

# **Impact of Introducing Cortical Auditory Evoked Potentials (CAEP) into the Newborn Hearing Assessment Pathway**

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**Thesis submitted to University College London in  
fulfilment of the requirements for the degree of Doctor  
of Philosophy in Brain Sciences**

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**Declaration**

I, Kinjal Mehta confirm that the work presented in the thesis is my own. Where information has been derived from other sources, I confirm that this has been indicated in the thesis. Signed:

.....

## **Abstract**

Cortical Auditory Evoked Potentials (CAEPs) are electrophysiological responses in the cortex in response to sounds. In recent years it has become possible to measure CAEPs and the technology is clinically available.

The aim of this research was to demonstrate if CAEPs are feasible to use in a clinical setting and to see if the CAEPs were helpful in optimising hearing aid use for infants under 6 months by showing if a child could detect speech tokens when aided. Recording of CAEPs with speech tokens presented in the free field was introduced into the audiology pathway for infants with a PCHI in 2011-2015 at a UK clinical service. Thirty-four children had followed an audiology pathway prior to CAEP introduction and forty-four children followed a pathway after the introduction of CAEP (using unaided and aided responses). Results showed that the median age at hearing aid fitting prior to CAEP introduction was 9.2 months and after the inclusion of CAEPs reduced to 3.9 months. The current study demonstrates that the inclusion of CAEP recording in the pathway facilitated earlier hearing aid fitting for milder-impairments.

Two studies were conducted to better understand these findings. The first was a focus group to determine the factors that influenced parents' acceptance of early aiding or early referral for cochlear implants. Eight sets of parents participated and they discussed all factors that affected their decision making process. The findings showed that hearing the speech tokens and seeing the clear pass/fail response was helpful for parents. The second study evaluated clinicians' viewpoints. A questionnaire was developed and completed by 49 clinicians around the world (including the UK). Clinicians used the CAEPs to verify or modify hearing aid fittings and to counsel parents, reinforcing the need for hearing aids. The impact of the research has resulted in earlier hearing aid fitting, cochlear implant referrals and improved hearing aid use for children under the age of 6 months and allowed for honest, informative discussions with families.

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## **Impact Statement**

The findings reported in this thesis are relevant for academic and clinical applications. The main finding is that use of cortical auditory evoked potential (CAEPs) add information to paediatric audiology management. The findings provide unequivocal evidence for the usefulness of CAEPs in clinical protocols.

Within academia, this research has contributed to the evidence-base on the usefulness of CAEPs in reducing the social impact that hearing loss can have on a child and her/his family. The study has demonstrated the worth of undertaking CAEP recording in children with a permanent childhood hearing impairment (PCHI) identified by the Neonatal Hearing Screening Programme (NHSP). The recording of CAEPs is beneficial for the use of hearing aids in the first 6 months of infancy. We have been able to demonstrate that the introduction of CAEPs resulted in a reduction in the age at intervention, resulting in earlier hearing aid fitting, cochlear implant referral and improved hearing aid use.

In the clinical domain, we have shown that CAEPs can be implemented into mainstream clinical practice successfully and have a positive impact. These findings will contribute to professional practice, clinical procedures and policies. The findings have established that CAEPs are quick to administer and practical to use in busy clinics.

CAEP measurements have been positively received by both patients' families and clinicians. The inclusion of CAEPs as a routine test gives improved efficiency, with clinicians reporting that they were more confident in the management of young deaf children. The results also provided a valuable cross-check method of hearing aid benefit. The study has shown that the benefits of hearing aids were apparent to parents because of the clear way that CAEP responses were indicated. This allowed clear communication of these non-language based on outcomes, permitting families to understand their infant's hearing loss and the consequent benefit of amplification or cochlear implants.

Our findings have raised clinical questions about the importance of managing mild-moderate hearing losses. Within the current NHSP protocol, identification and management of milder hearing loss has not been regarded as a priority for

detailed assessment and intervention. Evidence from the studies reported here indicate clearly that such losses should be diagnosed in a timely manner. The impact of the findings have been important to the individual children and families taking part in the studies, and will undoubtedly be advantageous to future generations of deaf children in our care. The study also supports the inclusion of CAEP recording in the audiology pathways, undertaken after NHSP identification and before behavioural verification of hearing aid fitting is possible.

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## **Abbreviations**

ABR	Auditory brainstem response
ACA	Aided Cortical Assessment
ANSD	Auditory neuropathy spectrum disorder
ASSR	Auditory Steady State Responses
dB SPL	Decibel sound pressure level
BPVS	British Picture Vocabulary Test
BSA	British Society of Audiology
CAEP	Cortical auditory evoked potential
CELF	Clinical Evaluation of Language Fundamentals
DSL	Desired Sensation Level
EAL	English as additional language
E1L	English as first language
HOP	Hearing Outcome Project
IHC	Inner hair cell
LOCHI	Longitudinal Outcomes of Children with Hearing Impairment
NAL-NL1	National Acoustic Laboratories nonlinear
NHSP	Newborn Hearing Screening Programme
OAE	Otoacoustic Emissions
OHC	Outer hair cell
PCHI	Permanent childhood hearing impairment
PPVT	Peabody Picture Vocabulary Test
PSW	Parent Support Worker
PTA	Pure Tone Audiogram
RECD	Real-ear-to-coupler difference
REM	Real ear measurement
SES	Socio-economic status
SNHL	Sensorineural
TEOAE	Transient Otoacoustic Emissions
TOD	Teacher Of the Deaf
UNHS	Universal Neonatal Hearing Screen

## **Relevant Publications**

Mehta, K., Watkin, P., Baldwin, M., Marriage, J., Mahon, M. & Vickers, D. (2017). Role of Cortical Auditory Evoked Potentials in Reducing the Age at Hearing Aid Fitting in Children with Hearing Loss Identified by Newborn Hearing Screening. *Trends in Hearing*, 21. 233121651774409. DOI:10.1177/2331216517744094.

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## **Thesis overview**

The main aim of the studies reported in this thesis was to investigate the use of electrophysiological testing and to see whether it improved the ability to understand whether infants could/couldn't hear speech tokens with or without aids and refer for cochlear implants wherever necessary. Furthermore, we wanted to determine if the use of CAEPs influences hearing aid use for those under 6 months of age.

In this thesis, Chapter 1 reviews the diagnosis and intervention process in the UK for children referred from the NHSP and provides a review of the existing literature. Chapter 2: (study 1) validates the use of cortical auditory evoked potentials (CAEPs). It introduces the use of recording of free-field CAEPs to speech tokens for infants with hearing loss and presents evidence from a study to investigate this. The results show how the use of CAEPs reduced the age of hearing aid fitting recommendation and improved family engagement in the process of hearing aid management. Following the introduction of CAEP, children with profound hearing loss were referred for cochlear implant assessment at a significantly earlier age. Chapter 3: (study 2) is a qualitative study of parents' perspectives on the value of CAEP recording, describing how the data collected from a focus group was analysed and results presented. The focus group provided insight into the parental perception of the process of hearing assessment and hearing aid fitting or cochlear implant referral for their child in infancy. Chapter 4: (study 3) is the views of clinicians on the potential use of CAEPs in the infant audiology pathway. Findings from a questionnaire demonstrated that CAEPs were considered valuable for clinical practice. Chapter 5 summarises the main findings of the previous chapters, discusses implications of the results for clinical practice, providing recommendations for future research.

# **CHAPTER 1: Overview of the auditory pathway & infant audiology pathway**

## **1.1 Introduction**

In the UK, the incidence of congenital bilateral hearing impairment greater than 40 dB HL is estimated to occur in 1.18/1000 cases (Fortnum et al., 2001). In the cases where this is permanent childhood hearing impairment (PCHI), it can have a devastating impact on communication skills, education and quality of life, with a high cost to society if effective intervention is not offered before 6 months of age (Davis et al., 1997; Fortnum et al., 2001, Yoshinaga-Itano., 1998). Typically, the term “hearing impaired” is used for people whose main mode of communication is spoken language. To improve outcomes for hearing-impaired children and to be able to offer early intervention, the universal neonatal hearing screen (UNHS) was introduced to reduce the age of confirmation of PCHI (Kennedy et al., 2006; Dalzell et al., 2000). The screening technique was nationally implemented as the Newborn Hearing Screening Programme (NHSP) from 2006. (Davis et al., 2003; [www.hearing.screening.nhs.uk/audiology](http://www.hearing.screening.nhs.uk/audiology))

### **1.1.1 The Newborn Hearing Screening Programme: Age of identification**

The intention of the NHSP was to identify cases of hearing loss within the first few weeks of life and no later than 3 months of age, and to provide appropriate early intervention. Since the roll-out of the NHSP, the most efficient sites have found typical prevalence levels of 0.96/1000 with bilateral, and 0.49/1000 with unilateral, PCHI (Uus et al., 2006; Watkin & Baldwin., 2011). The introduction of the screening programme has resulted in the mean age of diagnosis being reduced from 2.75 years to 3 months nationally (Fortnum et al., 1997; Dalzell et al., 2000; Watkin & Baldwin., 2012).

The early identification of cases with hearing loss have allowed hearing aids to be fitted in the first months of the child’s life (Yoshinaga-Itano, 2004). Acoustic hearing aids are a vital part of rehabilitation with the aim of improving hearing and speech comprehension in hearing impaired people by amplifying the inaudible parts of the speech spectrum, improving perceived intelligibility (McCreery et al.,

2013). Early fitting of hearing aids can reduce long-term effects by providing auditory stimulation in the critical period when neural plasticity is greatest, allowing the auditory pathways to develop similarly to normally hearing infants (Sharma et al., 2002; Ponton et al., 1996).

The predicted benefits of the NHSP was that 80% of infants identified with a hearing loss, who were fitted with hearing aids, would achieve spoken language milestones within the same time window as normal-hearing babies (Yoshinaga-Itano., 2000). However, in contrast to this prediction the Positive Support study from the UK ([www.positivesupport.info](http://www.positivesupport.info)) showed that the percentage of hearing impaired children meeting the typical speech and language milestones at the appropriate age was as low as 10% on some of the measures.

In 2001, a systematic review by the U.S. Preventive Service Task Force concluded that it was unclear whether UNHS and early identification of PCHI were associated with improved language abilities (Thompson et al., 2001). The review was updated in 2008 (Nelson et al., 2008) and included two further studies (Wake et al., 2005 & Kennedy et al., 2006). The Hearing Outcomes Project (HOP; Kennedy et al., 2006) studied 120 children with bilateral permanent hearing impairment. In this group 61 children were born during periods with UNHS and 57 had hearing impairment that was confirmed by nine months of age. This study showed higher scores for receptive language but no significant difference in expressive language or in speech production between children identified by UNHS and children diagnosed in the later group. Similar findings were reported by Wake et al. (2005) who showed that the mean language and reading scores of 86 hearing impaired children aged 7-8 years old did not vary significantly with age of diagnosis. However, the mean age of diagnosis in this study was 21.6 months, with only 11 children diagnosed before the age of 6 months. The mean age of hearing aid fitting was 23.2 months. Even in the HOP project where the hearing losses were confirmed by 9 months of age, only half of the children were actually fitted with hearing aids around the time of diagnosis. This resulted in a delay in amplification which could have been a contributing factor decreasing the potential effect of early detection on children's outcomes. There are other contributing factors affecting children's outcomes than the age at identification and the use of hearing aids. The Longitudinal Outcomes of



Children with Hearing Impairment (LOCHI) study by Ching et al. (2013) enrolled 401 children and 56% of them were fitted with hearing aids before the age of 6 months. This revealed that on average the outcomes were well below population norms and that hearing aid fitting was not significantly associated with better outcomes at 3 years of age. Their findings indicated that there are other significant predictors that contribute to a child's outcomes and this included: presence/absence of additional disabilities, severity of hearing loss, gender, maternal education; together with age of switch-on for children with cochlear implants. A more recent study by the LOCHI team (Ching et al., 2018) showed that better language outcomes were associated with milder hearing loss, use of oral communication, higher levels of cognitive ability and maternal education, and earlier device fitting.

### **1.1.2 The Requirements for Early Auditory Stimulation**

Central auditory pathways need to be developed in the early years of life. It is recognised that early auditory stimulation is important, and that there is a sensitive period for hearing development (Kral et al., 2013). Therefore, those with PCHI need the appropriate auditory stimulation as soon as possible to enable the pathways to develop. The introduction of the NHSP typically has resulted in earlier hearing aid fitting.

Research has shown that a normal hearing infant's auditory system and pathways start to be shaped in-utero (Litovsky, 2015). The unborn foetus can hear and respond to sound during the third trimester of pregnancy (Granier-Deferre et al., 2011), so that, for example, the normal hearing infant is born with a preference for their mother's voice, underpinning a secure attachment relationship (Granier-Deferre et al., 2011; Thompson, 2008). A normal hearing cochlea is functionally mature by full term, partially facilitated by this in-utero stimulation. It is evident that the language which the foetus has been exposed to in-utero, affects perception of their native language and this has been demonstrated soon after birth (Moon et al., 2013). Moon et al. (2013) showed that, for example, infants respond differently to native vs non-native vowels (the English /i/ or Swedish /y/) at birth. This indicates that development of the neural

structures for processing speech sounds starts prenatally, as the auditory system begins to lay down the neural structures for processing speech sounds.

The organisation and development of cortical auditory pathways is driven by timely experience of signals from the cochlea, so that these pathways are refined. If sensory input is reduced or absent during these early developmental periods, the functional properties of the neurons can be reduced or degraded (Sininger et al., 1999). This implies that a delay in functional organisation can be already present when a hearing impaired child is born because they would not have had comparable levels of auditory stimulation as a normal hearing infant. Therefore, the effective hearing age for hearing speech sounds may be delayed even more than is often realised. This detrimentally affects the development of the auditory system.

In addition to language delay arising from reduced auditory stimulation for an infant born with hearing loss, there are also important differences in physiological aspects of the impaired cochlea. Sensorineural hearing loss (SNHL) causes poorer detection thresholds but also reduces frequency resolution in the cochlea (Khanna & Leonard., 1982; Govaerts et al., 2006). It has been established that cochlear hearing loss reduces ability to resolve components in the frequencies of complex tones (Moore & Moore., 2003). Cochlear hearing loss may involve damage to, or loss of, function of the outer hair cells (OHCs) and inner hair cells (IHCs).

Damage to the OHCs results in a reduced sensitivity to weak sounds (elevated thresholds), reduced frequency selectivity and a reduced dynamic range (Moore & Oxenham., 1998; Stenfelt., 2008). Impaired frequency selectivity reduces a hearing-impaired child's ability to discriminate between sounds, making it difficult for them to hear different speech sounds and therefore, impairs speech perception. If there is damage to the IHCs, this results in less efficient stimulation of the auditory nerve. IHCs at certain places on the basilar membrane can be damaged or missing, and these areas are called "dead regions". Vibrations that occur within dead regions are not detected by the appropriate neurons. Moore (2001, 2004) has suggested that dead regions are likely to be present at frequencies where thresholds are greater than 90 dB HL, and that this this

reduces intelligibility of speech because of the distortion caused by off-frequency listening in neighbouring hearing regions.

Thus, a hearing impaired child may be able to detect a sound with hearing aids but due to the physiological characteristics of the cochlea, the child may not be able to discriminate between speech sounds as well as normal hearing peers, thereby constraining speech perception. Although hearing aids can improve detection thresholds, they cannot improve poor frequency discrimination, giving rise to upward spread of masking and poorer speech understanding in noise. Thus, despite early intervention and the fitting of hearing aids with new technology, the devices cannot provide the same quality of sound as in a normal hearing cochlea. Therefore, the foundations for spoken language development depend on the clarity of sounds being conveyed to the brain. The benefits of early detection of hearing loss are reliant, not only on timely identification of hearing impairment, including the type and extent of hearing loss but also by the quality of hearing aid technology and fitting.

The age of intervention for a child is not taken from the date of diagnosis but the date of effective hearing aid fitting (i.e. amplification provided). Watkin et al. (2007) reported on a study of 120 children with PCHI. This group was made up of 41 children who were enrolled in the audiological management plan but only 29 were fitted with hearing aids at the age of 9 months. The study indicated that children with PCHI managed before the age of 9 months, had higher adjusted language scores at 5-11 years of age, compared with children managed later than 9 months. Wake et al. (2005) found no relationship between age of identification or age at intervention on subsequent speech outcomes. This may be because their mean diagnosis was 18.5 months with amplification occurring at 19.5 months. The relatively late age of diagnosis for this group and the later intervention implies a reduced opportunity to benefit from an early management plan and would lead to a delay in timely stimulation of pathways. This may explain the findings that there was no significant relationship between age of diagnosis and outcomes with the Peabody Picture Vocabulary test (Dunn & Dunn 2007). However, Sininger et al. (2010) reported that age of fitting of amplification was the most important contributing factor in predicting speech outcomes. The mean age of fitting amplification in this study was 5.7 months, (13.8 months less

than Wake et al., 2005). These studies demonstrate that after diagnosis of hearing loss, immediate intervention is necessary for maximum benefit. Children with earlier access to speech signals through amplification are more likely to have better outcomes.

