocols at all children's HA departments. The most recent National Deaf Children's Society (NDCS) report on children's hearing services in England requested information from all 123 HA services in England, with a response rate of 87% (National Deaf Children's Society, 2021a). Of services that participated, most reported that their staff were able to access all continuous professional training needed for their roles. However, financial limitations prevented 4% of services from allowing staff to access necessary training and 11% were unable to adequately train their staff for other reasons (National Deaf Children's Society, 2021a). Also, it cannot be assumed that access to training was similar for the 13% of services who were approached but did not participate in the NDCS report.

Aside from access to training, there was an overall decrease in full-time equivalent paediatric audiology staff of 8% across England between 2019 and 2021 (National Deaf Children's Society, 2021a). The decrease in staffing levels varied geographically. The East Midlands was least affected, with only one service (14%) having decreased staffing. In contrast, the proportion of departments with reduced staff was 25% in London, 36% in the South West, 40% to 53% in East England, the North East, North West and South East, 67% in the West Midlands and 80% in Yorkshire and the Humber. This may reduce the capacity of services that are also recovering from the impact of the COVID pandemic to offer longer and/or more frequent HA reviews to the children whose NICE eligibility is not yet known.

If issues of access to training and /or inadequate staffing led to variable levels of adherence to a new testing protocol, this could produce a "postcode lottery" regarding which children had enough HTLs measured by HA services to establish if they should be referred for CI candidacy assessment. Considering also the potential impact of this guidance change on how paediatric audiologists allocate their time during clinical appointments, it would be essential to consult with HA service providers to establish the feasibility of implementing this option.

Option 2: Change the BCIG referral guidance

An alternative approach could be for the BCIG referral guidance to change, to include either a lower decibel limit and/or fewer HTLs to be known before referral for CI eligibility assessment was advised. This could be implemented without a change to the NICE audiometric candidacy guidance for

implantation. The CI centres would receive a greater number of referrals for eligibility assessments and would remain responsible for assessing whether children meet TAG566 (2019).

One potential advantage of this method is that local HA services would not need to change the way they manage children during HA appointments. Their training need would be awareness of the revised referral guidance rather than a protocol change that asked them to assess additional HTLs not clinically needed for immediate HA management. This might help the change to be more broadly and consistently adopted. A further benefit for local audiology services would be that less additional time was needed during HA clinics to establish whether a child warranted referral for CI assessment.

However, in this scenario an increased workload would be generated for the CI services, which could be perceived as a disadvantage to them. They would receive more referrals and need to carry out more eligibility assessments, requiring an increase in staff time and resource provision. A greater proportion of CI assessment appointments would likely result in discharge back to local audiology departments, making the process arguably less efficient. CI candidacy assessments take place at the 16 regional children's CI services across the UK (British Cochlear Implant Group website, accessed 28/09/2022) (12 centres within England, for ease of comparison to the 123 English HA services identified by the NDCS). No additional training or protocol changes would be needed at the CI services, since they would continue to implement the assessment methods already used, against the existing NICE guidance.

One potential down-side of this option for families could arise because there are far fewer CI than HA services, so families would need to travel further on

average to have their child's eligibility confirmed at a CI centre. This would involve a greater time and financial burden in comparison to visiting a closer, local HA centre. These burdens are known to present barriers to some families in accessing hearing services for their children (Noblitt et al., 2018) and may be seen as unacceptable for some, especially when children were found to be ineligible for CIs. However, the detriment caused by this decrease in the specificity of CI referral guidance (more children who do not need CIs would be referred) could be offset in terms of increased sensitivity, as a greater proportion of children who would hear better via CIs than HAs could be identified via this option.

Regardless of the outcome of each eligibility assessment at a CI centre, families might benefit from the specialist expertise in severe to profound deafness available at CI centres including specialist speech and language therapists, audiologists, teachers of the deaf, and surgeons (Martin & Raine, 2013) that they might not otherwise be able to access. Children for whom cochlear implantation was not immediately recommended could nevertheless have a comprehensive baseline assessment of their speech perception and language development to act as a benchmark for any future deterioration in their hearing (Warner-Czyz et al., 2022).

As with Option 1, the implications of Option 2 for staffing and resources at CI services means that it would be imperative for a national conversation and feasibility assessment to take place, including both CI and HA services and parents of children with hearing loss, before this possible approach was implemented.

Another important question that would need to be answered before implementing Option 2 would be deciding specifically what the revised referral guidance should be. The ACIA recommend unaided HTLs of >70 dB HL (Warner-Czyz et al., 2022) across an intentionally unspecified range of audiometric frequencies to allow for clinician judgment (Warner-Czyz, personal communication, 2022). Hypothetical options within the NHS could include only requiring HTLs routinely assessed at HA appointments, that fewer HTLs need be known, and possibly a lower decibel limit. For example at least one, rather than two, HTLs out of 0.5, 1, 2 or 4 kHz be ≥80 dB HL, or that any two HTLs out of 0.5, 1, 2 or 4 kHz should be ≥70 or 75 dB HL, or that HTLs average ≥70 dB HL across a range of frequencies. It is beyond the scope of this thesis to make a specific recommendation. Consultation with parents and professionals would help to clarify what degree of widening referral guidance compared to eligibility guidance would result in an acceptable trade-off between sensitivity and specificity within the context of the NHS, and what number of and which HTLs should be included. The data from Chapter 4 could support this process as it shows that this group had missing HTLs across the frequency range, but that data was most commonly available at 1 and 4 kHz (Figure 4.14). Once consensus was reached, a potential future use of the data collected in Chapter 4 could be to estimate the number of increased referrals that could be expected as a result of the change and to characterise the children affected.

7.2.2. Ethnicity, language, and socio-economic factors as barriers to accessing cochlear implantation

There are different possible explanations for why children using HAs who had previously been eligible for CIs included such a large proportion of families who needed spoken language interpreters. The UK receives immigrants from many countries and communities. The CRIDE report identified 206 newly arrived deaf children in England over its 12-month period, accounting for 0.5% of the adjusted total number of children, the same as the proportion of newly arrived children during 2019 (CRIDE, 2021). The NHS offers universal neonatal hearing screening to children born in the UK. This is known to facilitate early identification and intervention via HAs or CIs, leading to better oral/aural outcomes (Yoshinaga-Itano et al., 2018). If a child with congenital hearing loss is born in a country without access to early diagnosis or intervention, they might experience a long duration of severe-to-profound hearing loss before reaching the care of NHS services. This means they are likely to gain less benefit from either HAs or CIs than children without prolonged auditory deprivation (Ching et al., 2017; Nicholas & Geers, 2013; Yoshinaga-Itano et al., 2018). Congenitally deaf children who receive a CI above the age of 4 years are more likely to reject the device and become non-users (Archbold et al., 2009). These factors might contribute to the decision of a CI team to withhold cochlear implantation, led by the principles of the ChIP (Hellman et al., 1991), leaving the family no alternative but for their child to rely on HAs. However, it was not possible to determine country of birth from the eligibility screening forms and so the proportion of children born outside the UK was not known.