The age at which effective amplification is given is an important factor for management of hearing loss in children. It illustrates the importance of early intervention and supports the relevance of critical periods for auditory stimulation (Yoshinaga-Itano et al. 1998; Kennedy et al. 2006; Moeller 2000). Several authors indicate that the critical period for speech and language development is below 3.5 years (Kral et al., 2001; Sharma et al., 2002; Nicholas & Geers 2006).

It is known that infants with hearing loss are slower to develop spoken language than their peers with normal hearing (Moeller et al., 2007). The Eilers & Oller (1994) study described the onset of canonical babbling range in normal-hearing infants from 3-10 months. However, the onset of canonical babbling in hearing impaired infants ranged from 11-49 months. Thus, for children with hearing loss, the age at which canonical babbling starts is delayed and has reduced complexity, when compared to normal hearing children. Moeller et al. (2007) reported that infants with mild-profound hearing-impairment typically started their canonical babbling stage later than 21 age-matched infants with normal hearing. Even when amplification is provided to increase audibility of speech sounds, hearing-impaired babies show delayed speech and language and reduced frequency and complexity of vocal productions (Nott et al., 2009; Moeller et al., 2007; Oller & Eilers., 1988). The delays may arise partly from the lack of auditory stimulation in-utero. However, it is likely that a larger contributing factor is the lack of auditory stimulation following birth which leads to poorer development of neural networks.

### **1.1.3 The factors that affect the provision of adequate early auditory stimulation**

#### **1.1.3.1 Degree of Hearing Loss**

Several studies have demonstrated a relationship between the degree of hearing loss and language outcomes. Language delay increases with degree of hearing loss (Wake et al., 2004; Nicholas & Geers 2006; Fitzpatrick, 2007). Wake et al. (2005) found that the severity of hearing loss was the greatest predictor for speech and language ability in hearing impaired children aged at 7-8 years old; a finding reported by Fitzpatrick et al. (2011) in children of four to five year olds. Slinger et al. (2010) reported that severity of hearing loss was the second most important factor relating to speech and language outcomes; the first being age at fitting of amplification. The study also reported that level of hearing loss also affected the age of identification, milder losses were identified later than profound hearing losses.

Yoshinaga-Itano & Sedey (1998) showed that the degree of hearing loss was a significant predictor of the rate at which consonant and vowel inventories develop in hearing impaired children. Their study tested 147 children aged between 14-60 months to investigate the relationship between speech production and other demographic and developmental factors, one of them being the extent of hearing loss. The results showed that children with moderate-severe hearing loss developed their speech ability at a slower rate. The children with this degree of hearing loss lagged behind by approximately 1 year but by around 5 years of age their speech skills become age appropriate. The speech skills of those with profound hearing loss, however, never caught up and remained poor.

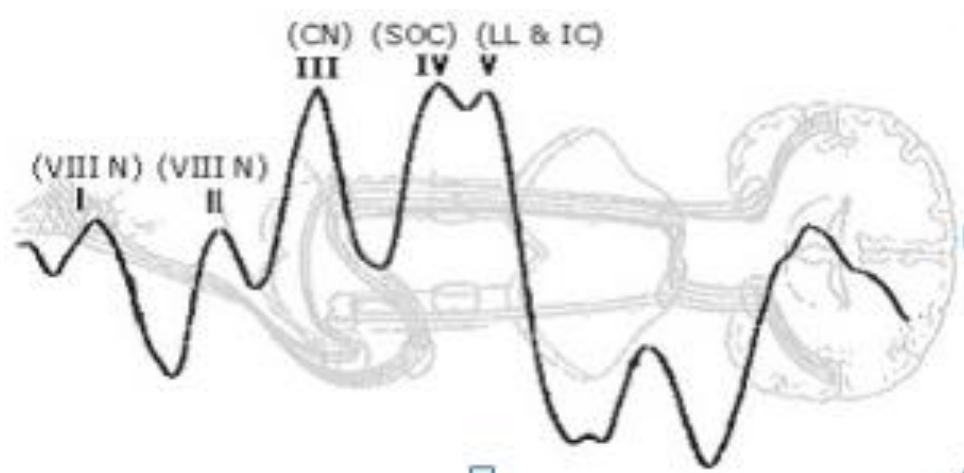
As the degree of hearing loss increases, the speech intelligibility decreases (Ching et al., 2013). This underlines the importance of determining accurately the type, degree and configuration of hearing loss of the individual child. It is essential to know the extent and nature of a child's residual hearing so that appropriate hearing aid selection and fitting can be made. Aiding infants with moderate or greater degrees of bilateral hearing loss has been standard practice for most clinicians (McKay et al., 2008).

Due to the poor sensitivity of the screening children with a milder level of hearing loss may not be detected. This does not mean that these children do not require intervention and support. In the past audiologists have not been overly concerned about these children with a milder level of hearing loss as they felt they were able to still obtain auditory stimulation (Porter et al., 2013; Wake et al., 2016). Đoković et al. (2014) compared 144 children with mild bilateral hearing loss with no aided experience and identified at 7-11 years of age, to a control group of 160 children with normal hearing. The results demonstrated the children with a milder hearing loss performed more poorly than the control group on measures of morph syntax and phonological short-term memory. Fitzpatrick et al. (2010) conducted a retrospective review of audiological services, between 1990-2006 for children with mild bilateral and unilateral hearing losses. They reported that 91.4% received a recommendation for hearing aid fitting and fewer than two thirds wore their hearing aids consistently. They did not report on speech or language outcomes in this study. Also, children with a mild hearing loss in this study were identified at 51.1 months. Their findings revealed uncertainty related to clinical recommendations of intervention for this population of children. The limitations of past studies have been that they classified mild hearing losses in the same category as unilateral hearing loss. Moreover, identification of these children is very late. Therefore, it has been difficult to ascertain the expected outcomes for mild hearing loss and what the clinical recommendations for them should be. Children with mild hearing loss compared to a unilateral hearing loss may have different difficulties and therefore, investigating them together could potentially alter findings. Audiologist require a method to demonstrate if these children with this level of hearing loss require aiding or intervention.

#### **1.1.3.2 Objective measures of infant hearing**

Objective measures are used to determine hearing levels in infants and children who cannot provide accurate and consistent behavioural responses. They can be used to determine the type, degree and configuration of hearing loss. The most accurate way of assessing hearing in newborns is with electrophysiological testing (Rodrigues & Lewis., 2010). In a normal auditory system, small electrical currents are generated by the process of hearing; electrophysiological testing

picks up and records some of these electrical responses. To record the responses, small surface electrodes are attached to the head at several places. A source of stimulation is placed in the ear via either an insert or headphones to present a specific sound source and the electrical signals across the electrodes are recorded. This generates certain neurological “markers” or patterns of responses as the hearing nerves respond. A neurological marker will only be apparent when the time-locked neural response is stimulated; therefore, different frequencies and intensities are used to generate a visible pattern of response (Figure 1.1). (<http://hearing.screening.nhs.uk/audiology>)



**Figure 1.1** The neural pathway for the auditory brainstem responses indicating where they are generated (Hall, et al., 1992). I (VIII N)=Cochlear, II(VIII N)=8<sup>th</sup> nerve, III (CN) = cochlear nucleus, IV (SOC) = Superior olivary complex, V (LL & IC) = Lateral lemniscus & Inferior colliculus

The NHSP guidelines (BSA: Auditory Brainstem Response (ABR), 2013) recommend using ABR or Auditory Steady State Responses (ASSR) and a two stage Transient Otoacoustic Emissions (TEOAE) test to screen the cochlear function (<http://hearing.screening.nhs.uk/audiology>; Baldwin & Watkin., 2012; Rance et al., 2005; Stapells et al., 1995). Otoacoustic Emissions (OAE) are not recorded by electrodes placed on the skin but by a microphone that sits in the ear canal. Perception of sounds are generated by the inner ear when the cochlea is stimulated with a sound. When a sound enters the cochlea, the basilar membrane vibrates causing the OHCs to move and in turn their stereocilia to be deflected, this causes an electromotility feedback loop which amplifies the

response of the basilar membrane and is considered to produce the energy that results in the emissions that are detected by the probe. (Kemp., 1978; Hamdan et al., 2008, Fettiplace. 2017). The objective measures recommended by NHSP, allow the main speech frequency range to be tested. Additionally, the protocol recommends testing the middle ear function with tympanometry, to determine the middle ear status and if there is a conductive component to the loss (Baldwin., 2006; Feldman., 1975; BSA Recommended Procedure Tympanometry, 2013).

### 1.1.3.3 Objective measures give an approximation of hearing levels.

The thresholds obtained via objective detection techniques (ABR/ASSR) are used to estimate behavioural thresholds in children with PCHI. However, it is important to determine how accurate the ABR/ASSR thresholds are in predicting an individual child's behavioural results. The ABR thresholds are used by audiologists for the initial fitting for amplification thus it is very important for this to be accurate.

A systematic review and Meta-Analysis by Stapells (2000) included 32 studies and demonstrated that tone-pip ABR thresholds in infants and young children with SNHL are typically 10 dB nHL lower or 10 dB nHL higher than pure-tone behavioural thresholds (Table 1.1).

**Table 1.1** Tone pip ABR. Results from Stapells (2000) meta-analysis showed mean elevation of the tone-pip ABR thresholds (dB nHL) over the pure-tone behavioural thresholds.

	Mean (95% CI of population mean) of difference between tpABR and behavioural thresholds (Stapells 2000)			
Subject group	0.5 kHz	1 kHz	2 kHz	4 kHz
Adults (normal hearing)	20.4 (18.8-21.9)	16.2 (14.9-17.4)	13.4 (12.3-14.4)	11.8 (10.7-12.8)
Adults (sensorineural)	13.4 (11.0-15.8)	10.3 (8.4-12.1)	8.4 (6.3-10.3)	5.2 (2.4-8.0)



Infants/young children (normal hearing)	19.6 (18.8-20.5)	17.4 (16.0-18.7)	13.6 (11.8-15.5)	15.5 (14.1-16.8)
Infants/young children (sensorineural)	5.5 (3.0-8.0)	4.9 (2.4-7.3)	0.6 (-1.6-+2.7)	-8.1 (-12.1—4.1)

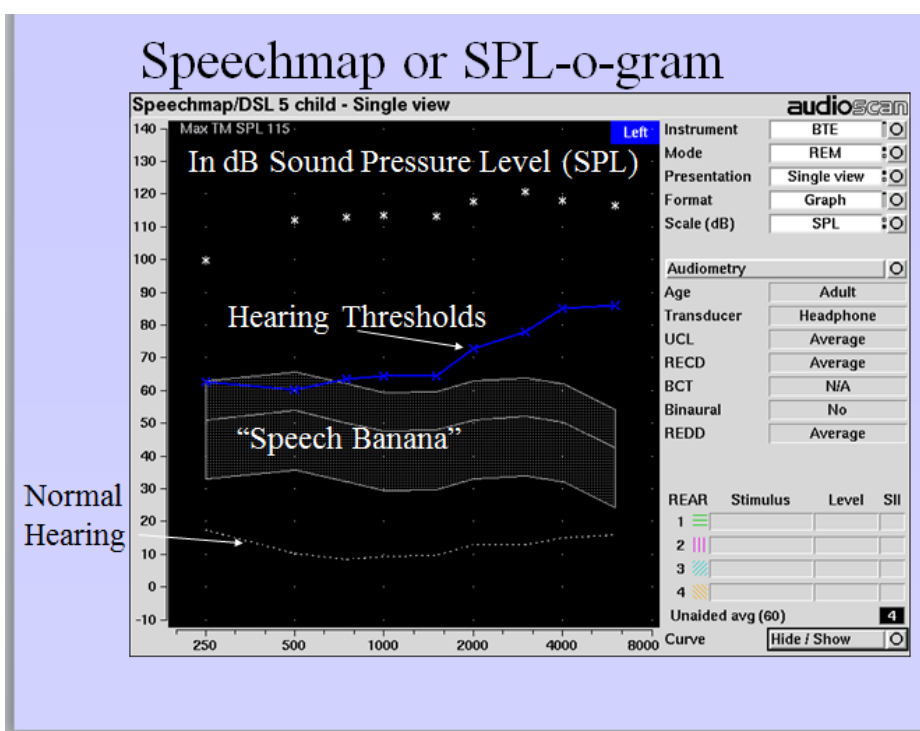
The confidence intervals for predicting absolute behavioural threshold from ABR results are large. It is known that 5% of babies will have true thresholds better than the lower limit of the confidence level given in Table 1, potentially leading to over-amplification where a hearing aid is fitted (BSA Guidelines for the early audiological assessment and management of babies referred from the Newborn Hearing Screening Programme, 2013). This emphasises the importance of using additional behavioural and subjective measures.

Once the degree of hearing loss is established, especially if it is a PCHI, it is essential that a management plan is put in place. The aim of intervention is to enhance access to sound through the use of hearing aids. The aim of the hearing aid is to improve audibility of speech information. The extent of the access to speech due to amplification depends on the thresholds entered into the hearing aid software. There is a known measurement error for pure tone audiometry so the thresholds can be wrong by approximately 10 dB HL, which can potentially lead to over or under amplification (Stevens et al., 2013; Feirn et al., 2014).

#### **1.1.3.4 Problems of procedural fitting of hearing aids using REMs**

The last decade has seen numerous improvements in hearing aid technology, producing sophisticated digital hearing instruments. New hearing aids have improvements in noise reduction by employing directional microphones (Ricketts & Galster., 2008; Ricketts et al., 2007), and other features such as feedback cancellation which does not reduce important speech cues, (Pittman., 2011; Stelmachowicz et al., 2010). Most recently, frequency lowering technology has been used to transpose high frequency sounds such as /s/ and /sh/ to lower frequency regions for patients with high frequency hearing loss (Glista et al., 2009; Wolfe et al., 2009).

There are specific protocols applied in the UK for hearing aid fitting and personalisation to the child's hearing needs. Audiologists are expected to verify the output of the hearing aid in the child's ear through real ear measurement (REM). REM requires the use of a probe microphone measurement in the ear canal to take into account the acoustic effect of an earmould in the ear. REM is the method of choice to ensure that the hearing aid amplification levels matches the targets specified by prescription formulae and predictable levels of amplified speech (Cox et al., 1990). One of the prescription methods used by clinicians is the Desired Sensation Level (DSL) (Seewald et al., 1993; Scollie et al., 2005) or National Acoustic Laboratories nonlinear fitting procedure NA-NL2) (Dillon & Storey., 1998; Keidser et al., 2011). Use of a published prescription method like DSL or NAL provides clinicians with a systematic, evidence-based approach to paediatric hearing instrument fitting that ensures potential audibility of amplified speech (Seewald et al., 2005). The prescriptive targets are derived from accurate hearing threshold measurements for each ear. The DSL prescription plots the converted audiogram in decibel sound pressure level (dB SPL) on an audiogram-like format called the SPL-o-gram (see Figure 1.3). The advantage of the SPL-o-gram approach is that it allows a close examination and understanding of the important interrelationship between hearing levels and amplification characteristics, facilitating the aural habitation/rehabilitation process (Seewald et al., 2005).