An alternative explanation for the high need for spoken language interpreters in this group is that there were disparities in children's access to CIs related to family ethnicity and/or English language proficiency for children born in the UK. A lack of equity in access to CIs associated with ethnicity has been reported in the USA for both adults and children (Stern et al., 2005; Tolisano et al., 2020). The most used spoken language category within this group was Eastern European, including Czech, Hungarian, and Slovak. The specific ethnic background of these families cannot be known from the data in this study. However, data from Swanwick et al., (2021) suggest that this cohort included some children from Roma communities, with 41 documented deaf or hard-of-hearing Roma children known to the local authority in Bradford. In total, 44 children from the Bradford caseload in the present study needed any kind of Eastern European interpreter. Roma families are known to experience barriers to accessing audiology and deaf education services and may have attitudes toward deafness that make them less likely to seek intervention (Swanwick et al., 2021). This could have contributed in part to children of families needing Eastern European interpreting making up 16% of this group. The findings highlight a need for children's hearing services to include family liaison officers with foreign language skills and cultural awareness of ethnic groups currently with lower uptake of CIs. This would help ensure that lower uptake is occurring through informed choice rather than comparative lack of access to evidence-based information and/or poorer awareness of educational services (National Deaf Children's Society, 2021b; Swanwick et al., 2018).

7.2.3. Additional needs as a barrier to accessing cochlear implantation

Chapter 4 revealed that the children using HA whose audiometric-eligibility for CIs is unknown under TAG566 (National Institute for Health and Care Excellence (NICE, 2019) has a disproportionately high number of children with additional needs compared to the wider population of HA users. There is evidence that CI outcomes for children with additional disabilities can be very variable, and some do not display benefits that are measurable via the speech perception and spoken language outcomes routinely used for their peers without additional needs (Mesallam et al., 2019). The risk of poorer outcomes might increase with the severity of the additional disabilities, for example global developmental delay (Edwards, 2007). However some children with additional disabilities can benefit from CIs (Rawes et al., 2021) and so it is important that these children receive equitable access to CI eligibility assessments, even if they are not all found to be suitable candidates once their unique circumstances are assessed by the multidisciplinary team. Chapter 4 indicated that children with additional disabilities are currently at risk of being missed from CI eligibility assessment referrals. Changes to the referral guidance as previously described might help to mitigate this risk.

7.2.4. The consequences of the COVID-19 pandemic as a barrier to accessing cochlear implantation

The latest BCIG Annual Data Collection report (Cullington, 2022) reveals interesting trends in the number of children receiving CIs over time. TAG566 (NICE, 2019) was initiated from spring 2019. The BCIG data collection reports run from 1st April to 31st March each year. As confirmed by the results

of Chapter 4, TAG566 (NICE, 2019) would be expected to lead to an increase in new paediatric CI surgeries that would be seen from the 2019-2020 report onward. Under normal circumstances, this might have begun with a large, transient increase as the cohort of children newly falling within criteria were referred, assessed, and implanted as appropriate, followed by a smaller but ongoing increase compared to before, reflecting the larger proportion of children with hearing loss now eligible for CIs. Considering both unilateral and bilateral surgeries, numbers of new paediatric CI recipients climbed from 2013 onward, peaking for the 2018-2019 report. Despite TAG566 (NICE, 2019), slightly fewer children were implanted during 2019-2020 than the year before (Cullington, 2022).

On one hand, it is extremely encouraging that surgeries continued to go ahead in similar numbers, reflecting the commitment and innovation of NHS CI services in delivering this time-sensitive intervention despite significant challenges (e.g. Lawrence et al., 2020; Mohammed et al., 2021). However, the 2020-2021 and 2021-2022 reports reveal a subsequent drop in the overall number of new CI recipients, with numbers more similar to those from 2014-2015 (Cullington, 2022). This suggests there might be both a backlog of children newly eligible for CIs who have not yet received them, and an accumulating backlog of traditional candidates who are not being implanted at the same rate as prior to the pandemic.

The exact causes for the declining implantation rates cannot be determined from the currently available data. The BCIG report does not include information on what proportion of new recipients were children who became newly eligible due to TAG566 (NICE, 2019) and so it is not known whether

this is associated with children's odds of receiving CIs. It is also not known whether the fall in implantation rate is due to lower referral rates from HA centres, and / or from reduced capacity at CI services. Either scenario is plausible, as the long-term effects of the pandemic remain significant for both staff and services across the organisation (Anderson et al., 2021; Gemine et al., 2021; Iacobucci, 2022).

7.2.5. Maximising the spatial listening via CIs of children newly eligible for cochlear implantation

The first NICE guidance for cochlear implant candidacy TAG166 (NICE, 2009) permitted children to receive a cochlear implant for both ears on the NHS. This followed growing evidence, from both adults and then children, that bilateral implantation could improve spatial listening skills such as sound-source localisation and speech perception in background noise (Ching et al., 2007; Litovsky et al., 2006; Muller et al., 2002; Verschuur et al., 2005). It became apparent that longer-duration auditory deprivation in the previously non-implanted ear was associated with poorer spatial listening outcomes following the sequential provision of the second CI to congenitally-deaf children (Chadha et al., 2011; Killan et al., 2019) because asymmetric hearing during critical periods of childhood leads to long-term impaired central representation of the poorer-hearing ear, known as aural preference syndrome (Anderson et al., 2022; Gordon et al., 2015).

Chapter 4 revealed that around one quarter of the children who fell outside TAG166 (NICE, 2009) but were confirmed to be within TAG566 (NICE, 2019) had asymmetric hearing loss. Should these children now progress to

simultaneous bilateral cochlear implantation, they are vulnerable to gaining poorer outcomes from their worse-hearing ear than their other ear. They are consequently likely to have worse long-term binaural speech perception and spatial listening skills than if NICE had permitted earlier unilateral cochlear implantation of their worse-hearing ear, with the option of subsequent sequential implantation of the other ear. Rehabilitation services need to be prepared to support these children with the kind of listening exercises used in the past for children transitioning from unilateral to sequential bilateral CI use, such as listening via the worse-ear alone for periods of time each day, and practising locating the sources of sound. These have been anecdotally reported to have some success, however little evidence is available to confirm their efficacy. Ongoing research is aiming to formally develop and validate spatial listening training for children and teenagers (Vickers et al., 2021). To limit the number of children experiencing preventable asymmetric auditory deprivation in the future would require NICE to revisit its decision not to allow sequential implantations.

7.2.6. The role of hearing interventions in children's early educational outcomes

Chapter 6 found that hearing loss, managed via standard NHS and educational support, was associated with significantly poorer educational outcomes for children during their first year of school. The cohort included children with unilateral and bilateral, sensorineural, mixed, and conductive losses ranging from mild to profound.

The British Society of Audiology has guidance on the management of children whose hearing loss is identified via neonatal hearing screening (British Society of Audiology, 2021). However, the document does not stipulate what the timing of device provision should be, nor specify any particular type of device, for any clinical scenarios including type, symmetry, permanence or degree of hearing loss. Rather, the guidance emphasises the importance of individual management plans devised in consultation with parents and multi-disciplinary teams. Similar to guidance on surgical intervention for conductive hearing loss due to middle ear effusion (NICE, 2008), a monitoring period during which no intervention is used is included among management options. The guidance states:

> *"Options include ongoing audiological assessment and monitoring, provision of amplification and referral to early intervention services. The actual management approach will depend upon the clinical findings including the likely degree and type of hearing loss, the developmental status of the baby including the existence of other disabilities and the views and wishes of the parent"*
>
> (British Society of Audiology, 2021, p24).

Within a child's management plan, audiologists are advised to ensure "Provision of amplification where appropriate". This non-specific approach may be due to the wide variety of clinical and family scenarios that can arise. However it also leaves open a risk of inconsistent service provision between audiology departments, dependent on the awareness and interpretation of the literature by individual teams. Poor outcomes for children with acquired hearing loss and geographical variation in referral to assessment time were flagged in the Action Plan on Hearing Loss (NHS England and Department of

Health, 2015), indicating that children do not currently receive consistent or timely care across the country.

For children with conductive hearing loss, uncertainty remains around the best management strategy. A Cochrane review of surgical intervention proposed increasing the period of "watchful waiting" prior to surgery from 3 to 6 months (Browning et al., 2010). The findings of Chapter 6 suggest that caution should be used in implementing this without the provision of some form of hearing device, due to the risk of poorer educational outcomes arising while the child is waiting. In terms of hearing devices, the Cochrane review suggested issuing HAs as an alternative to either surgery or watchful waiting. Another alternative is bone conduction aids. Innovative ways to provide bone conduction sound to children via relatively cheap headsets are under preliminary development in the UK (Holland Brown et al., 2019; Holland Brown et al., 2021) and more costly but validated clinical devices are also available that are reported to be both acceptable to children and effective in improving speech perception (Favoreel et al., 2020).

7.3. DIRECTIONS FOR FUTURE RESEARCH

7.3.1. Further comparative outcomes for children with and without access to cochlear implantation

The PEACH questionnaire is a measure of caregiver-reported functional oral/aural skills. It was a good fit for this project because it was applicable to children with a broad range of ages, degree of hearing loss, and developmental abilities. However, it cannot be expected to reveal other more complex and equally important differences between HA and CI users. These include many

of the outcome gaps in the knowledge base identified in Chapter 2, including pragmatic language skills, speech perception in complex background noise, and spatial listening. Further, a validated tool is now available to measure children's listening fatigue via caregiver- or self-report (Hornsby et al., 2022) and child-reported tinnitus outcomes would provide a more accurate measure of any differences in children's experience of tinnitus between the groups (Raj-Koziak et al., 2011). One important area for future research therefore remains in-person behavioural assessments of children's complex listening skills, and child-reported outcomes, as initially planned for this project prior to the pandemic. Data such as these would be needed in addition to this project's findings to more fully validate the existing guidance or to make decisions regarding future revisions to CI candidacy criteria. Future research with a sample including more families who use English as an additional language would be valuable, especially as comparison of the need for interpreters between the survey participants and wider eligible population showed that these families were underrepresented in the present study.

The gold standard method for comparing the interventions would be a randomised controlled trial of cochlear implantation versus HAs (Munro et al., 2021). Based on the available evidence, Chapter 5 found no significant difference between functional listening outcomes for children outside the current NICE guidance using HAs and children using CIs. Now that CIs are available via the NHS to children with at least two HTLs of 80 dB HL or greater at two frequencies bilaterally, it would be ethically questionable to withhold CIs from children meeting this criterion in order to create a control group of HA users. The increased risks associated with CIs compared to HAs

(Loundon et al., 2010; Scheperle et al., 2017) also present a challenge for any clinical trial randomising children to CIs or HAs. Further research would be needed to establish the ethical acceptability and feasibility of such trials.

7.3.2. Audit of the management of children with HAs eligible for CIs and whose eligibility is not known

The timing of this study, being conducted while a change in NICE guidance for cochlear implantation occurred, meant that it was possible to identify children using HAs who had recently become audiometrically eligible for cochlear implantation but had not yet been offered CIs. It is not known how many of these children have since, or will in the future, access CI eligibility assessments or cochlear implantation. As discussed in section 7.1.4, there is evidence, including that provided in this thesis, to suggest that not all audiometrically-eligible children are currently accessing CIs. It would be informative to revisit the HA caseloads screened for this study and identify which of the eligible, and possibly eligible, children have since been referred for CI candidacy assessment. Characterising those who have been referred and those who have not might help to identify barriers to referral. It could also identify any changes in local audiometry practice such as increasing the number of HTLs measured per child at HA appointments, that might have been initiated independently by local audiology staff in response to TAG566 (NICE, 2019). Understanding whether services have already adapted their HA management protocols to TAG566 (NICE, 2019) would help to inform which national strategies would be suitable or effective, as discussed in section 7.1.1.

Comprehensive identification of all barriers to cochlear implantation could be carried out by auditing the whole pathway for children referred for CI eligibility assessment due to TAG566 (NICE, 2019). Data extracted could include how many families have had conversations with their local audiology staff about onward referral for CI assessment, to identify both the rate at which CI assessment has been offered by local HA teams and the proportion and characteristics of families that do or do not accept onward referral. Next, of those referred for CI candidacy assessment, a review of records at the CI centres would reveal the proportions and characteristics of children who are a) found to be audiometrically-eligible by the CI team; b) deemed to have "insufficient benefit from HAs" (fulfilling the other requirement of TAG566 (NICE, 2019)) and c) meet the other requirements of the CI team's eligibility process such as medical fitness for surgery, duration of auditory deprivation, realistic expectations and family support, and d) the proportion and characteristics of families who then accept the offer of CIs. Those who reach the end of this process will be a group of children who were not eligible for CIs prior to 2019 but who access CIs due to TAG566 (NICE, 2019). The benefits of measuring their outcomes before and following implantation will be discussed in the following section.

7.3.3. Before and after outcomes of children receiving CIs whose HTLs fell outside TAG166 but within TAG566

Monitoring this group of children's outcomes before and after implantation would further help to validate TAG566 (NICE, 2019), because children would act as their own controls in terms of the potentially confounding variables

discussed throughout this thesis. Monitoring outcomes of this group would thereby fill gaps in the evidence base.

An alternative before and after comparison of HA and CI outcomes for this group would be possible if uptake of CIs among those newly eligible was not universal. It would then be possible to recruit both children who receive CIs and children who are offered CIs but whose families decline them. The group of children who received CIs could be assessed before and after implantation, and those who continued to use HAs could be monitored at equivalent time intervals. A similar method was used to form a study group of CI users and control group of HA users with equivalent hearing loss in a study of cognitive function pre- and post-implantation in adults (Mertens et al., 2020). The control group were adults who were not eligible for implantation in their country's healthcare system, or who were on long waiting lists for implantation. For children within the NHS, any changes in outcomes for the control HA group over time could be used to differentiate between changes in the outcomes of the CI group caused by the intervention and potentially confounding changes due to e.g. increased age, maturity, and duration of listening experience.