**Figure 1.2** Screenshot of speech mapping of SPL-o-gram. The blue line is the hearing threshold and the area labelled “speech banana” is the region where speech phonemes fall in this case they are not audible. (DSL: [www.dslio.com/page/en/dsl/history.html](http://www.dslio.com/page/en/dsl/history.html))

Real ear verification requires multiple measurements to ensure that speech is audible over a range of different levels of stimulus. As this method needs time and cooperation, REM is not a practical or feasible method of choice in very young children (Bagatto et al., 2005). An alternative method is by the recording of the real-ear-to-coupler difference (RECD). This method only uses a single probe microphone measurement with the child’s earmould in place. The ear-canal response is compared to 2-cm<sup>3</sup> coupler (2CC coupler which acts like an average adult ear canal). The difference is used to estimate the response of the hearing aid in the child’s ear for verification which can then be completed in the 2CC coupler. The values vary depending on age and frequency. This is an important measurement because research has shown that even age-appropriate average RECD values collected from a sample of children are not as accurate for use as individual measures (Bagatto et al., 2010; King 2010). The RECD allows the audiologist to accurately convert threshold information collected with insert phones from dB HL to dB SPL (ear canal level) for use within the hearing aid selection and fitting process. It also allows the audiologist to determine the difference between the output in the real-ear and the output in the 2CC coupler used in the hearing aid fitting and verification process. This strategy, however, may not be easily implemented while attempting to acquire an RECD measurement on a young infant due to them being uncooperative or noisy. Although there are age-related average RECD values available in cases where the response cannot be reliably measured on the child (Bagatto et al., 2002). Despite the common goal of these methods to integrate individual ear-canal acoustics into the hearing aid verification process, the variability could influence the consistency of the hearing aid fitting. Furthermore, a key component to measuring the RECD is accurate placement of the probe-tube in the individual’s

ear canal (Bagatto, 2006). This can be difficult in moving and active children and can result in inaccurate targets.

The aim of this process and fitting to prescribed gain targets is to ensure that adequate amplification is given to the child for their specific hearing loss and that the speech spectrum is both audible and comfortably loud. However, reliance on the hearing aid fitting to the targets alone, does not directly reflect how much of an average speech signal is audible to a child through their hearing aids (McCreery et al., 2013). The hearing threshold levels may be inaccurate, particularly if derived from objective ABR testing in the first weeks of life (as discussed in section 1.1.3.3). If the hearing thresholds are inaccurate then the DSL/NAL targets that are generated will also prescribe inaccurate targets meaning inadequate amplification resulting in poor speech discrimination. Signals which are inaudible cannot be detected, discriminated, recognised or learned especially in the real-world where listening can be highly complex. Quantifying the adequacy of hearing aid fitting is challenging in infants. McCreery et al. (2013) looked at hearing aid fitting data for 195 children with mild-severe hearing losses and found that 55% of children in the study had at least one ear that deviated from prescriptive targets by more than 5dB on average. This indicates that some other means of evaluating adequacy of the hearing aid fitting is required to ensure that speech is audible.

#### **1.1.3.5 Evaluation of Hearing Aid fitting**

For a reliable PTA to be achieved, behavioural confirmation of hearing thresholds is necessary. In the first few months of life there are challenges in assessing hearing using behavioural responses. At present the recommended protocol for UK NHS audiologists, in the first six months of life is to use estimated thresholds derived from ABR tests or ASSR to fit hearing aids ([www.hearing.screening.nhs.uk/audiology](http://www.hearing.screening.nhs.uk/audiology)). Therefore, hearing aid fittings are typically based on the best estimate, but incomplete hearing threshold information. This can result in audiologists fitting the aids “conservatively” and giving less amplification than required to avoid over amplification. This leads to a reduction in the quality of the sound input through the child’s hearing aids, so that not all speech sounds are audible and distinct to the infant.

Early assessment is reliant on objective testing until behavioural testing is feasible at approximately six to eight months of age when Visual Reinforcement Audiometry (VRA) becomes developmentally appropriate (BSA recommended procedure for Visual Reinforcement Audiometry BSA, 2014). VRA is a classical-conditioning technique which capitalises on a child's natural inclination to turn towards a sound source. It is routinely used with children aged from 6-8 months and upwards to assess hearing levels and is shown to be a reliable and efficient procedure at this young age (Thompson & Wilson., 1984; Sabo., 1999). This method of testing is not appropriate for children less than 6 months old because the child need to be able to sit unsupported and be able to turn their head side to side. Therefore, by the time a hearing impaired child is able to demonstrate behavioural responses via VRA in clinic, they may have had sub-optimal stimulation of their auditory pathways, if inadequate amplification (based on estimated thresholds) was provided.

#### **1.1.3.6 Types of intervention: cochlear implantation vs hearing aids**

For children with profound hearing loss, acoustic hearing aids are unable to give sufficient audibility of speech. For these children, cochlear implants are an alternative way of delivering sound to the auditory system. A number of studies have demonstrated that cochlear implant users have more effective speech perception, production and spoken language skills than acoustic hearing aids (Tomblin et al., 1999; Dettman & Dowell., 2010; Moog & Geers, 2003). Sininger et al. (2010) demonstrated better outcomes in speech perception and speech production in 16 children who used cochlear implants. They reported that for both speech production and expressive and receptive language, cochlear implant users had significantly improved outcomes. Having a cochlear implant was associated with an increase in speech production by 0.64 standard deviations and with 12 months improvement in receptive and almost 18 months improvement in expressive language. This shows that cochlear implants provide better auditory input than traditional hearing aids for severe-profound hearing loss. Many studies have shown that the fitting of cochlear implants for infants who are profoundly deaf has allowed them to develop their spoken language. However, Fitzpatrick et al. (2011) studied 88 children; 26 children used cochlear

implants, 25 used hearing aids and 37 had normal hearing and all were aged between 4 to 5 years of age. The degree of hearing loss ranged from mild to profound, the results indicated those with cochlear implants did not obtain better results on outcome measures than children who used hearing aids. There are several reasons why Sininger et al. (2010) demonstrated better outcomes than Fitzpatrick et al. (2011). Firstly, there was a greater number of cochlear implant users enrolled in the Fitzpatrick et al study. Secondly, fewer children with mild and moderate-severe hearing loss were enrolled in the Fitzpatrick et al study; 35.3% enrolled versus 65.9% in Sininger et al. As mentioned previously, greater severity of hearing loss is associated with lower levels of spoken language performance. Hence, the study by Fitzpatrick et al demonstrated worse communication outcomes in his cohort because the study had a greater proportion of children with severe-profound hearing loss. Studies have shown that degree of hearing loss is a predictor for the benefit of cochlear implants. No matter how early intervention is initiated, the level of speech perception ability provided by hearing aids to profoundly deaf children is rarely sufficient to support normal rates of spoken language development (Geers et al., 2007). The results showed that cochlear implantation was likely to be clinically effective for children with a 4-frequency pure tone audiometry (PTA)  $\geq 80$  dB HL. (Lovett et al., 2015). Recent studies have also demonstrated that cochlear implants should be fitted before the age of 12 months. Results have shown that children under the age of 12 months show better language development compared with children who receive their cochlear implant between 13 and 24 months (Holman et al., 2013). This supports the provision of a cochlear implant within the first year of life to ensure that a child with severe-to-profound hearing loss will commence school with age appropriate language skills (Vlastarakos et al., 2010; Leigh et al., 2013).

The studies emphasise that careful fitting of hearing aids is essential. If adequate amplification is not achieved the quality of the sound stimulation is compromised and this can have an impact on the child's development of speech and language.

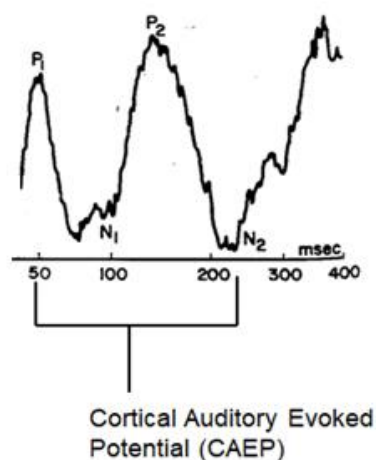
#### **1.1.3.7 Hearing aid usage**

A recent study by Walker et al. (2015) investigated hearing aid use on 290 children with mild to severe hearing loss and they found that data logging values

were lower than parental report, suggesting that parents overestimated daily hearing aid use. They also found that maternal education levels influenced longitudinal trends in daily hearing aid use. They found that children whose mothers had a bachelor's degree were approximately 14 times more likely to be routine users, compared to mothers who had some college education. One of the limiting factors of this research was some data on hearing aid usage was collected from parental report rather than objectively from the hearing aid. This was due to the sample size not having enough data. One of the recommendations from their study was that any future research should incorporate data logging to investigate hearing aid use. Munoz et al. (2015) looked at data logging for 29 children (7 months to 6 years of age) and revealed large variability in hours of hearing aid use, with an increase in hours of use with age and severity of hearing loss. Data logging is a reliable way to estimate the child's hearing aid use and does not have any adverse effects on the child. Walker et al. (2013) in an earlier study collected data logging information on 133 children, ranging in age from 8 months to 8 years. Nineteen percent wore hearing aids for 4 hours or less per day and 12% used their hearing aids for more than 12 hours per day. The research found that longer hearing aid use related to older age, those with poorer hearing, and higher maternal education. Their results suggested that families benefit from counselling. Their research highlighted that counselling benefitted consistent hearing aid use especially for those with milder degrees of hearing loss and for families with lower levels of education. Moeller et al. (2009) correlated higher maternal education and SES to higher device usage and this could be down to parental understanding and knowledge about potential impact. McCreery et al. (2014) reported that additional counselling for families with lower educational levels should be given so they can understand the relationship between consistent hearing aid use, learning, and brain development. Sjoblad et al. (2001) demonstrated that 65% of parents in the early stages of the hearing device fitting questioned aided benefit and 30% of parents requested better education around the benefit that the aids provided to their child. This shows the importance of parental participation in the hearing aid pathway.

#### 1.1.4 The availability of Cortical Auditory Evoked Potential recording to assist in hearing aid management in infancy

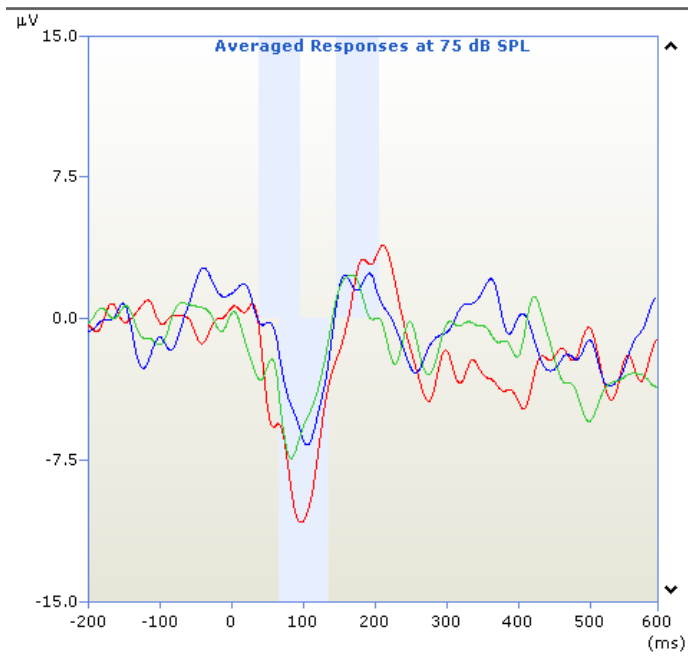
One method for assessing aided hearing thresholds in children under 6 months old is to use electrophysiological techniques. These include testing such as aided ABR or ASSR tests and Cortical Auditory Evoked Potentials (CAEP). CAEPs are a specific type of electrophysiological testing, which records neural responses generated at higher levels of the auditory pathway. The responses originate from neurons at the level of the primary auditory cortex, and from the auditory association areas in the temporal lobe (Cone-Wesson et al., 2003). The latency and amplitude characteristics of CAEP responses are determined primarily by the acoustic parameters of the stimulus and the integrity of the primary auditory pathway. There are three major components in the recorded response referred to as the P1-N1-P2 complex (Cone-Wesson et al., 2003). For an adult, the response waveform is characterized by a small positive peak (P1) about 50 ms after stimulus onset, a large negative peak (N1) about 100 ms after stimulus onset, and a second large positive peak (P2) about 180 ms after stimulus onset (Figure 1.3). The shape is very different, and more variable, for infants, often comprising just a single broad peak around 200 ms after stimulus onset (Figure 1.4). The shape changes as the auditory cortex matures, right through the teenage years up to early adulthood (Van Dun et al., 2016). Therefore, if the neural signals are reaching the auditory cortex it could be assumed that perception of sound is reaching the brain (Purdy et al., 2005).



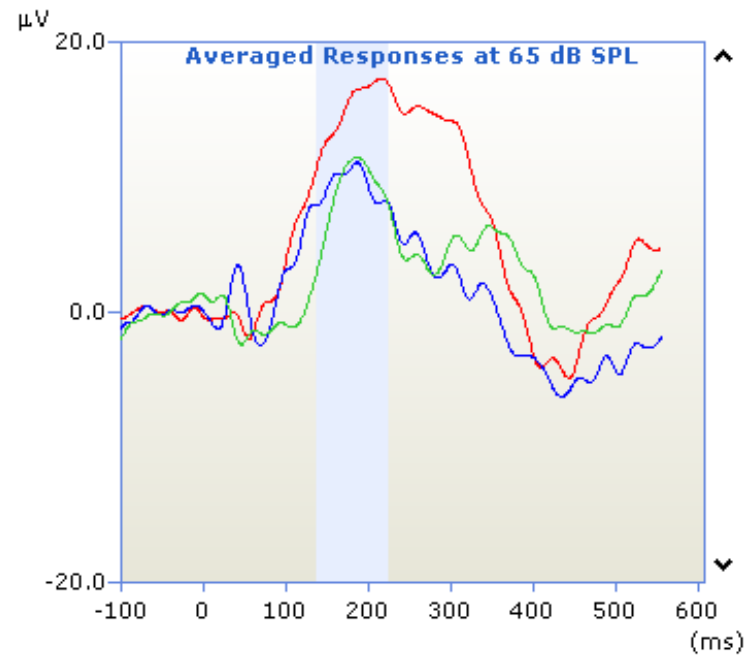
**Figure 1.3** A typical CAEP waveform (Katz. 2001).



A)



B)



**Figure 1.4** A) Example from HEARLab of CAEP waveform morphology of an adult B) Example from HEARLab of CAEP waveform morphology of an infant

There are several reasons why CAEP may be an appropriate test to be used for evaluating adequacy of hearing aid fittings. Firstly, CAEP can be recorded in response to a variety of complex stimuli such as clicks, tones and speech sounds (Dillon., 2005). Wunderlich et al. (2006) showed that using words as a stimulus resulted in evoking frequent and larger responses at all ages than the use of tones. Using speech sounds as stimuli allows the hearing aid to transmit the sounds in the same way as for everyday speech signals. Any other sounds used may be amplified differently by the hearing aids if recognised as noise e.g. they might be attenuated. The use of speech stimuli is more meaningful to parents as they can relate these to everyday speech. Secondly, the duration of speech sounds allows hearing aid amplification to be comparable to real life signals. The signals have a presentation time long enough to activate the compression circuits of a hearing aid, just like in continuous speech. This makes it an effective tool to determine if the amplification produced by the hearing aid provides a signal that is detectable at the cortical level. Thirdly, the responses are generated at the higher perceptual level of the auditory pathway. Therefore, any effects to the signal due to the hearing aid (i.e. any change in the frequency response of the signal) will be taken into account (Souza & Tremblay., 2006; Dillon., 2005; Hood et al., 1994). Fourthly, the response waveform appears irrespective of whether the person receiving the sound attends to the sound or completely ignores it and the responses can be recorded when the subject is awake making it easier to use on infants (Cone-Wesson et al., 2003).

Several research studies report that neural detection and audibility of aided and unaided sound can be assessed using aided CAEPs (Glister et al., 2012; KorczaK et al., 2005; Purdy et al., 2005). CAEP morphology changes are seen in neural activity associated with auditory deprivation and auditory stimulation (Ponton et al., 1999). The underlying assumption is that if a response is present, for a specific sound, then it must be audible (Purdy et al., 2008). CAEP can be used to look at the differences in the pattern of cortical responses to auditory speech signals, giving an indication of discrimination between the two sounds (Golding et al., 2007). KorczaK et al. (2010) tested consonant-vowel speech stimuli (/bi/vs/bu/, /ba/vs/da/ and /da/vs/ta/) showing vowels generate larger amplitude and earlier latencies than consonant stimuli which they interpreted as suggesting that the brain may find it easier to process vowel stimuli (Kurtzburg et al., 1986;

Tiitinen et al., 1993). Additionally, Kolkaila et al. (2012) and Tremblay et al. (2003) reported that speech CAEPs are as reliable as tone-evoked CAEPs. They showed consistent re-test reliability from the same individual and concluded that if there are any significant alterations in the morphology that this may be due to neural changes. CAEPs have been used to assess the impact of auditory deprivation and auditory stimulation on neural function over time.