7.3.4. Improving the quality of case-control matching for future analyses of the impact of hearing loss on the early educational outcomes of children in the Born in Bradford cohort

Case-control matching can be used to compare the outcomes of children with hearing loss to their hearing peers while controlling for other potentially confounding variables (Di Stadio et al., 2018; Walker et al., 2020). Using this

method, Chapter 6 found significantly poorer EYFSP outcomes for all sub-groups of children with hearing loss compared to their matched controls. To increase the quality of case control matching, a future project could request the hospital numbers of the whole cohort, not just those who had been screened for the CUSP study. This would allow the creation of a more-matched comparator group, because it could be confirmed that matched cases had no history of otological or audiological problems. Previously published work on the relationship between the presence of autism and EYFSP outcomes of the BiB cohort (Wright et al., 2019) suggests that data regarding children's autism diagnoses, not present within the data dictionary, might be available for linkage also via collaboration with the authors and BiB executive group. Matching for autism and confirming the absence of hearing problems for the controls, in addition to controlling for the variables already used in Chapter 6, would strengthen the ability of the comparisons to reflect only the effects of hearing loss on children's outcomes.

7.3.5. Studies to determine the optimal intervention for children with unilateral, mild, or conductive hearing loss

Chapter 6 found that any degree of hearing loss was associated with poorer early educational outcomes for children. Practice guidance documents are insufficient to guide audiologists and surgeons in knowing the optimum management strategy for children with conductive losses (British Society of Audiology, 2021; Browning et al., 2010; National Institute for Health and Care Excellence, 2008) and the management of mild losses with HAs can be difficult (Lin et al., 2022). Randomised trials of hearing device interventions,

either provided in isolation or in conjunction with other strategies such as surgical and middle ear aeration approaches, could be beneficial in informing future practice (Tham et al., 2018). Chapter 6 indicates that EYFSP scores could be a useful outcome measure to include in the assessment battery.

7.3.6. Longer-term follow up of educational attainment in children with hearing loss

The EYFSP is conducted toward the end of the academic year during which children turn five years of age, much earlier than the educational assessments that will influence access to further or higher education, or employment. It is important to know whether or not the attainment gap between children with hearing loss and their controls can narrow as children progress through the education system. In the general population, there is evidence that such gaps can remain over time. Decreasing but persistent correlations were found between the pre-2013 Foundation Stage measure and exam grades up to the age of 16 years for a cohort of over 500,000 UK pupils, with Foundation Stage score modelled to account for 33% of the long-term attainment gap for children from the most deprived backgrounds (Treadaway, 2019). Among the BiB cohort, reaching the GLD on the EYFSP used for the present study was found to be predictive of reading ability at the end of Year 2 (two years later), most strongly for children receiving SEN support (Atkinson, 2021). These results suggest it cannot be assumed that children with hearing loss who have underperformed on the EYFSP compared to their typically-hearing peers will necessarily catch up in subsequent years, and that children from more deprived backgrounds may be the least likely to do so. Further work is needed to see if

EYFSP scores of children with hearing loss are predictive of later attainment and whether hearing interventions for this group could narrow the gap.

7.3.7. Potential for national pooling of data

Twenty years ago it was feasible for Fortnum et al (Fortnum et al., 2001; Fortnum, Marshall, & Summerfield, 2002) to study all children with permanent bilateral hearing loss of moderate degree or greater. They asked clinical and education professionals to extract and share comprehensive data on their whole caseloads of children at one point in time, with complete national participation. Though consulting with heads of department and implementing the CUSP study, and considering evidence of less than complete compliance with other national reports such as the CRIDE and NDCS reports (CRIDE, 2021), it is clear that this is not feasible in the current NHS. However, a national registry of hearing implants (BCIG Website, 2022) such as that established in France (Artières-Sterkers et al., 2020) is under development in Britain. Its main focus will be on monitoring safety, similarly to the UK's National Joint Registry (National Joint Registry, 2022) and is likely to include the consistent, national monitoring of some clinical outcome measures (Jenny Townsend, personal communication, 2022). Coordination at this level has the potential to provide a range of outcome measures for a truly representative cohort of children newly accessing cochlear implantation in greater numbers than in the present study. If the national registry could also include data on children referred for CI assessment but not implanted, and children's outcomes via HAs prior to implantation, then this could increase its

potential to provide evidence to support HA and CI candidacy decision making in the future.

7.4. ABILITY OF THE STUDY TO DETECT MEANINGFUL DIFFERENCES

As explained in chapter 3, a prospective power calculation was only performed for the word perception in noise test originally planned as the primary outcome measure but not ultimately collected. This section provides a retrospective discussion of whether sample sizes were adequately powered to detect meaningful differences in the measures used, beginning with the key outcome measure of the thesis, children's PEACH scores.

The survey sample size was sufficient to identify differences between CI users and each group of children with HAs not confirmed to fall outside TAG566, as evidenced by the significant findings of the regression model in section 5.3.2. The effect sizes associated with NICE eligibility group compared to children with CIs, adjusted for caregiver education and additional needs, were 13% for children with HAs newly eligible for CIs, 19% for children of unknown eligibility, and 42% for children previously eligible for CIs but using HAs. In the present study, only five out of six items from the original quiet sub-scale and four out of five items from the noise sub-scale were included, so some caution is needed when comparing these effect sizes to the literature. However, Ching & Hill (2007) reported that a difference in total score of 16.5% was equivalent to the difference between a hearing loss of 50 dB HL and 80 dB HL averaged across 0.5, 1 and 2 kHz. This would be considered highly meaningful in terms of children's language development (Tomblin et al, 2015). In

conclusion, the sample size for this key outcome measure was sufficient to detect meaningful group differences in children's parent-reported aural/oral outcomes.

Similarly, statistically significant differences were found in Chapter 6 between the GLD pass rates of study versus control groups, indicating that this chapter was sufficiently powered. The GLD pass/fail criterion is used for schools to allocate targeted teaching resources to children, meaning it has an immediate and meaningful effect on children's education and UK education expenditure. GLD pass/fail rate has also been associated with children's later academic attainment, indicating that the difference between the groups was meaningful in terms of children's odds of achieving academic success up to the age of 16 years (Atkinson et al., 2022; Treadaway, 2019).

With regard to the other parent-reported outcomes in the survey, there were no previously published studies on the comparative prevalence of tinnitus or need for recovery from listening on which to base power calculations. It cannot be known whether the lack of significant group differences in parent reported prevalence in the present study was due to an insufficient sample size and/or a true similarity between the groups.

7.5. CONCLUSION

This thesis adds to the evidence base to guide clinicians when choosing whether to offer acoustic hearing aids or cochlear implants to children with hearing loss within or close to 2019 NICE guidance (TAG566). It tentatively supports TAG566 (NICE, 2019) since only children using HAs confirmed to fall outside this guidance had equivalent caregiver reported aural/oral

outcomes to children using CIs. The thesis provides novel evidence revealing that TAG566 (NICE, 2019) has increased the number of children for whom eligibility for both CI assessment referral and CI candidacy is not known. It also provides novel measurement of the impact of single-sided, mild, or moderate hearing loss on children's early years educational outcomes, confirming that standard clinical and educational practice was not sufficient to overcome the detriment associated with hearing loss. This work can impact clinical practice via suggestions around CI candidacy assessment referral that would decrease the uncertainty regarding which children might benefit from implantation. It also highlights a need for changes to clinical and educational practice for young children with any degree of hearing loss. Consultations and research are needed to clarify the optimum approaches to CI eligibility assessment referral and the timing, type of hearing device, and type of educational support offered to young children in the UK.

APPENDICES

Appendix A: Hearing aid eligibility screening form

CUSP Level 1 Screening Data Extraction Form – HA users

Data not recorded mark "NR"
Data recorded as not tested mark "NT"
Data not needed mark "NN"
Data recorded that could not be extracted mark "CNE"
M = masked; N = not masked; ABR = Auditory Brainstem Response
You can use the tab key to move from box to box
Do not record any information that could identify the patient

CUSP
COMPARING OUTCOMES FOR SEVERELY AND PROFOUNDLY DEAF CHILDREN

Name of person extracting data:	Date of extraction:
Type your name here	Choose today's date.

Signature of person extracting data: _____

CHILD'S DETAILS: Please make sure that the study code exactly matches the code for this child on the key document. All cells must contain some response.

Study Code			Sex	Has acoustic hearing aid(s)?		Age at date of screening:		Interpreter needed for audiology appointments?		For children aged 5 or older, any record of additional needs that affect their ability to do hearing tests?	
Site	No.		M/F	Right	Left	years	months	Y/N	Language / "NN"	Y/N	Brief comment
Choose your site	e.g. 007	HA	Choose	Choose	Choose	Choose age in years	Choose months.	Choose	Interpreter language	Choose	Summarise additional needs and how they affect hearing tests, or type "NR" if none recorded, or "NN" if under 5.

CHILD'S UNAIDED HEARING LEVELS: Enter the most recent unaided hearing levels (Audio 1). If Audio 1 is complete, enter "NN" for Audio 2. If any A/C frequencies were not tested at Audio 1, go to the next most recent test (Audio 2). If any frequencies not tested at Audio 1 were tested at Audio 2, enter full data for Audio 2. Otherwise, choose "NT" for frequencies missing from both Audio 1 and Audio 2 and you do not need to enter the other Audio 2 thresholds, put "NN". Enter ear-specific results if available. If only soundfield thresholds were recorded, enter these in the "RIGHT" A/C columns, and enter "NN" in the "LEFT". Enter bone conduction results for the same test dates.

Audio 1						Audio 2					
Test date 1	date 1	dB scale 1	scale	A/C transducer 1	Choose	Test date 2	date 2	dB scale 2	scale	A/C transducer 2	Choose

Test	RIGHT Unaided OR SOUNDFIELD unaided AIR CONDUCTION (A/C) thresholds (kHz)																LEFT unaided A/C thresholds (kHz) OR enter "NN" if previous column has soundfield results															
	0.25			0.5			1			2			3			4			0.25			0.5			1			2			3			4		
Audio 1 A/C																																		
Audio 2 A/C																																		
	RIGHT BONE CONDUCTION (B/C) thresholds (kHz)																LEFT BONE CONDUCTION thresholds (kHz)															
	0.5			1			2			3			4				0.5			1			2			3			4			
Audio 1 B/C																																		
Audio 2 B/C																																		

Appendix B: CUSP survey questions

1. ABOUT YOUR CHILD AND THEIR COMMUNICATION

C1. How many years old is your child? _____ years

C2. Is your child female or male? Delete as appropriate: Female / Male

C3. What stage of life is your child currently in? (If you are filling this in during the summer holiday, please choose the year group your child has just completed). Please choose one.

- ❑ Infant / Pre-school (aged 0 to 3)
- ❑ Early Years (aged 3 to 4)
- ❑ Early Years Reception (aged 4 to 5)
- ❑ Key Stage 1 (aged 5 to 7, school years 1 and 2)
- ❑ Key Stage 2 (aged 7 to 11, school years 3, 4, 5 and 6)
- ❑ Key Stage 3 (aged 11 to 14, school years 7, 8 and 9)
- ❑ Key Stage 4 (aged 14 to 16, school years 10 and 11)
- ❑ A-levels at a school sixth-form

If you chose one of these, please go to C3.1

- ❑ A-levels at a college
- ❑ Further Education (studying for a college course)
- ❑ Higher Education (studying for a degree at college or university)
- ❑ Training / Apprenticeship
- ❑ Employment

If you chose one of these, please go to C4

- ❑ Not in education, employment or training

If you chose this, please go to C5

C3.1 What kind of care / education provision does your child have? Choose one.

- ❑ Pre-school aged, supported only at home
- ❑ School aged and supported only at home (i.e. home-schooled)

If you chose one of these, go to C5

- ❑ Pre-school aged with carer, e.g. nursery, nanny, or other kind of day-care
- ❑ Mainstream state-funded school (including academies and free schools)
- ❑ Mainstream independent school (private school, not state-funded)
- ❑ Resource provision in a mainstream school
- ❑ Special school for deaf pupils (whether state-funded or not)
- ❑ Other special school, not specifically for deaf children (whether state-funded or not)

If you chose one of these, please go to C4

C4. How do your child's carers / teachers / work colleagues communicate with your child at home, day-care, pre-school, school or work?

- ❑ Spoken language only
- ❑ British sign language only

- ❑ Spoken language together with signed support
- ❑ Other

If you selected Other, please specify:_____

C5. How do *you* communicate with your child at home?

- ❑ Spoken language only
- ❑ British sign language only
- ❑ Spoken language together with signed support
- ❑ Other

If you selected Other, please specify: _____

C6. How does *your child* communicate with you at home?

- ❑ Spoken language only
- ❑ British sign language only
- ❑ Spoken language together with signed support
- ❑ Other

If you selected Other, please specify: _____

C7. How do *other family members* communicate with your child?

- ❑ Spoken language only
- ❑ British sign language only
- ❑ Spoken language together with signed support
- ❑ Other

If you selected Other, please specify: _____

C8. How does *your child* communicate with other family members?

- ❑ Spoken language only
- ❑ British sign language only
- ❑ Spoken language together with signed support
- ❑ Other

If you selected Other, please specify: _____

C9. What is the main spoken language used in your home?

- ❑ None, we only use British Sign Language
- ❑ English
- ❑ Other spoken language

If you selected "other spoken language", please specify: _____

C10. How many different spoken languages does your child listen to during a typical week?

_____

C11. Apart from their hearing, does your child have any additional needs? Please choose all that apply.

- ❑ No additional needs apart from hearing loss
- ❑ Learning difficulty
- ❑ Behaviour, emotional or social difficulties
- ❑ Speech, language and communication needs
- ❑ Vision impairment
- ❑ Physical disability
- ❑ Autistic spectrum disorder
- ❑ Other difficulty/disability (please specify):

If you answered "Other difficulty / disability", please specify _____

C12. What is the cause of your child's hearing loss? (options continue on the next page)

- ❑ Meningitis
- ❑ Cytomeglovirus (CMV)
- ❑ Wide vestibular aqueduct syndrome
- ❑ Auditory Neuropathy Spectrum Disorder (ANSD)
- ❑ Usher's Syndrome
- ❑ Another syndrome
  Please give the name of the syndrome your child has:
  _____
- ❑ Childhood deafness runs in the family, but we don't know what gene
- ❑ Known genetic cause, e.g. GJB2
  Please give the name of the gene, if you know it:
  _____
- ❑ Ototoxicity (e.g. a medicine like strong antibiotics caused the hearing loss)
  Please give the name of the ototoxic medicine or treatment, if you know it:
  _____
- ❑ Other cause
  Please specify what the other cause was:
  _____
- ❑ The hospital told me that the cause is unknown
- ❑ The hospital told me the cause, but I can't remember what it was

C13. How old was your child when their hearing loss was first picked up?