The latency changes in CAEPs have been used to look at auditory system plasticity and recovery from auditory deprivation following cochlear implantation (Sharma & Dorman., 2005; Ponton et al., 1996). Cardon et al. (2012) has shown early intervention with appropriate auditory input increases the likelihood of normal auditory cortical development in children with congenital deafness. Bakhos et al. (2014) conducted a pilot study on 8 to 12 year olds and looked at the relationship between CAEP responses and language ability. They demonstrated that children with hearing loss who had no language impairment showed normal CAEPs and children with hearing loss and impaired language showed atypical temporal patterns in their CAEPs. CAEP therefore, provides an insight into brain processing and many aspects of auditory perception (Stapells., 2009). Hence, this objective test offers possibilities for evaluating and validating audibility through hearing aids, ensuring that infants have access to speech via hearing aid amplification. Results from Billings et al. (2011) found conflicting results. They demonstrated no significant effect of amplification on latencies or amplitudes on the cortical responses measured with hearing aids, providing 20dB of gain compared to unaided responses. This could be due to the use of normal hearing participants who, it could be argued, hear internal noise from the aid which could affect the cortical results. The effects of amplification on CAEPs are largely unknown and have been one of the reasons why they have not been widely used.

One of the reasons for the lack of uptake of CAEPs clinically, for evaluating the effective fitting of hearing aids, has been due to the difficulty for clinicians to ascertain if a response was present. To address this problem commercial CAEP machines now include automated statistical algorithm techniques for determining the presence of a response, which are more reliable than previously used. Carter et al. (2010) and Golding et al. (2009) compared cortical responses using the

automated statistical algorithm (Hotelling T2) versus visual detection by experienced examiners. They found that the Hotelling T2 could differentiate a cortical response from random electrical activity at the level of an experienced examiner. The use of this method gives more certainty and reduces human error in the results obtained. Hotelling's T2 calculates the probability for each response (a "p-value") and automatically calculates the likelihood that a CAEP has been detected in response to the test signal.

Several studies have measured aided CAEPs in infants with hearing loss and have shown that a small proportion of these infants do not exhibit a strong CAEP even when the stimulus is audible. Two small-scale studies by Chang et al. (2012) and Van Dun et al. (2015) have shown that it was not possible to detect CAEPs in 30 to 40% and 22 to 28% of participants in Chang et al. and Van Dun respectively. In both these studies, participants were older (8 to 30 months) compared to the current study. A number of studies have demonstrated that CAEP detection rates increase with sensation level. A retrospective review of clinical data obtained using the HEARLab (Gardner-Berry et al. 2016) demonstrated that CAEP detection increased with sensation level; however, even at sensation levels greater than 20 dB, around 30% of children with SNHL do not have a detectable CAEP. Although this number of absent responses and increased sensation level seems high, this uncertainty is reduced by combining the results of the same subject for other speech sounds and intensities and using the CAEPs in combination with other tests. The current HEARLab ACA model cannot be used for threshold estimation, as the stimuli are presented in the free field and there are limitations to presentation level and stimuli. Furthermore, research conducted by Munro et al. (2011) showed that the HEARLab cannot be used to evaluate speech discrimination between different sounds. Their research showed that the speech stimuli /t/ and /g/ which are relatively similar in spectral content did not reliably result in different CAEP waveforms.

Despite this there is information showing that the relationship between pure tone audiometric and CAEP thresholds correlate well (Cody et al., 1967, Davis & Roberts., 1965, Tsu et al., 2002). Lightfoot and Kennedy (2006) identified that 94% of individual cortical electric response audiometry threshold estimates were within 15 dB of the behavioural threshold. These findings suggest that cortical

electric response audiometry has a performance that is as good as or better than the ABR for threshold estimation in adults. This strengthens and validates the use of CAEP in clinics, especially in children who are not able to give responses through behavioural testing. Furthermore, another benefit of CAEPs is that it can be recorded while the child is awake unlike ABR where audiologists have to either wait for natural sleep or use sedation methods.

#### **1.1.5 The value of the CAEP in pathway post NHSP**

In Australia CAEPs have been routinely integrated into the infant audiological pathway. Punch et al. (2016) reported a retrospective review of 83 infants with PCHI, fitted with hearing aids using evoked potential tests and prescriptive targets. The findings indicated that CAEP testing influenced the effectiveness of rehabilitation and that CAEP testing was well received by parents.

The CAEP can be used as a counselling tool for parents and families. The importance for parents and family members to see their child responding or not responding to an auditory stimulus, may be crucial in helping them through the process of diagnosis of deafness and towards auditory communication development. The absence of cortical responses when unaided can provide evidence to the parents of the importance of wearing hearing aids, illustrating the benefits, and showing them improvements in hearing when their child uses hearing aids. The results have implications for effective hearing aid management by adjusting hearing aid gain across frequency, to make sounds audible for communication at the appropriate time for auditory learning. At the early stage in audiology intervention, audiologists are primarily concerned about making speech sounds detectable. Therefore, it is important that hearing aids amplify speech so that it is audible at a comfortable sensation level and this can be directly tested by measuring cortical potentials evoked by speech stimuli. The development of CAEP equipment allows clinicians to view the child's access to speech sounds, aided and unaided, using relevant speech tokens. This can, therefore, be used to evaluate the effectiveness of individual hearing aid fittings. The presence of CAEP responses elicited by speech sounds at equivalent conversational levels has been shown to correlate well with how the infant hears in real life, as subjectively reported by the parents (Golding et al., 2007). Van Dun et al. (2016)

conducted a study with 12 normal hearing and 12 hearing impaired adults and looked at the detection of the speech sounds /m/, /g/ and /t/. The study recorded CAEPs and analysed the amplitudes of the waveform. They demonstrated that CAEPs can potentially be used to assess hearing aid gain in hearing-impaired users. Furthermore, cortical testing may provide information that is relevant to the decision whether or when to refer for assessment for cochlear implantation. Thus, the CAEP can be used to bridge the gap between hearing aid fitting at around 3 months and behavioural testing at around 7 months developmental age.

## **1.2 Aims of research**

Despite recent improvements in identification of hearing loss and implementation of early intervention even with provision of early amplification (Wood et al., 2015), children with hearing loss continue to experience delays in the acquisition of spoken language. The limitations of subjective testing in early life means that hearing aids are fitted on estimated audiograms. Inaccurate hearing aid fitting targets can lead to under-amplification and potential under-stimulation of the auditory system. To avoid delays in providing effective stimulation of hearing potential, the use of CAEP may play a role for evaluating hearing aid fittings for children under the age of six months and to demonstrate aided benefit to families.

The primary aim of this research was to determine the usefulness of the introduction of CAEPs in the NHSP pathway for children under six months of age. The study evaluated the worth of implementing the test on the use of hearing aids fitted to infants in a clinical service. The secondary aim of this research was to validate hearing aid fitting with the use of electrophysiological testing using CAEPs at a younger age with the objective of achieving timely amplification, showing aided benefit to clinics and audiologists. This aim is addressed in the first experimental chapter (Chapter 2). In the second experimental chapter (chapter 3) the aim was to investigate parental perspectives on the value of CAEP recording and to highlight factors that influenced parental uptake of hearing devices for their infant in the period immediately following their child's diagnosis of deafness from the NHSP. The aim of the final experimental chapter (chapter 4) was to obtain clinicians' views on the use of CAEP in the clinical pathway to find out how it is being used around the world and with which populations.

## **CHAPTER 2: The Role of Cortical Auditory Evoked Potentials (CAEP) in reducing the age at hearing aid fitting in Children with Hearing loss identified by Newborn Hearing Screening.**

### **2.1 Abstract**

Recording of free-field cortical auditory evoked potential (CAEP) responses to speech tokens was introduced into the audiology management for infants with a permanent childhood hearing impairment (PCHI) in 2011-2015 at a UK service. Children with bilateral PCHI were studied from two sequential cohorts. Thirty-four children had followed an audiology pathway prior to CAEP introduction and forty-four children followed a pathway after the introduction of CAEP and were tested with unaided and aided CAEPs. Data analysis explored the age of diagnosis, hearing aid fitting and referral for cochlear implant assessment for each of these groups. CAEP offered a novel educative process for the parents and audiologists supporting decision-making for hearing aid fitting and cochlear implant referral. Delays in hearing aid fitting and cochlear implant referral were categorised as being due to the audiologist's recommendation or parental choice. Results showed the median age at hearing aid fitting prior to CAEP introduction was 9.2 months. After the inclusion of CAEP recording in the infant pathways it was 3.9 months. This reduction was attributable to earlier fitting of hearing aids for children with mild and moderate hearing losses, for which the median age fell from 19 to 5 months. Children with profound hearing loss were referred for cochlear implant assessment at a significantly earlier age following the introduction of CAEP. Although there has also been a national trend for earlier hearing aid fitting in children, the current study demonstrates that the inclusion of CAEP recording in the pathway facilitated earlier hearing aid fitting for the milder-impairments.

### **2.2 Introduction**

Universal neonatal hearing screening (UNHS) is well established within the health care system in the United Kingdom (UK) and in many other countries across the world. The introduction of UNHS has successfully reduced the median age at diagnosis of permanent childhood hearing impairment (PCHI) to 3

months or less from 2.75 years (Kennedy et al., 1998; Uus and Bamford, 2006; Watkin & Baldwin, 2012). The Hearing Outcome Project (HOP; Kennedy et al., 2006) reported that UNHS was associated with better language scores for hearing impaired children. The early management of hearing loss through hearing aid fitting is considered an essential initial step for improving the communication and auditory abilities of children born with PCHI (Yoshinaga-Itano et al., 1999). However, speech and language outcomes still vary widely for these children due to a number of child and family factors (Ching et al., 2013). Child factors include non-verbal intelligence, educational input, additional disabilities, the aetiology and severity of deafness, and the age at which amplification (hearing aids and/or cochlear implant) was provided. Sininger et al. (2010) reported that the age at fitting of amplification had the single largest effect on outcomes. The quality of hearing aid provision in the UK was transformed by the Audiology Modernisation project (Bamford et al., 2004; 2005). However, delays to hearing aid provision and cochlear implant referral remained although many researchers have highlighted the importance of early cochlear implantation in children to improve speech and language outcomes (Ostojić et al., 2011; Geers et al., 2013; Ching et al., 2013).

Family factors include participation in rehabilitation, mother–child interaction, maternal education level, SES and communication mode. Watkin et al. (1990) demonstrated that parents often had difficulties recognising the impact of hearing impairment through observation of their infants in the home, especially for mild or moderate impairments. They concluded that the delays in hearing aid fitting and consistent use could in part be attributable to the lack of parental awareness of the hidden difficulties their infants have in hearing speech. Further support should be provided to demonstrate difficulties and improve parental understanding.

Cortical Auditory Evoked Potentials (CAEPs) give an objective measure of a response to auditory stimuli, including speech sounds. The responses originate from neurons at the level of the primary auditory cortex, and from the auditory association areas in the temporal lobe (Cone-Wesson et al., 2003). The latency and amplitude characteristics of CAEP responses are determined primarily by the acoustic parameters of the stimulus and the integrity of the primary auditory



pathway. There are three major components in the recorded response referred to as the P1-N1-P2 complex (Cone-Wesson et al., 2003). The response can be recorded in infants who are too young to respond behaviourally both unaided and also with hearing aids on to assess improved detection of speech stimuli (Barnet, 1971; Dillion, 2005; Korczak et al., 2005; Purdy et al., 2005; Glister et al., 2012). The assumption is that if a CAEP response is present for a specific sound, then it must be audible (Purdy et al., 2008; Stapells, 2009). CAEP responses have been recorded in cases of auditory neuropathy spectrum disorder (ANSD) when early latency electrophysiological responses (ABR responses) are absent (Pearce et al., 2007). Rance et al., (2002) showed CAEPs to be present for children with ANSD who had open-set speech perception ability and benefit from amplification.

CAEPs are not always consistently present in hearing impaired children. Van Dun et al. (2012) showed that around 25% of hearing impaired children do not evoke a response to speech stimuli presented at 10 dB sensation level (SL) or 10 dB above auditory threshold. This may indicate that late latency evoked potentials are less stable than the early evoked potentials (Carter et al., 2010). However the infant's state of arousal is known to affect the morphology and detection of a CAEP response (Suzuki et al., 1976) and therefore they are not always detectable even at low and medium sensation levels when it would be expected that the sounds are audible. This highlights the importance of normative data, carefully controlled recording conditions and confidence measures for intensity levels and different types of stimuli. With this, CAEPs may have a role for systematically evaluating aided and unaided responses. Research undertaken by National Acoustic Laboratories used short speech sounds with low-, mid-, and high-frequency content, presented in the free field, to record unaided and aided hearing in infants as part of their clinical assessment (Dillon, 2005; Pearce et al., 2007; Chang et al., 2012).

Audiologists face a challenge when recommending hearing aids for milder degrees of hearing loss (Bagatto et al., 2013; Fitzpatrick et al., 2014) and it can be difficult to confidently prescribe hearing aids without behavioural testing which is typically conducted from around 6 months of age. The CAEP response to speech sounds can be recorded from 3 months and therefore potentially offer earlier confirmation of an infant's access to speech sounds.

The implementation of CAEP using the HEARLab device began in Australia in 2011. Punch et al. (2016) report a retrospective review of 83 infants with PCHI, fitted with hearing aids using evoked potential tests and prescriptive targets according to their national protocol. In addition aided CAEP responses were recorded within 8 weeks of the initial fitting. CAEP results were used to confirm unaided hearing capability and to modify the initial hearing aid fittings. Their findings indicated that CAEP testing influenced the effectiveness of rehabilitation and that CAEP testing was well received by parents. This led to integration of CAEP testing into their routine infant fitting program.

In recognition of the potential contribution of CAEP testing CAEP recordings were incorporated into the infant audiology pathway at one of the London Hospitals. CAEP recording was undertaken for infants after a PCHI had been confirmed by diagnostic auditory brainstem response (ABR) and/or auditory steady-state response (ASSR) assessments but before hearing aids were fitted.

The goal of the current research was to determine the impact on patient management of introducing CAEP assessments into the infant audiology pathway for infants. Specifically the age of hearing aid fitting and cochlear implant referral were analysed and influential factors of family engagement, audiologist decision making and extent of hearing loss were explored.

## **2.3 Methods**

### **2.3.1 Research and Development Approval**

The study was registered as a clinical service evaluation with a NHS Trust in East London research and development department: registration number 1275. It was a cohort comparison study comparing a retrospective group, cohort A and a prospective group, cohort B and following the pre-school audiological management.

### 2.3.2 Participants

The study was undertaken in a paediatric audiology service in East London. Data were analysed from children who were referred from the NHSP (Newborn Hearing Screening Programme) hearing screen and who were identified with a PCHI between 2008 and 2015. A review of data from diagnosis of PCHI to school entry (at 5 years) was conducted. At the time of the analysis the youngest child in the study with PCHI had reached 2.3 years of age. Children from all ethnic backgrounds and all spoken languages were included. Children with a primary diagnosis of ANSD, those with long term mixed hearing loss due to otitis media or with additional learning or sensory disabilities and comorbidities were excluded from the current analysis. This was to remove cases with known progressive or fluctuations in hearing loss following the recording of CAEP results. The study sample was divided into two separate cohorts based on the date of their audiological assessments. Cohort A consisted of babies born from January 2008 to August 2011 when the clinical pathways did not include the recording of CAEP measurements and Cohort B consisted of babies born between September 2011 and April 2015 when CAEP recordings were included in the infant audiology pathway. The infants with PCHI were identified from the Easy Screening Programme database (Stevens et al., 2013; British Society of Audiology, 2007). The severity of hearing loss was categorised using the British Society Audiology (BSA) descriptors (British Society of Audiology, 2011) as mild (20 to 39 dB HL), moderate (40 to 69 dB HL), severe (70 to 94 dB HL) and profound (>95 dB HL), using the average of hearing thresholds across 500, 1000, 2000, 4000 Hz. This four-frequency average hearing loss (4FAHL) was derived from detection thresholds assessed with Visual Reinforcement Audiometry (VRA) using warble tones through inserts to obtain ear specific thresholds at 6-8 months.

### 2.3.3 Enrolled participants

**Cohort A** consisted of 34 children with PCHI who were identified from the cohort of 31,373 babies born during the period from January 2008 to August 2011. There were 21 female and 13 male participants. In this cohort 24 children had English as a second language and 10 had English as their first language. The UK

follows the NHSP guidelines to diagnose children with hearing loss. These guidelines were the same for children prior to and post 2011.