- ❑ _____ years and _____ months.
- ❑ I can't remember

C14. How old was your child when they first got a hearing aid?

- ❑ _____ years and _____ months.
- ❑ I can't remember

C15. How old was your child when they first got a cochlear implant? [CI survey only]

- ❑ _____ years and _____ months.
- ❑ I can't remember

ABOUT THE PERSON FILLING OUT THIS SURVEY

P1. What best describes your relationship to the child?

- ❑ Mum
- ❑ Dad
- ❑ Grandmother
- ❑ Grandfather
- ❑ Other

If you selected Other, please specify:

_____

P2. What best describes your own hearing?

- ❑ I have good hearing
- ❑ I am hard of hearing
- ❑ I am Deaf

P3. Do you use anything to help you hear better? Choose all that apply

- ❑ No, I don't use anything to help me hear better
- ❑ I use a hearing aid
- ❑ I use a cochlear implant
- ❑ I use another device

Please specify what kind of device:

_____

P4. What level of education do you have? Please choose all that apply

- ❑ School
- ❑ A-levels
- ❑ College course
- ❑ Professional qualification
- ❑ Undergraduate degree, e.g. BA, BSc
- ❑ Post-graduate degree, e.g. MA, MSc
- ❑ Doctorate, e.g. PhD
- ❑ I prefer not to say
- ❑ Other

If you selected Other, please specify

_____

P5. Does your child have another main care-giver?

- ❑ Yes — **Please answer P6, P7, P8 and P9.**
- ❑ No — **Please go to the Listening questions.**

P6. Thinking about the other main care-giver for your child, what best describes this person's relationship to your child?

- ❑ Mum
- ❑ Dad
- ❑ Grandmother
- ❑ Grandfather
- ❑ Other

If you selected Other, please specify _____

P7. What best describes this person's hearing?

- ❑ They have good hearing
- ❑ They are hard of hearing
- ❑ They are Deaf
- ❑ I don't know

P8. Do they use anything to help them hear better? Choose all that apply.

- ❑ No, they don't use anything to help them hear better
- ❑ They use a hearing aid
- ❑ They use a cochlear implant
- ❑ They use another device

Please specify what kind of device

_____

❑ I don't know

P9. What level of education does this other main caregiver have? Please choose all that apply

❑ School
❑ A-levels
❑ College course
❑ Professional qualification
❑ Undergraduate degree, e.g. BA, BSc
❑ Post-graduate degree, e.g. MA, MSc
❑ Doctorate, e.g. PhD
❑ I prefer not to say
❑ Other

If you selected Other, please specify _____

❑ I don't know

HOW YOUR CHILD'S HEARING AFFECTS THEIR EVERYDAY LIFE
LISTENING QUESTIONS

Think about your child's behaviour OVER THE PAST WEEK in relation to each question. Give a rating, based on the estimated percentage of time that your child displays the described behaviour.

L1. Has your child been wearing his/her hearing aid(s)/cochlear implant(s)? Choose one:

❑ Yes ❑ No

L2. Has your child been well/healthy? Choose one:

❑ Yes ❑ No

L3. Have your child's hearing aid(s) been working properly? Choose one:

❑ Yes ❑ No

L4. How often has your child worn his/her hearing aid(s)/cochlear implant(s)? Choose one:

❑ Never (0%)
❑ Seldom (1 - 25%)
❑ Sometimes (26 - 50%)
❑ Often (51 - 75%)
❑ Always (76 - 100%)

L5. How often has your child complained or been upset by loud sounds? Choose one:

❑ Never (0%)
❑ Seldom (1 - 25%)
❑ Sometimes (26 - 50%)
❑ Often (51 - 75%)
❑ Always (76 - 100%)

L6. When you call, does your child respond to his/her name in a quiet situation? Choose one:

❑ Never (0%)
❑ Seldom (1 - 25%)
❑ Sometimes (26 - 50%)
❑ Often (51 - 75%)

❑ Always (76 - 100%)

L7. When asked, does your child follow simple instructions or do a simple task in a quiet situation? Choose one:
❑ Never (0%)
❑ Seldom (1 - 25%)
❑ Sometimes (26 - 50%)
❑ Often (51 - 75%)
❑ Always (76 - 100%)

L8. When you call does your child respond to his/her name in a noisy situation when he/she can't see your face? (examples of responses include looks up, turns, answers verbally). Choose one:
❑ Never (0%)
❑ Seldom (1 - 25%)
❑ Sometimes (26 - 50%)
❑ Often (51 - 75%)
❑ Always (76 - 100%)

L9. When asked, does your child follow simple instructions or do a simple task in a noisy situation? Choose one:
❑ Never (0%)
❑ Seldom (1 - 25%)
❑ Sometimes (26 - 50%)
❑ Often (51 - 75%)
❑ Always (76 - 100%)

L10. When you are in a quiet place reading with your child, how often does he/she pay close attention to what you are saying? OR if your child is listening to stories/songs on the TV or CD when there is no other background noise, how often can he/she follow what is being said? Choose one:
❑ Never (0%)
❑ Seldom (1 - 25%)
❑ Sometimes (26 - 50%)
❑ Often (51 - 75%)
❑ Always (76 - 100%)

L11. How often does your child initiate / participate in conversation in a quiet situation? Choose one:
❑ Never (0%)
❑ Seldom (1 - 25%)
❑ Sometimes (26 - 50%)
❑ Often (51 - 75%)
❑ Always (76 - 100%)

L12. How often does your child understand what you say in the car / bus / train? Choose one:
❑ Never (0%)
❑ Seldom (1 - 25%)
❑ Sometimes (26 - 50%)
❑ Often (51 - 75%)
❑ Always (76 - 100%)

L13. How often does your child recognise people's voices without seeing who was talking? Choose one:

- ❑ Never (0%)
- ❑ Seldom (1 - 25%)
- ❑ Sometimes (26 - 50%)
- ❑ Often (51 - 75%)
- ❑ Always (76 - 100%)

L14. How often does your child successfully use a phone? Choose one:

- ❑ Never (0%)
- ❑ Seldom (1 - 25%)
- ❑ Sometimes (26 - 50%)
- ❑ Often (51 - 75%)
- ❑ Always (76 - 100%)

L15. How often does your child respond to sounds other than voices? Choose one:

- ❑ Never (0%)
- ❑ Seldom (1 - 25%)
- ❑ Sometimes (26 - 50%)
- ❑ Often (51 - 75%)
- ❑ Always (76 - 100%)

L16. Please give any extra comments you would like to about these listening questions:

_____

TINNITUS QUESTIONS

T1. Has your child ever told you that they hear noises in their ears or head, that don't come from their hearing aids, cochlear implants, or surroundings (tinnitus)?

- ❑ Yes

Please answer T1.1, T1.2, T1.3, T1.4 and T1.5.

- ❑ No
- ❑ My child is not able to tell me

Please go to the Need for Recovery questions

Please answer these questions thinking about the noises your child has told you they hear in their ears or head that don't come from their hearing aids, cochlear implants or surroundings.

T1.1. Does your child say that they find these noises annoying?

- ❑ Yes, my child has told me they find the noises annoying.
- ❑ No, my child has told me that they do not find the noises annoying.
- ❑ I don't know, we haven't talked about it.

T1.2. Does your child say that these noises disturb their sleep?

- ❑ Yes, my child has told me that these noises disturb their sleep.
- ❑ No, my child has told me that these noises do not disturb their sleep.
- ❑ I don't know, we haven't talked about it.

Please give any extra comments you would like to about these tinnitus questions.

_____

NEED FOR RECOVERY QUESTIONS

R1. Following their usual activities, such as childcare / school / studying / work, how often does your child want a break from using their **RIGHT** hearing aid/cochlear implant?

- ❑ Never
- ❑ Hardly ever
- ❑ Sometimes
- ❑ Frequently
- ❑ They don't have a hearing aid for their right side
- ❑ My child is unable to tell me

R2. Following their usual activities, such as childcare / school / studying / work, how often does your child want a break from using their **LEFT** hearing aid/cochlear implant?

- ❑ Never
- ❑ Hardly ever
- ❑ Sometimes
- ❑ Frequently
- ❑ They don't have a hearing aid for their left side
- ❑ My child is unable to tell me

R3. How often does your child complain of headaches related to listening?

- ❑ Never
- ❑ Hardly ever
- ❑ Sometimes
- ❑ Frequently
- ❑ My child is unable to tell me

R4. How often does your child complain of tiredness related to listening?

- ❑ Never
- ❑ Hardly ever
- ❑ Sometimes
- ❑ Frequently
- ❑ My child is unable to tell me

R5. Please give any extra comments you would like to about these need for recovery questions.

_____

YOUR OPINIONS AND EXPERIENCES OF HEARING AIDS AND COCHLEAR IMPLANTS

HA1. What device has your child been given for their RIGHT ear? Please select the type of device they have been given, even if they do not currently use it

- ❑ Hearing aid / cochlear implant — **Please answer HA1.1, HA1.2 and HA1.3.**
- ❑ They do not have a device for their right ear — **Please go to HA2**

HA1.1. On a typical day, for how many hours does your child use their RIGHT hearing aid / cochlear implant switched on?

- ❑ _____ hours

❑ They don't use their right hearing aid

HA1.2. How easy is it for you to get your child to use their RIGHT hearing aid / cochlear implant?

❑ Very easy
❑ Quite easy
❑ Quite difficult
❑ Very difficult

HA1.3. Why is it as easy or difficult as you have said?

_____

HA2. What device has your child been given for their LEFT ear? Please select the type of device they have been given, even if they do not currently use it

❑ Hearing aid / cochlear implant — **Please answer HA2.1, HA2.2 and HA2.3.**

❑ They do not have a device for their left ear — **Please go to CI1**

HA2.1. On a typical day, for how many hours does your child use their LEFT hearing aid / cochlear implant switched on?

❑ _____ hours
❑ They don't use their left hearing aid

HA2.2. How easy is it for you to get your child to use their LEFT hearing aid / cochlear implant?

❑ Very easy
❑ Quite easy
❑ Quite difficult
❑ Very difficult

HA2.3. Why is it as easy or difficult as you have said?

_____

[The following 'CI' questions were only asked to children using HAs]:

CI1. Has your child ever been seen at a cochlear implant centre to find out if cochlear implants would be an option for them?

❑ Yes — **Please answer CI2**

❑ No — **Please go to CI3**

If your child has been assessed more than once, please give your answers based on your most recent assessment.

CI2. What was the outcome of your child's cochlear implant assessment? Choose one:

❑ The cochlear implant department decided not to offer cochlear implantation for my child

- ❑ The cochlear implant department offered cochlear implantation for my child, but we decided not to go ahead
- ❑ We decided not to complete the assessment process
- ❑ We have accepted cochlear implantation for our child
- ❑ We are currently in the middle of the assessment process, which has not been completed

CI3. Have you ever thought about having a cochlear implant assessment for your child?

- ❑ I have asked a health professional if we could be referred for a cochlear implant assessment
- ❑ I have thought about asking for a cochlear implant assessment but not yet talked about it
- ❑ I have not thought about a cochlear implant assessment for my child

S1. Are you satisfied that using [hearing aids / cochlear implants] is in the best interests of your child at the moment?

- ❑ Very satisfied
- ❑ A little satisfied
- ❑ A little dissatisfied
- ❑ Very dissatisfied

Appendix C: Original PEACH

	Question	Never 0%	Seldom 1 - 25%	Sometimes 26 - 50%	Often 51 - 75%	Always 75-100%
1.	How often has your child worn his/her hearing aids and/or cochlear implant?	0	1	2	3	4
2.	How often has your child complained or been upset by **loud** sounds?	4	3	2	1	0
3.	When you call, does your child respond to his/her name in a **quiet** situation?	0	1	2	3	4
4.	When asked, does your child follow simple instructions or do a simple task in a **quiet** situation?	0	1	2	3	4
5.	When you call does your child respond to his/her name in a **noisy** situation when he/she can't see your face? (examples of responses include looks up, turns, answers verbally)	0	1	2	3	4
6.	When asked, does your child follow simple instructions or do a simple task in a **noisy** situation?	0	1	2	3	4
7.	When you are in a **quiet** place reading with your child, how often does he/she pay close attention to what you are saying? OR if your child is listening to stories/songs on the TV or CD when there is no other background noise how often can he/she follow what is being said?	0	1	2	3	4
8.	How often does your child initiate/ participate in conversation in a **quiet** situation?	0	1	2	3	4
9.	How often does your child initiate/ participate in conversation in a **noisy** situation?	0	1	2	3	4
10.	How often does your child understand what you say in the car/bus/train?	0	1	2	3	4
11.	How often does your child recognise peoples' voices without seeing who was talking?	0	1	2	3	4
12.	How often does your child successfully use a phone?	0	1	2	3	4
13.	How often does your child respond to sounds other than voices?	0	1	2	3	4

Appendix D: Assessing the quality of the final PEACH model

The R square value indicates that the final model accounted for 48.5% of the variance in the sample data. The difference between this value and the adjusted R square value indicated that if the model were derived from the whole population instead of the study sample, it would account for approximately 3% less variance in the outcome. The F value was greater than 1 and highly significant, indicating that the model significantly improved predictions of the PEACH score compared to assuming each child would have a mean score (Field, 2000). The Durbin-Watson test statistic was close to the value of 2, strongly suggesting that the model satisfied the assumption of independent errors.

The expectation in linear regression modelling would be that approximately 5% of cases would have standardised residuals less than -2 or greater than +2 (Field, 2000). Casewise diagnostics of the final model confirmed that only four participants had standardized residuals outside this range, comprising 3.6% of the 109 participants. This indicates that the model provided predictions that were accurate for an acceptable proportion of cases. No case had a Cook's distance greater than 1, suggesting that no one case was exerting undue influence over the model.

Variable types: All predictor variables were quantitative or categorical, and the outcome variable was quantitative, continuous and unbounded, meeting the first assumption.

Non-zero variance: All predictors had some variation in value, meeting this assumption.