**Cohort B** consisted of 44 children with PCHI identified from a cohort of 32,941 babies born between September 2011 and April 2015. There were 21 female and 23 male participants. Of the 44 children, 33 children had English as a second language and 11 had English as their first language.

The overall prevalence of PCHI in this group was 1.21/1000 (95% CI 0.96-1.50). The prevalence by degree of hearing loss and by cohort is provided in Table 1. There were no significant differences in the prevalence of PCHI by degree of impairment between the two cohorts ( $\chi^2$  analyses summarised in table 2.1). At the time of enrolment into the study the median age of Cohort A was 1.3 months (IQR 0.9-2.6) and for Cohort B was 0.9 months (IQR 0.7-1.5).

**Table 2. 1** The prevalence of PCHI by degree of PCHI in the two cohorts.

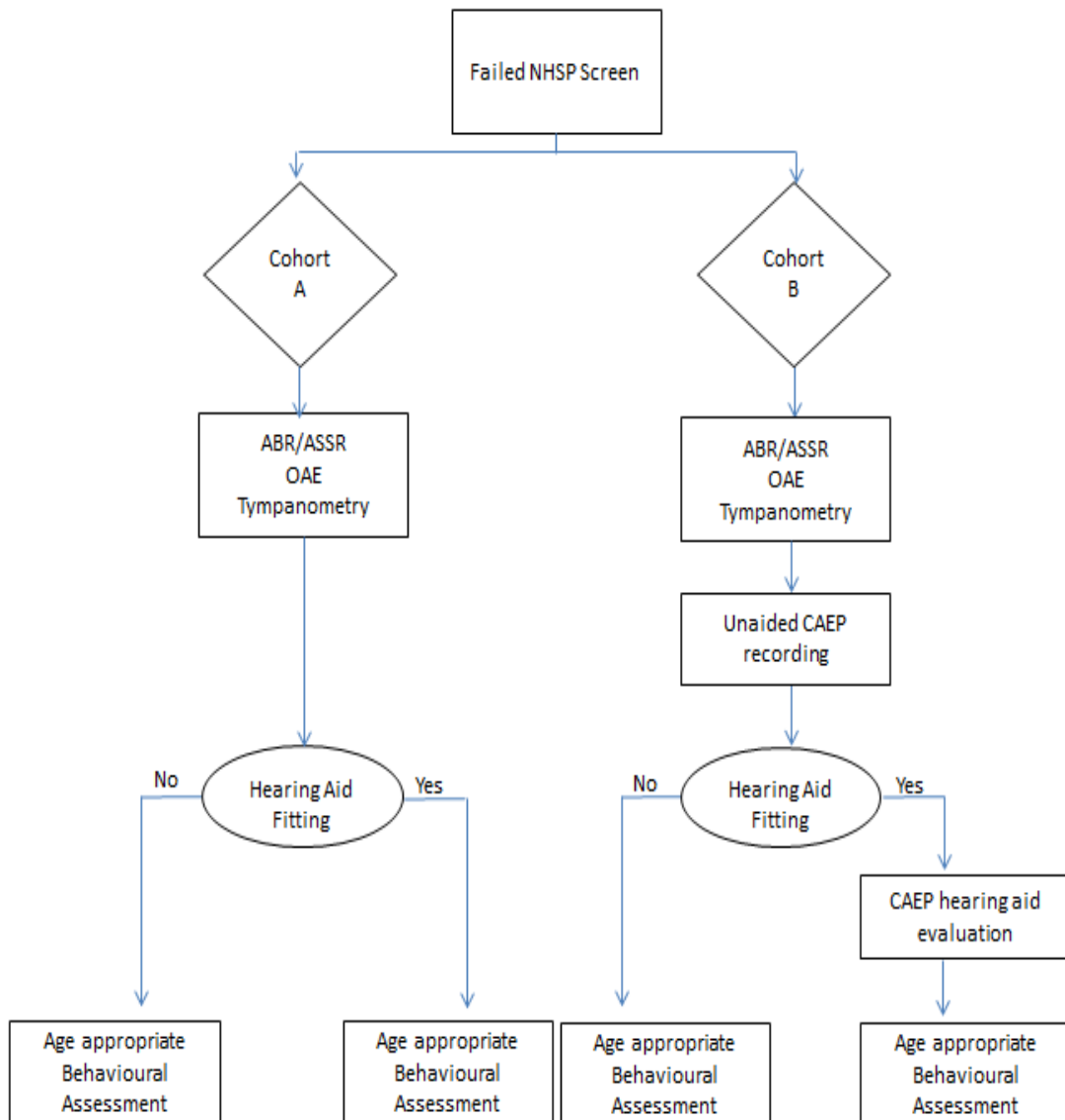
PCHI = Permanent Childhood Hearing Impairment

	Cohort A N. of births =31373			Cohort B N. of births =32941			$\chi^2$ ; df ; p
	N=	Prevalence /1000	95% Confidence Interval	N=	Prevalence /1000	95% Confidence Interval	
<b>Mild PCHI</b>	6	0.19	0.05 – 0.35	6	0.18	0.04 – 0.33	0.130; 1; 0.711
<b>Moderate PCHI</b>	17	0.54	0.28 – 0.80	21	0.64	0.36 – 0.91	0.249; 1; 0.618
<b>Sev/Prof PCHI</b>	11	0.35	0.14 – 0.56	17	0.52	0.27 – 0.76	1.011; 1; 0.315
<b>Total PCHI</b>	34	1.08	0.75 – 1.49	44	1.34	0.94 - 1.73	0.635; 1; 0.426

### **2.3.4 Procedures**

With both cohorts the only change that was introduced in the infant audiology pathway was the use of CAEP. Staff and clinical facilities remained the same and the follow up procedures were conducted according to the NHSP guidelines.

All the infants who were referred from the neonatal hearing screen followed a post-screen diagnostic test protocol which included ABR/ASSR, high frequency tympanometry and otoacoustic emissions (OAE). This test battery was established and delivered by experienced audiologists (Baldwin & Watkin, 2014). Identification of PCHI prompted immediate referral for both peer to peer and educational support for families. The support was undertaken both within the home and in a centre run by the Early Support Team for families of deaf and hearing impaired children. This support was established in 2008 and remained unchanged throughout the period. From 2011 an unaided CAEP recording was offered within 6-8 weeks of the PCHI confirmation and those with a more severe loss were seen by 4 weeks, followed by hearing aid fitting. Aided recording of CAEP was offered within 4-8 weeks of the hearing aid fitting to determine the effectiveness of the amplification. Throughout the period of the study all the children identified with a PCHI were regularly offered follow up appointments with continued assessment using behaviour observation audiometry (BOA), visual reinforcement audiometry (VRA) and eventually conditioned play audiometry. Multidisciplinary input was given by the teacher of the deaf (TOD) and the peer to peer support worker. Figure 2.1 shows the flow chart summarising the patient pathways for the two cohorts.



**Figure 2.1** Shows the patient pathway for Cohort A and Cohort B.

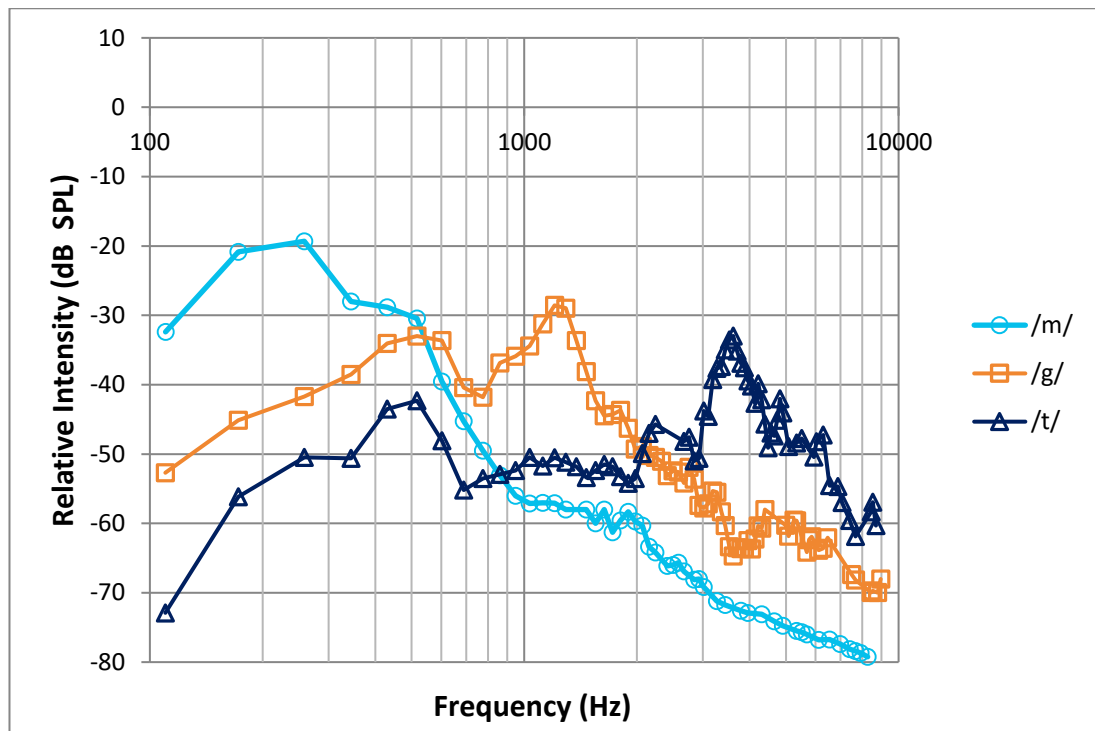
NHSP = Newborn Hearing Screening Programme, ABR = Auditory Brain Stem Response, ASSR = Auditory Steady State Response, OAE = Oto-Acoustic Emissions, PCHI = Permanent Childhood Hearing Impairment

### 2.3.5 Cortical Auditory Evoked Potential (CAEP)

All cortical measurements and calibrations were made in a sound-treated and electrically screened test room using the HEARLab system (Frye Electronics, 2013). The stimuli were the speech tokens /m/ (duration of 30ms), /g/ (duration of

20 ms), /t/ (duration of 30 ms), which were presented at nominal intensity levels of 55, 65, 75 dB SPL from a sound field speaker. These speech sounds have been described and justified in detail in research publications (Golding et al., 2009; Carter et al., 2010; Chang et al., 2012). The speech tokens were presented with an inter stimulus interval of 1125 ms, as described in Munro et al. (2011) using the HEARLab system.

The speech tokens were analysed to determine the spectrum and energy peaks of each sound (see figure 2.2). Stimuli were recorded using a B&K reference microphone 4192 and a B&K nexus amplifier. The speech tokens were analysed using the Audacity 2.0.6. software package.



**Figure 2.2** Spectra of the three HEARLab stimuli.

Automated calibration of the HEARLab system was performed at 75 dB SPL in the sound field before each testing session (in accordance with manufacturer's recommendations, Frye Electronics, 2013). Electroencephalography (EEG) band pass filter settings and artefact rejection levels were set to 0.2 to 30 Hz and  $\pm 150$   $\mu$ V, respectively. The child's vertex, forehead and right mastoid were prepared using Nuprep™ gel to ensure good contact between the skin and electrode.



Snap-on disposable electrodes were positioned at vertex as positive, right mastoid as negative, to keep consistency and reduce any experimental bias, with the forehead as ground with electrode impedance below 5 kOhms. A headband and surgical tape were used to stop the electrodes from slipping. Children were seated on their carer's lap, 1 meter from the loudspeaker. Responses were collected whilst the infants were alert, awake and quiet throughout testing and whilst watching a subtitled DVD with the sound disabled or using visual toys to engage the child. The child's EEG was recorded and the residual electrical noise was minimised to optimise the signal to noise ratio. The residual noise was displayed by the HEARLab software using a traffic light indicator method. The colour red appeared when residual noise voltage in the average waveform was  $>3.6\mu\text{V}$ , and green when the residual noise was  $<3.2\mu\text{V}$  and orange when in between. Testing was paused if the residual noise was  $>3.6\mu\text{V}$  and testing recommenced when the child had settled. Speech token stimuli were initially delivered at 65 dB SPL, and if no response was recorded the presentation level was increased to 75 dB SPL. If a CAEP response was evident at 65 dB SPL the presentation level was decreased to 55 dB SPL. This procedure was performed for each of the 3 speech stimuli for all the infants tested at both the initial unaided appointment and the subsequent CAEP hearing aid fitting evaluation. For each speech stimulus tested a total of 150 artefact free epochs were recorded from each child with an epoch of -200 to 600 msec. (Munro et al., 2011). The decision-making criterion for determining the presence of the cortical response was made objectively by the HEARLab using the Hotelling's T-squared ( $T^2$ ) statistical method (Flury & Riedwyl, 1988; Chang et al., 2012). The Hotelling's  $T^2$  statistical analysis divides each accepted response into nine 50 ms time bins starting from 101ms. All the samples within each bin are averaged to produce a single value and the Hotelling's  $T^2$  calculation determines if the response is significantly different from noise i.e. that an evoked response is present (significance level set at  $\alpha=0.05$  for rejecting the null hypothesis that the amplitude of the response and noise are not different).

### **2.3.6 The clinical implementation of the CAEP tests**

The initial unaided CAEP measurement appointment lasted between 90-120 minutes. The CAEPs were explained as for all tests and measurements, and the

parents observed the CAEP responses to the speech sounds. Following unaided CAEP recordings, early aiding was not recommended for children with a PCHI who had CAEPs responses at 55 dB SPL for speech tokens /g/ and /t/. Early hearing aid fitting was recommended if there was an absent response for /g/ or /t/ at 55 dB SPL if this was consistent with the diagnostic ABR/ASSR assessment. Although responses to /m/ were recorded and considered alongside the /g/ and /t/ speech tokens, the absence of an isolated response confined to the /m/ speech token did not prompt a recommendation for early hearing aid fitting. Van Dun et al. (2012) reported a lower detection sensitivity for the /m/ speech token with a sensitivity of 63% at a 20 dB SL and the early aiding in infancy of isolated low-frequency PCHI was not considered clinically necessary. All the children, for whom early hearing aid fitting was not recommended, were subsequently followed up with further assessment of their PCHI by behavioural testing. This prompted the audiologists to counsel parents when speech needed to be amplified and a hearing aid fitting appointment was made. If parents agreed to this a hearing aid was fitted. A second CAEP assessment was undertaken 4 to 8 weeks after the hearing aid fitting to assess the benefit of the amplification for speech sound detection, to the parents. When the CAEP was absent at 75 dB SPL in children optimally fitted with hearing aids, and if there were no contraindications, referral for cochlear implant assessment was recommended.

### **2.3.7 Hearing Aid Fitting**

Throughout the study period hearing aids were fitted following recommendations from the NHSP guidelines (British Society of Audiology, 2007; Feirn et al., 2014). The department followed the required NHSP protocol which was to use ABR to obtain two frequencies and added the additional procedure of using ASSR for the other two frequencies. ABR is a lengthy procedure compared to ASSR and there is a time limitation in testing with infants. The ABR thresholds for 1000 Hz & 4000 Hz and ASSR thresholds for 500 Hz & 2000 Hz were used to predict hearing levels for each child. This combination gave 4 frequency thresholds for more accurate hearing aid fitting. Correction factors for both ASSR and ABR were applied according to the NHSP guidelines (Stevens et al., 2013). The estimated behavioural threshold values were entered into the Audioscan Verifit real ear measurement (REM) system (Audioscan Verifit, 2014). Target gain

values were generated using the Desired Sensation Level (DSLv5) hearing aid prescription method (Seewald et al., 2005; Scollie et al., 2010). Hearing aid outputs were verified using Real Ear Aided Response (REAR) measures, and individually corrected using Real-Ear-To-Coupler Difference (RECD) or normative values for RECD for a child of the same age. Hearing aid outputs for 55, 65 and 75 dB SPL speech input were adjusted to match prescription targets (Bagatto et al., 2002; British Society of Audiology, 2007). The amplification was matched to targets within +/- 5 dB at frequencies of 500, 1000 and 2000 Hz, and of +/- 8 dB at 3000 and 4000 Hz, in accordance with national guidelines.

Regularity of hearing aid use was recorded at post-fitting assessments using the data logging facility in the hearing aids. For Cohort A, data logging was measured retrospectively from the fitting sessions at their first VRA appointment. For Cohort B the data logging was recorded at the appointment after the CAEP hearing aid fitting evaluation but before the VRA assessment. The data logging represented the duration of aid use that was achieved following the CAEP evaluations and before behavioural assessments had reinforced the need for amplification.