No perfect multicollinearity: There was no correlation greater than R = 0.9 between any predictor variables in the final model. The strongest correlation between any predictor variables was R = -0.301 between caregiver highest qualification classification and the presence of additional needs, however this was not statistically significant. All tolerance and variance inflation factor values were consistent with an acceptably low degree of multicollinearity between predictor variables.

The exploratory model of all potential predictor variables found that these had low degrees of multicollinearity, suggesting that the final model met the assumption that predictors were uncorrelated with external variables.

The scatterplot of standardised residuals against standardised predicted values (Figure D.1) shows points were dispersed around zero, indicating that the final model met the assumption of homoscedascticity, namely that at each level of the predictor variables the variance of the residual terms should be constant.

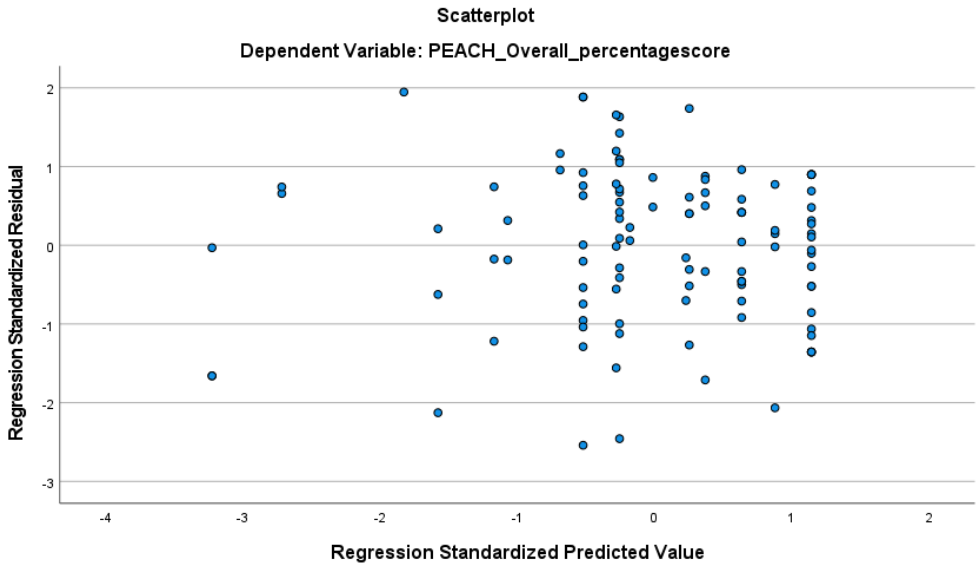


Figure D.1: Final PEACH model standardised residuals versus standardised predicted values

Normally distributed errors: Figure D.2 shows that the distribution of the standardised residuals is close to normal, with no residuals exceeding a value of ±3.

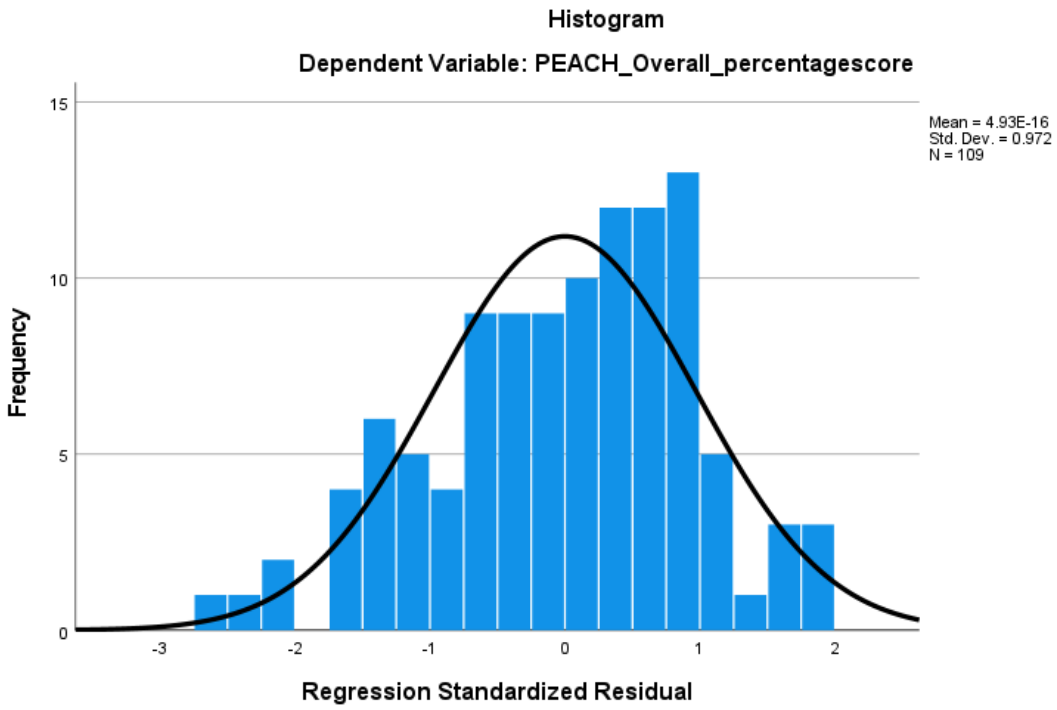


Figure D.2: Final PEACH model distribution of standardised residuals

Figure D.3 is a normal probability plot of the standardised residuals of the final model. Individual observed residuals are plotted as circles. They lie close to the horizontal line that represents a perfectly normal distribution.

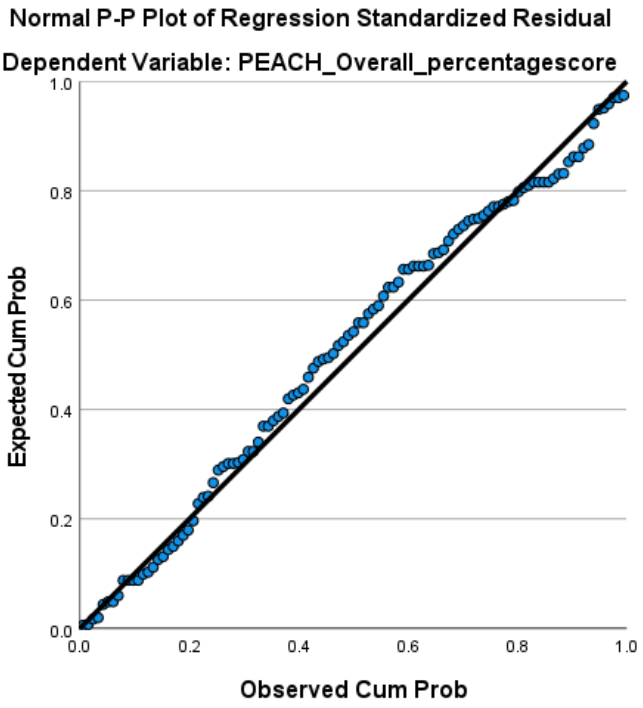


Figure D.3: Final PEACH model normal probability plot of the standardised residuals

Last, the model met the assumption of independence, because each PEACH score came from a separate participant. In conclusion, the final model of PEACH scores appears to be accurate for the sample and also generalisable to the population.

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