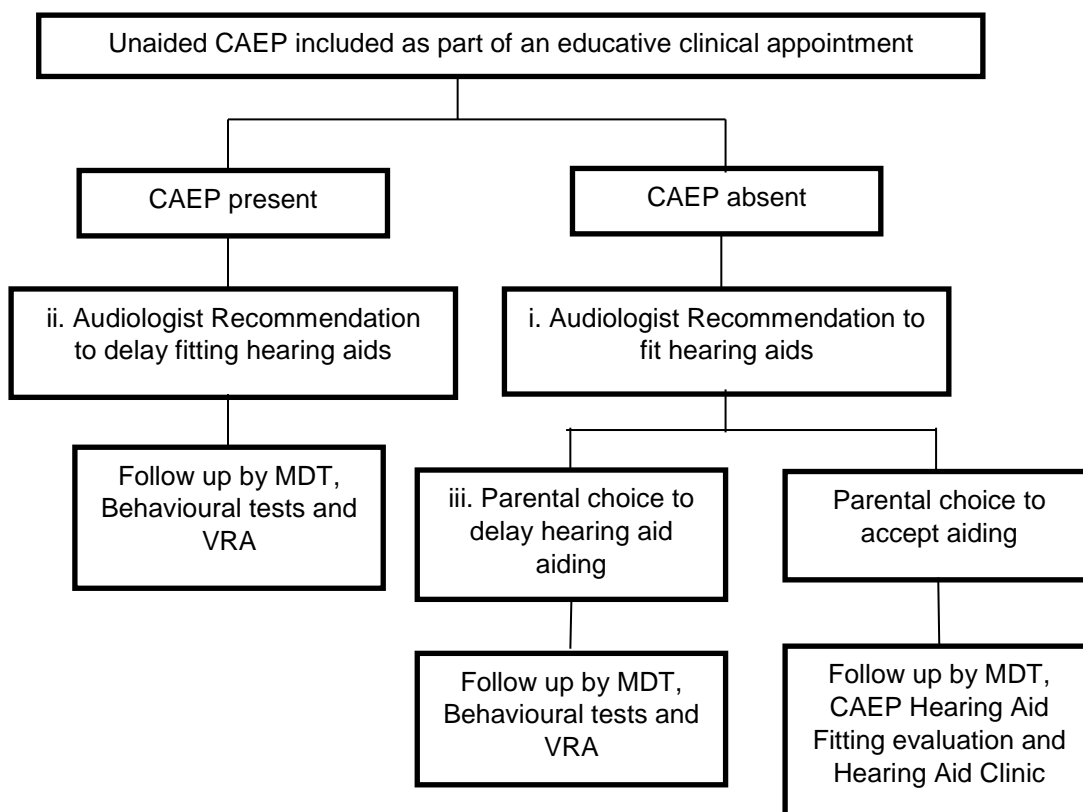
### **2.3.8 Data Analysis**

Demographic data were obtained from individual patient records and from the Easy Screening Programme to ensure that all eligible children diagnosed were included. Age at confirmation of PCHI, the age at hearing aid fitting and age at referral for cochlear implant assessment were recorded. Following the unaided CAEP assessment, decisions about the rehabilitative process were made through discussions between the audiologists and the parents. The children were categorised into one of three decisions groups (Figure 2.3).

Children who had an early hearing aid fitting (children fitted with hearing aids without any delay and at the appropriate age) undertaken as a result of the *Audiologist's Recommendation* made in the initial hearing evaluation appointments. For cohort A the assessment included ABR/ASSR thresholds and for cohort B it additionally included the unaided CAEP testing.

Children for whom hearing aid fitting was delayed as a result of the *Audiologist's Recommendation* following the initial hearing evaluation appointments.

Children for whom hearing aid fitting was delayed as a result of *Parental Choice* despite the audiologist's recommendation at the initial hearing evaluation appointments.



**Figure 2.3** Shows the management pathway following the unaided CAEP recording.

CAEP = Cortical Auditory Evoked Potential, MDT = Multidisciplinary Team, VRA = Visual Reinforcement Audiometry.

Distribution of data for age at hearing device fitting violated the assumptions of normality (Shapiro-Wilk;  $W=0.701$ ,  $p<0.01$ ) so the Mann-Whitney U test was used to make comparisons between the two cohorts.

## **2.4 Results**

### **2.4.1 The age of confirmation of PCHI**

The 34 children in Cohort A had PCHI confirmed at a median age of 1.3 months (IQR 0.9-2.6). The 44 children in Cohort B had PCHI confirmed at a median age of 0.9 months (IQR 0.7-1.5). The median ages of confirmation of PCHI were not significantly different in the two cohorts (Mann Whitney U = 590.50; p = 0.11).

### **2.4.2 The Unaided CAEP Recording**

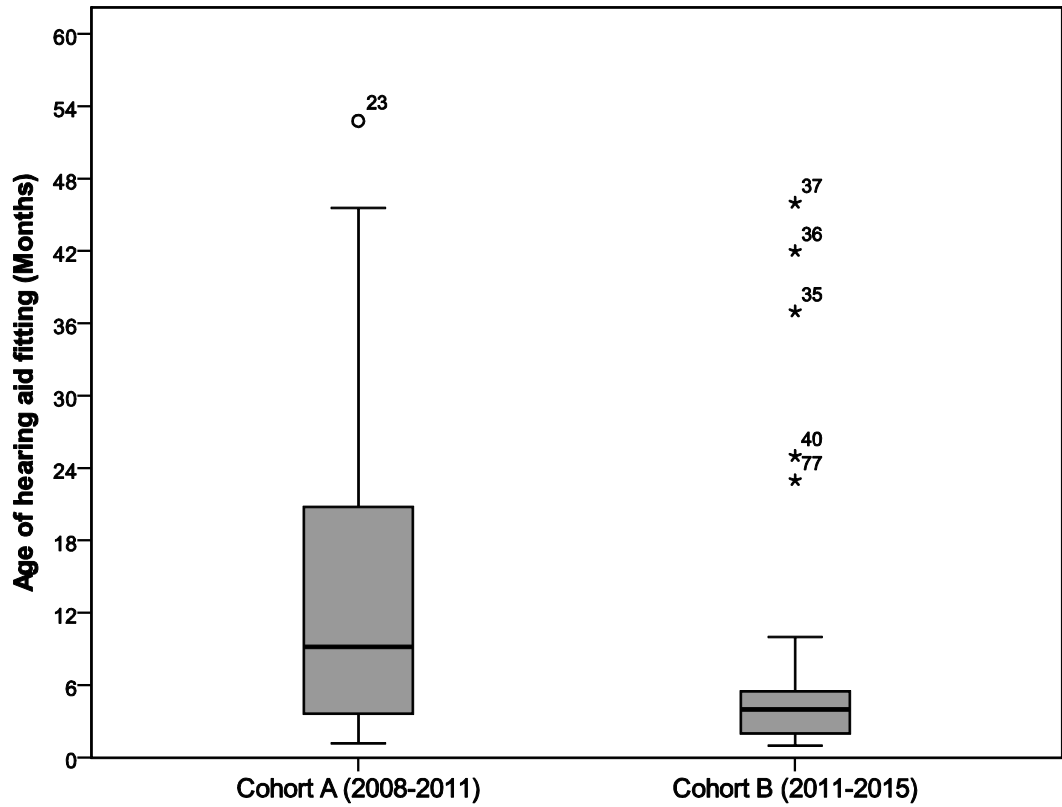
All 44 children in Cohort B had recording of unaided CAEP responses. Eight children had a second appointment because of illness or because they weren't sufficiently settled for testing. They were seen for unaided CAEP recording at a median age of 3.5 months (IQR 2.5-4.6) with a median delay of 8.8 weeks (IQR 5.1-14.9) after diagnostic confirmation of PCHI.

### **2.4.3 The age of hearing aid fitting**

Of the 34 children enrolled from Cohort A, 31 (91%) were fitted with hearing aids by the age of school entry. In Cohort B, 43 of the 44 children (97%) had been fitted with hearing aids by the time of this analysis. The single child who had not been fitted with hearing aids from Cohort B was aged 2.4 years at the time of the analysis. Figure 2.4 shows the median age of hearing aid fitting and the interquartile range (IQR) for children in the two cohorts. By 52 weeks 56% of children in Cohort A had been fitted with hearing aids compared to 90% from Cohort B. The outlier in Cohort A, (number 23) was a child with a moderate PCHI whose parents chose to delay the use of hearing aids until the age of 53 months. There were five outliers in Cohort B. Outlier 40 was a child with an asymmetrical PCHI with a 70 dB HL high frequency loss at 4 kHz in his better hearing ear and 80 dB HL 4FAHL in his worse hearing ear. He was fitted at 25 months. Outlier 77 had a severe PCHI and was fitted at 23 months. Both children had been identified by the CAEP recording as requiring hearing aids but their parents chose to put off the fitting until a delay in their child's speech and language prompted them to accept the audiologist's recommendation for hearing aid fitting.

Outliers numbered 35, 36 and 37 were children who had two or more CAEP responses at 55 dB SPL. The initial audiologist recommendation at the unaided CAEP recording for each of these children was to continue monitoring their hearing but not to undertake hearing aid fitting in infancy and they were eventually fitted at 39 months, 42 months and 46 months respectively. All 3 children showed delay in their speech and language which was reported by the TOD. The 39 month old child was showing a delay of 13 months and the other two children were showing a delay of 11 months. At this point hearing aid fitting was advised hearing aid fitting.

The median ages of hearing aid fitting by degree of PCHI and by cohort are presented in Table 2.2 Comparison of the cohorts confirmed that there had been no significant change in the age of hearing aid fitting for those with a severe or profound extents of hearing loss. However, the median age of hearing aid fitting for those confirmed with a mild-to-moderate hearing loss was significantly reduced from 19 months in the early cohort A to 5 months in the later cohort B (Mann Whitney U = 102.0;  $p < 0.01$ ).



**Figure 2.4** Box-and-whisker plots of age of hearing aid fitting for cohort A (N=31) and cohort B (N=43). Values are expressed as the median (horizontal line in each box), with the lower strand upper edges of the boxes showing the interquartile range (IQR 25th to 75th percentile) and range (T bar), outliers are shown by a circle and asterisk (\*).

**Table 2.2** The median age of hearing aid fitting and IQR in the two cohorts by degree of hearing loss. The grey box represents results which are significant  $p < 0.05$

	Cohort A			Cohort B			Mann Whitney U; p
	N=	Median age in months	IQR	N=	Median age in months	IQR	
<b>Mild-to-moderate PCHI</b>	20	19.00	14.58- 27.99	26	5.00	4.27- 14.58	U= 102.0; p<0.01
<b>Severe/Profound PCHI</b>	11	2.80	1.97- 4.82	17	3.00	1.70- 6.89	U= 99.0 ; p>0.10
<b>All with PCHI</b>	31	9.20	3.69- 20.76	43	3.90	2.20- 6.00	U=410.5; p<0.01

#### 2.4.4 Delays in the Fitting of Hearing Aids

The children were assigned a decision group category based on timing of hearing aid fitting following the unaided CAEP recording (Table 2.3). The groups were assessed by cohort and by degree of hearing loss. The median age of hearing aid fitting and IQR are presented. Delays typically occurred for infants with mild or moderate PCHI. In cohort A, this occurred in 16 out of 20 cases (80%) and in cohort B to 8 out of 26 (31%). The decision group category and the eventual age of fitting, are detailed in table 3.

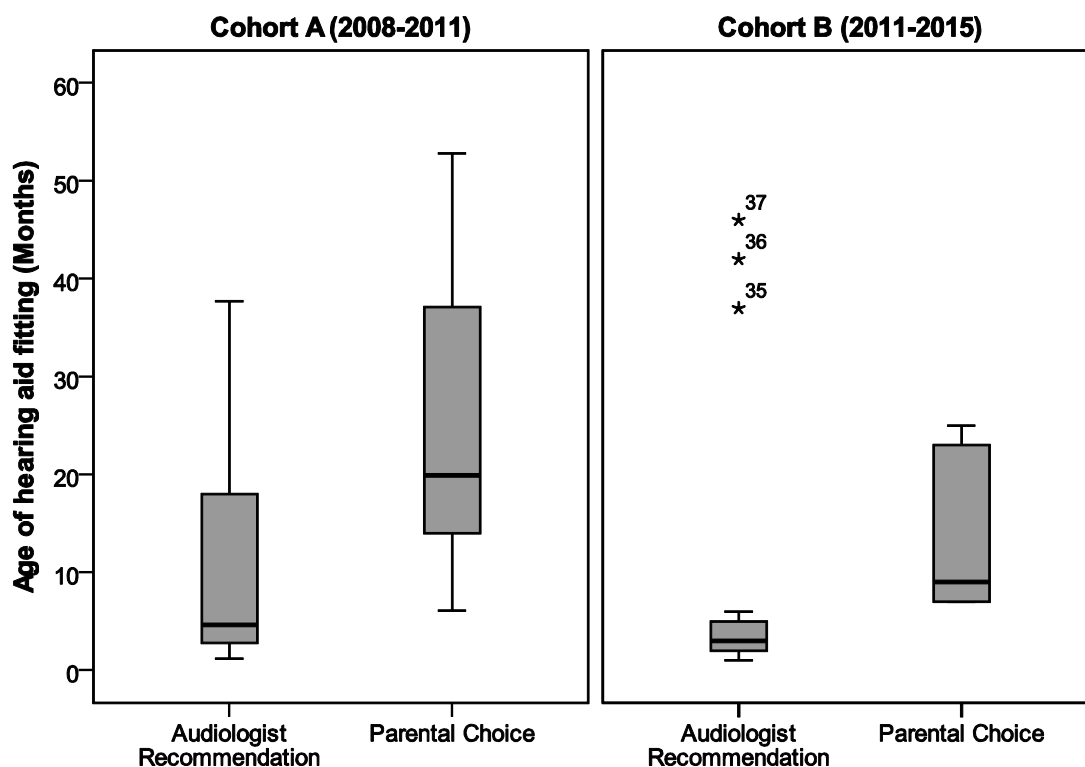


**Table 2.3** PCHIs in Cohort A and Cohort B, grouped by the severity of the hearing impairment and decision group categorisation.

PCHI = Permanent Childhood Hearing Impairment.

Cohort	Degree of PCHI	N. aided	Early Hearing aid fitting following Audiologist's Recommendation (i)		Delayed Hearing aid fitting following Audiologist's Recommendation (ii)		Delayed Hearing aid fitting due to Parental Choice (iii)	
			N (%)	Median age (IQR) in months	N (%)	Median age (IQR) in months	N (%)	Median age (IQR) in months
Cohort A.	Mild/ Mod	20	4 (20)	5.19 (2.95-8.44)	8 (40)	20.80 (14.10-29.97)	8 (40)	26.90 (14.75-42.14)
	Sev /Prof	11	10 (91)	2.80 (2.16-3.47)	0 (0)	-	1 (9)	9.20
Cohort B.	Mild/ Mod	26	18 (69)	3.40 (2.16-4.32)	3 (12)	41.50 (39.09-43.56)	5 (19)	8.40 (7.49-10.32)
	Sev /Prof	17	16 (94)	3.00 (2.30-3.94)	0 (0)	-	1 (6)	23.00

Those infants whose hearing aid fitting followed the audiologists' recommendation were grouped together. They included those where the fitting was undertaken early in infancy and those where fitting was delayed because of the audiologists' recommendation. In all, 22 of the 31 aided children (71%) in Cohort A were fitted as recommended by the audiologists. For 9 children hearing aids were fitted at an age determined by parental choice. In Cohort B, 37 of the 43 aided children (86%) were fitted in accordance with the audiologists' recommendation with 6 children having hearing aids fitted at an age delayed by parental choice. The median and IQR for the age of hearing aid fitting as grouped by the reason for the delay and by cohort, are presented in Figure 2.5. In Cohort A the median age of hearing aid fitting by the audiologist's recommendation was 4.6 months (IQR 2.9-16.6 months). The median age of the children fitted by parental choice was 19.8 months (IQR 14.0-37.1 months). In Cohort B the median age of fitting with hearing aids by audiologist recommendation was 3.2 months (IQR 2.0-4.6 months) and age at fitting by parental choice was 9.4 months (IQR 7.7-22.1 months).



**Figure 2.5** Box-and-whisker plots of those children with PCHI in cohort A (left panel) and cohort B (right panel), showing the age at which hearing aid fitting occurred based on the audiologists' recommendation (Cohort A N=22; Cohort B N=37) or when intervention took place later because of parental choice (Cohort A N=9; Cohort B N=6).

#### 2.4.5 A comparison of unaided and aided CAEPs for hearing aid fitting evaluation and hearing aid use

All 34 infants with hearing aid fitting following the unaided CAEP test attended for a repeat CAEP recording to evaluate the fitting. The median age of aided CAEP was 5.0 months (IQR 3.9-6.2 months). This represented a median delay of 7.1 weeks from the unaided CAEP (IQR 3.0-8.9 weeks). Twelve infants (35%) demonstrated an improvement of  $\geq 10$  dB in CAEP thresholds for all 3 speech tokens when wearing hearing aids, with 11 (32%) showed an improvement for 2 speech tokens, and 5 (15%) for a single token. No responses were recorded at

75 dB HL to any of the 3 tokens in 6 of the infants (18%) wearing functioning hearing aids with a high gain. This indicated that hearing aids were not providing sufficient amplification for sound to be audible at 75 dB HL suggesting that hearing levels were insufficient for good speech development. The aids may have provided information about environmental sounds at higher levels, but they were just inadequate for effective amplification of speech so the infants were referred for cochlear implant.

For cohort A, hearing aid usage demonstrated median usage of 4 hours 6 minutes each day and 22% of infants wearing the hearing aids for 7 hours and longer. Cohort B data logging demonstrated a median usage each day of 4 hours 25 minutes with 25% of the infants wearing the hearing aids for 7 hours 9 minutes or longer. This shows an improvement in duration of use from Cohort A to Cohort B (Mann Whitney U = 214.0; p=0.021).

#### **2.4.6 Behavioural Audiological Assessments**

In order to evaluate the accuracy of the hearing level estimates for the infants in cohort B with mild to moderate PCHI, the hearing loss categorisation based on audiometric values from the behavioural visual reinforcement audiometry (VRA) at 6-8 months were compared to the categories estimated from their ABR and ASSR at 3-4 months. This was checked to ensure that the thresholds were not over estimated which could have resulted in inappropriate hearing aid gain levels being provided. The estimates from ABR/ASSR thresholds and the behavioural VRA examination for the 26 children with a mild or moderate PCHI in Cohort B are presented in Table 2.4. The findings showed that all children had been appropriately classified according to category of hearing loss i.e. a 100% accuracy for this population suggesting that the infants were appropriately aided. Within this group of infants there were three infants with the mildest hearing impairment, who had unaided CAEP responses at 55 dB SPL, for these cases the audiologist had estimated a mild loss using the ABR/ASSR protocol but made the decision not to fit these children with hearing aids in infancy but they did eventually receive hearing aids at a median age of 42 months. The single child from Cohort B who had not been fitted with hearing aids on the basis of audiologist recommendation by the time of the analysis had a 4FAHL of 35 dB HL

in one ear and 36 dB HL in the other. At the point of data collection this child remained unaided and his early support monitoring protocol scores at the age of 32 months for attending, listening and vocalisation were age appropriate.

**Table 2.4** Categorisation of hearing impairment for children with mild or moderate PCHI based on early estimates from ABR/ASSR thresholds and the behavioural VRA examination undertaken at 6-8 months.

PCHI = Permanent Childhood Hearing Impairment, VRA = Visual Reinforcement Audiometry, ABR= Auditory Brainstem Response, ASSR= Auditory Steady State Response

		VRA threshold: N = Children	
		Mild	Moderate
ABR/ASSR : N= Children	Mild	5	0
	Moderate	0	21

The 18 infants with a mild or moderate PCHI in the early hearing aid fitting group, had a median 4FAHL of 58 dB HL (SD 13.7 dB). The median 4FAHL for the 5 infants where the parents delayed the hearing aid fitting until the VRA assessment was 56 dB HL (SD 15.8 dB) and the median 4FAHL was 41 dB HL (SD 9.1 dB) for the 3 children in the audiologist delayed hearing aid fitting group where responses had been present to 2 or more of the speech tokens at 55 dB SPL. The Mann-Whitney U-test showed that the 4FAHL for the group where the audiologist delayed hearing aid fitting was significantly lower than the 4FAHL in early hearing aid fitting group ( $U = 7.00, p = 0.044$ ). All other comparisons of 4FAHL between groups were not significant.

#### **2.4.7 The age of Referral for Cochlear Implant Assessment in cases of profound hearing loss**

Table 2.5 shows the age of referral for cochlear implant for Cohorts A and B. Referral was recommended for all those children with a PCHI who had hearing thresholds at 90dB HL or greater, unaided at 2 and 4 kHz and had poor audibility

demonstrated by behavioural tests when optimised hearing aids were being used. Referral rate for the 2011-15 Cohort B had increased to 9 cases (23% of those with a PCHI) and although there had been no reduction in the age of confirmation of their PCHI, or of the age of their initial hearing aid fitting, the median age of referral for cochlear implant assessment had fallen significantly to 8.2 months (Mann-Whitney U = 1;  $p < 0.01$ ). In Cohort A, one cochlear implant referral had been delayed through parental choice (20% of those referred), and in Cohort B one of the nine referrals were delayed for the same reason (11% of those referred). Table 2.6 shows the 9 individual aided and unaided CAEP responses. Eight of the nine infants referred for cochlear implant assessment in Cohort B had absent unaided CAEP responses at 75 dB SPL to all three speech tokens. Five of these had no demonstrable aided benefit and 3 had limited aided benefit. Child number 9 had an asymmetrical PCHI with a profound loss in her worse hearing ear. Aided CAEP responses were recorded from her better hearing ear at 55dB SPL to the 2 speech tokens /t/ and /g/. Behavioural assessments were consistent with these findings but these free field responses were from the better hearing ear and she was eventually referred for cochlear implant at 18.6 months because of absent responses in her profoundly deaf ear. One infant with absent aided CAEP responses was a candidate for cochlear implant referral but referral was rejected by Deaf parents. The percentage of the cochlear implant referrals that had followed the Audiologist's recommendation remained very similar in the two cohorts. The reduction in the age of referral from 20.2 months to 8.2 months had therefore apparently been driven by earlier audiologist recommendation.

**Table 2.5** The median age of confirmation of hearing loss, fitting of hearing aids and referral for cochlear implant assessment in cohort A and cohort B. The grey box represents results which are significant  $p < 0.05$ .

PCHI = Permanent Childhood Hearing Impairment

	Cohort A		Cohort B		Mann Whitney U; p
	N=	Median age (IQR) (months)	N=	Median age (IQR) (months)	
<b>Age of confirmation of PCHI</b>	5	0.8 (0.19-1.77)	9	1.7 (0.68-3.11)	U=11.5; $p > 0.10$
<b>Age of hearing aid fitting</b>	5	3.7 (0.41-6.63)	9	3.1 (1.91-4.18)	U=18.0; $p > 0.5$
<b>Age of referral for cochlear implant</b>	5	20.2 (15.15-29.32)	9	8.2 (6.26-13.74)	U=1.0; $p < 0.01$

**Table 2.6** The aided and unaided CAEP responses in the children who were referred for Cochlear Implant. The grey boxes represent a response is present.

CAEP= Cortical Auditory Evoked Potential.

Children	CAEP /m/		CAEP /g/		CAEP /t/	
	Unaided	Aided	Unaided	Aided	Unaided	Aided
1	Absent at 75dB SPL	Absent at 75dB SPL	Absent at 75dB SPL	Present at 65dB SPL	Absent at 75dB SPL	Absent at 75dB SPL
2	Absent at 75dB SPL	Absent at 75dB SPL	Absent at 75dB SPL	Absent at 75dB SPL	Absent at 75dB SPL	Absent at 75dB SPL
3	Absent at 75dB SPL	Absent at 75dB SPL	Absent at 75dB SPL	Absent at 75dB SPL	Absent at 75dB SPL	Present at 75dB SPL
4	Absent at 75dB SPL	Absent at 75dB SPL	Absent at 75dB SPL	Absent at 75dB SPL	Absent at 75dB SPL	Absent at 75dB SPL
5	Absent at 75dB SPL	Absent at 75dB SPL	Absent at 75dB SPL	Absent at 75dB SPL	Absent at 75dB SPL	Absent at 75dB SPL
6	Absent at 75dB SPL	Absent at 75dB SPL	Absent at 75dB SPL	Absent at 75dB SPL	Absent at 75dB SPL	Absent at 75dB SPL
7	Absent at 75dB SPL	Absent at 75dB SPL	Absent at 75dB SPL	Absent at 75dB SPL	Absent at 75dB SPL	Absent at 75dB SPL
8	Absent at 75dB SPL	Absent at 75dB SPL	Absent at 75dB SPL	Absent at 75dB SPL	Absent at 75dB SPL	Present at 65dB SPL
9	Present at 65dB SPL	Absent at 75dB SPL	Present at 55dB SPL	Present at 55dB SPL	Absent at 75dB SPL	Present at 55dB SPL

## 2.5 Discussion

This study demonstrates a significant reduction in the age at hearing aid fitting in children with a PCHI following the inclusion of speech sound CAEP measurements within the infant audiology pathway. This analysis compared the age of hearing aid fitting in sequential cohorts, before and after the CAEP assessment had been introduced. The median age of hearing aid fitting for children with all degrees of PCHI was reduced from 9.2 months in Cohort A to 3.9 months in Cohort B with a marked reduction from 19.0 months to 5 months in those with a mild or moderate PCHI. In the earlier cohort (Cohort A), even though



the presence of a mild or moderate PCHI had been confirmed by the neonatal diagnostic tests, the audiologist recommended delaying the hearing aid fitting for 40% of the infants until behavioural tests were available. However, for Cohort B, the recording of CAEP in response to short speech sounds enabled the audiologist to evaluate hearing detection for low, mid and high frequency speech sounds. In the later cohort this resulted in a recommendation for hearing aid fitting in early infancy in 88% of children with mild or moderate hearing losses. The observation that CAEP responses to speech sounds were absent influenced the parents who were able to hear the stimuli themselves. The CAEP evaluation was used as an educative process and the percentage of parents of infants with a mild or moderate PCHI who chose to defer hearing aid fitting fell from 40% to 19%. Even when they elected to wait, the delay in hearing aid fitting was reduced with the median fitting age of 26.9 months for Cohort A, falling to 8.0 months for Cohort B. In 82% of cases, aided CAEPs demonstrated hearing benefit for detecting the speech tokens, this motivated the parents to support their child's consistent use of hearing aids. As well as reducing the age of fitting of the hearing aids, hearing aid use average increased, where 25% of the infants wearing the hearing aids for 7 hours or longer, in cohort B. This compares favourably with median data logging of 3.7 hours in children aged 7 to 35 months reported by Munoz et al. (2014). These findings are similar to the children in cohort A who showed hearing aid use on average of 4.6 hours each day. The absence of responses to short speech sounds of 75 dB SPL prompted further behavioural assessment and discussion about referral for cochlear implant assessment. The age of cochlear implant referrals reduced from a median age of 20.2 months to 8.2 months from Cohort A to Cohort B respectively.

The age at hearing aid fitting is recognised to be an important factor on the outcomes of children with hearing loss (Moeller et al., 2007). The early studies supporting the need for UNHS in the United States reported higher language scores when neonatal identification was accompanied by the early provision of hearing aids and enrolment into particular, well developed intervention programs (Yoshinago-Itano, 1999; Moeller, 2000; Calderon & Naidu, 1999). A subsequent longitudinal study of children with mild to profound hearing loss reported by Sininger et al. (2010) reported that age at hearing aid fitting had the largest effect on auditory based communication outcomes. Despite the importance of early

fitting and consistent hearing aid use, this does not necessarily follow early identification.

Difficulties have been encountered when providing optimal hearing aid intervention for some populations. The New York State Universal Neonatal Hearing Screen programme reported variation in the age at which children who were identified neonatally were fitted with hearing aids (Dalzell et al., 2000). The median age at diagnosis of PCHI was 3 months for the entire group of 85 infants but only 36 were fitted with hearing aids and this minority were fitted at a median age of 7.5 months. The challenges encountered were: delays in obtaining parental agreement for early intervention, non-compliant parents, milder hearing losses and audiologist uncertainty that amplification was required. The difficulties encountered in New York mirrored the findings from an earlier cohort in East London. Watkin et al. (1990) reported that because children with a mild-to-moderate hearing loss responded to broad band environmental sounds, parents often found it difficult to recognise the impact of a hearing impairment by observing behaviours in the home. Unsurprisingly, if the effect of hearing loss is not apparent, the motivation for parents to use hearing aids is lower. Sjoblad et al. (2001) found that 65% of parents questioned hearing aid benefit and 30% of them emphasized the need for better education on hearing aid use. The value of providing hearing aids is reliant upon parents understanding how hearing aids can benefit their child (Ching et al., 2013; Munoz et al., 2014). In the reported cohorts, the inclusion of CAEP recording in the infant audiology pathway offered a novel opportunity to inform parents. CAEP testing facilitated discussion about the hearing impairment and the benefit of hearing aids in a pragmatic and audio-visual, heuristic manner.

The educative value of CAEP was recognised by the introduction of HEARLab into the Australian Hearing infant audiology pathway in 2011 (Punch et al., 2016). There were some initial concerns about the routine introduction of CAEP recording because sensitivity studies had demonstrated that in around 25% of cases CAEP responses were not detectable even when the sounds were audible. This reinforced the need to interpret results across a test battery within the infant audiology pathway. The CAEP aided assessment within 8 weeks of the initial hearing aid fit was used to check or guide modifications of the fitting, or to confirm

that despite optimal aiding, hearing responses to speech presentations were absent. The absence of recordable responses helped parents and professionals to consider the need for cochlear implant evaluation including children with ANSD. Our study adds evidence that CAEP can be a useful assessment and counselling tool both to reinforce the need for aiding and when no responses are observed, to consider the need for progressing to cochlear implant assessment.

Despite the concerns that absent CAEP responses may not always signal inaudibility of sound, no infant for whom hearing aid fitting was recommended, was found at the later behavioural tests to have been incorrectly aided. It is important to note, that CAEP alone was not used and ABR/ASSR assessment were used to derive hearing thresholds. In cases where absence of responses to speech tokens at 55 dB SPL had prompted the audiologist to recommend early aiding, average hearing thresholds were 58 dB HL. In cases where the audiologist recommended delaying the fitting because unaided CAEP responses were present at 55 dB SPL the median hearing levels were 41 dB HL compared to a median of 55 dB HL in the group where aiding was recommended, suggesting that greater uncertainty was present for infants with lower levels of hearing impairment. CAEP responses at 55 dB SPL speech sound may have a 10 dB sensation level when the hearing threshold is 45 dB HL and therefore predictably those with an average hearing loss of 41 dB HL would have shown responses to speech tokens of 55 dB SPL, and those with a loss of 58 dB HL would not (Punch et al., 2016). Those with an average loss of 41 dB HL did eventually require hearing aid fitting and it is important that the presence of unaided CAEP responses in infancy does not offer false reassurance that hearing aid fitting will not be required during the pre-school period. However, for the majority of infants the introduction of unaided CAEP assessment had offered additional information both to the audiologist and the parents for audibility of short speech sounds and the benefit of using hearing aids. This presented an opportunity for counselling the parents which was considered pivotal in the marked reduction in the age when hearing aids were fitted to those children who were identified neonatally and who had anything less than a severe PCHI.

### **2.5.1 Limitations of the study**

In this study one of the cohorts was historical and this poses inherent problems. Changing attitudes of audiologists, the hearing support team and the parents over the period of the study may have been a contributory factor in reducing the age at hearing aid fitting. In the UK, Cross (2011) looked at trends in the age of referral for cochlear implant between 2001 and 2010 which had significantly decreased over this period. However, although a culture of earlier referral for cochlear implant evaluation had been nationally fostered, the CAEP test provided a clinical rationale to support both parents and audiologists in the need to make such referrals. Such changing attitudes had also influenced the age at hearing aid fitting following the establishment in England of the NHSP. Wood et al. (2015) reported this national trend in children with moderate and worse hearing impairment and this is likely to have contributed to the reduction in age at hearing aid fitting found in the current study. Nonetheless the staff and clinical facility had remained constant over the period reported and the single major difference in the infant audiology pathway between the two sequential cohorts had been the introduction of the CAEP recording. The CAEP test had provided a means of demonstrating the clinical need for hearing aid fitting to both parents and audiologists and it was considered a significant factor especially in those with milder impairments. Unfortunately, there is the potential for missing mild hearing losses which cannot be addressed through testing in the free field. In order to assess levels below 55 dB SPL, insert earphones or similar are necessary, however this would not allow comparison between aided and unaided measures. The use of sequential cohorts imposed some limitations on follow up. The early cohort consisted of children born between January 2008 and August 2011 and it was followed up until all the children had entered primary school. The later cohort consisted of children born between September 2011 and April 2015 and at the time of analysis one child remained unaided and had not reached primary school age so had the potential to still receive a hearing aid, however this one child has a minimal effect on the findings. The moderate sample size, single centre and narrow demographics represented in the sample, limit generalizability of the findings. However optimising hearing aid management is the foundation for improving auditory-based communication and use of CAEP recording can provide information about speech sound detection and the benefit

of hearing aid use encouraging timely parental engagement. The reason for this will be further explored with the families via questionnaires and focus groups (Mehta et al., 2019).

## **2.6 Conclusion**

This study has shown that CAEP recording has the potential to be an important tool for inclusion in the infant audiology pathway for infants with confirmed PCHI. In a comparison between two consecutive cohorts, where the difference was that the latter followed an infant audiology pathway containing CAEP recording, it was shown that the cohort receiving CAEP testing had a lower age of hearing aid fitting. Interestingly, the most dramatic difference was observed for infants with mild or moderate hearing losses. It is postulated that this occurred because the CAEP is an educative tool for parents to understand their child's hearing impairment and for audiologists to have an additional objective tool to increase confidence in their assessments. This study also indicated an earlier age at referral for cochlear implantation for those in the group receiving CAEP testing, however this is in line with the National trend.

The theories to explain the findings will be evaluated further and research conducted to evaluate language outcomes for children receiving hearing aids at a younger age.

## **CHAPTER 3: A qualitative review of parents' perspectives on the value of CAEP recording in influencing their acceptance of hearing devices for their child.**

### **3.1 Abstract**

The aim of this research was to obtain a parental perspective on how audiological tests, including recording cortical auditory-evoked potentials to speech sounds (CAEP), influenced their uptake of hearing devices for their infant. A focus group was established of parents of hearing-impaired children. A facilitator explored how audiology tests influenced their understanding and management of hearing loss in their child and their acceptance of hearing aids or cochlear implant referral. The views were transcribed and thematic analysis was used to determine the key topics of importance to parents. Eight sets of parents participated. Their children had been enrolled in an infant audiology pathway that included CAEP testing. The sample included 6 children who were aided, 1 child who was going through the assessments for cochlear implants and 1 child who was already implanted. Parents reported that it was important for them to understand the test results because this influenced acceptance of hearing aids or move forward to cochlear implant assessments. Seven sets of parents had not understood ABR results whilst 6 reported that CAEPs had helped them to understand their child's hearing and need for intervention. Compliance with early hearing aid use and referral for cochlear implant depends upon parents' understanding of their infant's hearing loss by including CAEPs in the infant audiology pathway.

### **3.2 Introduction**

Babies born in the UK are screened for hearing loss through the newborn hearing screening programme (Davis & Hind, 2003). Those requiring diagnostic assessment are referred for auditory brainstem response (ABR) to estimate the degree of hearing loss. If hearing aids are required, then the prescription gains are also derived from these results. At this age parental observation of the behaviour of infants to sound is often ambiguous and so the ability of families to understand the impact of the hearing loss largely depends upon information

provided by the audiologist and hearing support team. Optimal day-to-day use of hearing aids depends on the commitment and determination of parents to place the hearing aids on their child, and to provide a good listening environment and spoken language input. This can only be achieved if families understand the impact of hearing loss and the importance of using amplification which provides access to speech (Muñoz et al., 2015). In those with a more severe loss the age at cochlear implantation is an important factor in determining speech and language outcomes (Sharma A et al., 2002; Nicholas & Geers 2006). Ching et al. (2013) in their prospective study of three-year olds found that better outcomes were associated with a younger age at cochlear implant switch-on. Other studies support this and report that children implanted within the first year of life can achieve age appropriate spoken language (Holman et al., 2013; Dettman et al., 2016). The possibility of achieving better outcomes in children who are implanted in infancy has encouraged the promotion of very early implantation (Sharma et al., 2017). However, parents need to be able to make informed decisions which require a good understanding of the severity and implications of their child's hearing loss, and the options that are available to them.

The recognition of hearing loss and the subsequent use of hearing devices by the family impacts upon the deaf child's language development. Watkin et al. (2007) who explored language ability in 120 children with Permanent Childhood Hearing Impairment (PCHI) and reported that family participation correlated positively with language development. Holzinger et al. (2011) quantified the effect suggesting that 60% of the variance in speech and language outcomes was explained by family-related factors. Thus, we can predict that the use of hearing aids may be less effective when parents do not understand the implications of their child's hearing loss or the purpose of hearing aids thus compromising management. Muñoz et al. (2015) reported the challenges faced by parents for achieving consistent aid use. The challenges they found was inconsistent hearing aid use, expectations not addressed by audiologist, lack of hearing aid benefit and psychosocial challenges. The children enrolled in their study had been issued with hearing aids for some 15 months, but as toddlers one-third were still wearing them for less than 5 hours a day. Walker et al. (2015) in a longitudinal study of 290 children found that although the majority of children had been identified neonatally, the mean age of hearing aid fitting was 11 months and in those aged

less than 2 years, over half were wearing their hearing aids for less than 4 hours a day.

Punch et al. (2016) reported a retrospective review of 83 infants with PCHI, who were fitted with hearing aids based on CAEP results that were used to determine prescriptive targets according to the Australian national protocol. Their findings indicated that professionals believed CAEP testing had a positive impact on the effectiveness of hearing aid provision and that CAEP testing was well received by parents as a counselling tool to help reassure them that the hearing aid was providing sufficient amplification. This led to integration of CAEP testing into the Australian routine infant hearing aid fitting program.

Mehta et al. (2017) reviewed data for an inner city UK clinic following the introduction of CAEPs into the infant audiology pathway. They observed an earlier age for cochlear implant referral for severe-profound hearing loss, and also an earlier age of amplification for infants with mild-to-moderate hearing loss compared to a cohort of children assessed prior to the introduction of CAEPs in the infant audiology pathway. They hypothesised that the important change for those infants with a mild-to-moderate hearing impairment arose in part because the CAEP recordings enabled parents to be better informed about both the effect of the hearing loss and the impact of hearing aids.

Mehta et al. (2017; also chapter 2) and Punch et al. (2016) and used a system that measured CAEP responses to speech sounds presented from a loudspeaker placed in front of the child. Three speech sounds with a spectral emphasis in the low, mid and high frequency regions were used (Van Dun et al., 2012). The CAEP test can be conducted with and without hearing aids to demonstrate the impact of amplification on the child's detection of speech sounds. With this approach parents can hear sounds of different frequencies and levels and can understand how quiet or loud they are. The CAEP recording to speech stimuli enables parents to observe their child's recorded responses with and without hearing aids and this can be used as a counselling tool enabling parents to make informed decisions about their child's needs.



As stated earlier, we hypothesise that the reduction in age at intervention with hearing devices reported by Mehta et al. (2017) was in part due to the use of CAEP recordings to help audiologist aid mild-to-moderate hearing losses and for parents to better understand their child's hearing loss and to engage with the use of the hearing devices that were available to them. To further investigate this particular hypothesis a focus group of parents was held to explore how different elements of the infant audiology pathway influenced parent's understanding of their child's hearing loss, which specific aspects were most important and whether this empowered them to make hearing device choices.

### **3.2.1 The infant audiology pathway**

The study took place in the paediatric audiology department in an inner city UK clinic. All the infants referred from neonatal hearing screening were placed on a post-screen diagnostic test pathway which included ABR and unaided CAEP recording. The post-screen diagnostic tests were undertaken by experienced paediatric audiologists. The initial audiological assessments were followed by hearing aid fitting with the recording of aided CAEPs to determine the effectiveness of the amplification. All the children identified with a hearing loss were offered regular follow-up appointments with continued assessment which consisted of visual reinforcement audiometry (VRA) using the Ling 6 sounds and eventually conditioned play audiometry. Identification of PCHI prompted immediate referral for parental support and educational counselling and guidance. Peer-to-peer support was given by a parent support worker (PSW) with experience as a parent of a deaf child. Parental counselling and guidance was provided by a specialist teacher of the deaf. Both supportive services were undertaken within the home and in a centre run by the Early Support Team for families of deaf and hearing-impaired children.

## **3.3 Methods**

### **3.3.1 Ethical Considerations**

The study was registered as a service evaluation with the Hospital Research and Development department: registration number 1275. Written consent was taken

from the parents to participate in the focus group and for the facilitator to make audio-recordings of the discussions and to record any comments that were made by the group.

### **3.3.2 Selection of focus group participants**

Parents were identified through a purposive selection process to include families whose children had been identified with a bilateral PCHI by the Newborn Hearing Screen Programme and who had been managed by an infant audiology pathway that included the recording of aided and unaided CAEP. Although not necessarily the first language of the home, 15 families who could speak English were invited by letter to participate in the focus group. The invitation was subsequently followed up by contacting the families with a text message. The invitation requested that when possible both parents or a parent and another primary carer attend the focus group. Very often decisions are made by the primary carers away from the clinical appointment and it was considered desirable to include the views of those decision makers who may not have been able to be present at pivotal audiological assessments. To facilitate the attendance of working members of the family the focus group was conducted in the early evening. Ten families responded to the invitation but two were not able to attend on the date of the focus group. Single parent and same sex parents were invited but did not attend. The group therefore consisted of the mother and father of eight children, 16 parents in total. The demographic details of the group participants are detailed in table 3.1.

**Table 3.1** Characteristics of the 16 parents who participated in the focus group.

Gender (n.)	Male: 8 Female: 8
Age in years	Mean: 36 Range: 24-45
Born in UK (n.)	8 parents (4 families)
English not spoken as the main language of the home (n.)	8 parents (4 families)
Level of Education received (n.)	
i. Higher Education	5
ii. High school examinations or apprenticeship/ equivalent	2
iii. Other foreign, high school or apprenticeship qualifications	6
iv. No qualifications	3

Both the number of participants not born in the UK and the number who did not speak English as their main language was substantially higher than the overall UK population, but typical for London, where the clinic was based. The proportion who had passed through a course of higher education was the same as the national average. Half of the parents participating in the focus group had high or secondary school qualifications gained in the UK, or the equivalent achieved in their country of birth. This was only marginally lower than the national average, but characteristic of the local resident population (UK profiles, 2017).

The eight children all had a bilateral PCHI. Their age when the focus group was held ranged from 6 to 36 months with a mean of 16.3 months. The severity of their PCHI was categorised using the British Society Audiology (BSA) descriptors as mild (20 to 39 dB HL), moderate (40 to 69 dB HL), severe (70 to 94 dB HL)

and profound (>95 dB HL), using the average of the better hearing ear at octave intervals from 500Hz to 4000 Hz. The degree of hearing loss and other characteristics of the 8 children are detailed in table 3.2. The median four-frequency hearing loss of the eight children was 61 dB SPL in the better hearing ear. The children were categorised using their hearing loss from the initial hearing aid fitting appointment. Seven children had a bilateral symmetrical PCHI with two having a sloping high frequency loss. The remaining child had a profound loss in one ear with a moderate loss across the frequency range in the other.

**Table 3.2** Characteristics of the 8 children whose parents participated in the focus group.

Gender (n.)	Female: 4 Male: 4
Age (n.)	6 months – 12 months: 4 12 months – 36 months: 4
Type of hearing devices (n.)	Phonak hearing aids: 3 Oticon hearing aids: 3 Referred for cochlear implants and using Phonak hearing aids at the time of focus group: 1 Cochlear implant: 1
Age of CI referral in months	Child 1: 4 months Child 2: 5 months
Degree of hearing loss in better hearing ear (n.)	Mild: 2 Moderate: 2 Severe: 2 Profound: 2

### **3.3.3 Focus Group**

The focus group was held during a single evening session. It took place in a quiet room in the centre where the children had been assessed. The group's facilitator was the first author of this paper and the paediatric audiologist who had assessed the children. The families sat around a small table allowing the participants to see each other. The facilitator explained the procedures to the group. It was emphasised that individual families had their own understanding, knowledge, experiences and viewpoints, and that all differences would be respected. Approximately 30 minutes were spent with the families introducing themselves and sharing their experiences about their child's diagnosis, level of hearing and use of hearing devices (hearing aids or cochlear implants).

The facilitator then explained that the aim of the group was to explore the parents' views of how the different clinical audiology tests had affected their understanding of the child's hearing and how it had guided their management of the hearing loss. The discussions aimed at exploring how the tests had influenced their use of the hearing aids and, when appropriate, how they had affected their decision to engage with further cochlear implant assessments. Leading questions about individual tests were avoided with the participants being asked to mention anything at all that they had found helpful. The facilitator used the information offered by the parents to further guide the discussions and to explore their perspective on the value of the different tests. References by the parents to specific audiological tests, assessments, events and support that they had received were recorded and if not clear the facilitator encouraged participants to give more details. All 6 of the following general questions were asked with the discussions being audio-recorded. Parents were encouraged to speak freely about topics and expand on information. Once the introduction has completed the remaining session lasted for approximately 40 minutes.

How confident are you about describing your child's hearing loss and could you describe your child's hearing loss for the group?

Do you know how your child's hearing loss will affect his/her hearing of speech and what effect that will have on them?

Can you remember any of the appointments/events that helped you to understand your child's hearing needs?

Which part of the process/appointments did you find most helpful to understand your child's hearing loss?

When did you feel your child needed hearing aids? What helped you realise that?

Do you know what your child can hear with and without their hearing aids?

The questioning prompted a wide range of discussions with the facilitator balancing the discussions and encouraging less confident contributors so that all opinions were heard. Parents had the opportunity not to answer the questions and at times were asked to discuss their responses.

### **3.3.4 Data Analysis**

The audio recordings were transcribed by the first author verbatim, for use in a thematic data analysis. Thematic analysis is a method used for identifying, analysing and reporting patterns (themes) within data (Braun and Clarke 2006). The qualitative research software NVivo 10 ([www.qsrinternational.com](http://www.qsrinternational.com); qualitative software) was used to assist with data handling, organisation and coding. The thematic synthesis was conducted in three stages:

1. Familiarisation with data: free line-by-line coding from the focus group,
2. The organisation of these 'free codes' into related areas to have general themes,
3. Defining and naming themes and report writing.

The parental discussions were wide-ranging and recurrent themes that were identified in the dialogue related to the parents' feelings about their child's hearing loss and their views of the support they had received. For the current study a "top down" (theoretical or deductive) thematic analysis was employed, and the coded data was organised into general themes that related to the discussions that were held about the clinical audiology tests with associated sub-themes of similar response by different parents focussing on how the tests had affected their understanding and management. Illustrative content was extracted from the transcribed text and is presented using quotations and includes the

child's gender and age at the time of the focus group. The facilitator's explanatory text is added to the transcribed text in brackets. Individual family members reinforced a unified view and therefore numeric analysis was by family rather than by parent.

### 3.4 Results

The analysis of the parental discussions topics are presented in order of where they fall in the infant audiology pathway.

#### 3.4.1 Auditory Brainstem Response

The parents discussed the ABR, but 7 of the 8 families felt quite negative about the test (table 3.3). Almost all had found it too long and although they had been shown the results by the audiologists and had them explained, they had failed to fully understand them. Two parents discussed the difficulties they had in "getting their heads around" lines shown to them on a page. Despite careful explanations by the audiologists the results were not meaningful to them. Another father asserted that his family's lack of understanding of the implications of the ABR test results had significantly hindered their engagement with further audiology appointments.

**Table 3.3** Parents' perspectives on ABR.

The number of families who expressed similar views are shown in brackets with the denominator being the total number of families.

Found the test too long. (6/8)
Did not fully understand the test. Very unhelpful. (7/8)
Found the whole experience very tiresome. (5/8)
Could not fully understand the results which were lines on a page. (2/8)

An example of one parent's comment:

*"..... it was our first audiology appointment, our child would not sleep, (we) had to come back to have the test repeated. We had no faith in the test until we came*

*back and had all the other various tests done with the puppets (here parents were referring to the VRA with speech sounds)” (Parents of 12 month old boy)*

*“The first hearing test (the ABR) was so difficult that we didn’t come back to any follow-ups.....because of this we didn’t discuss hearing aid fitting until the PSW came to our home” (Parents of 15 month old boy)*

### **3.4.2 Cortical Auditory Evoked Potential**

All 8 families made positive comments about the usefulness of the CAEP test in helping both the understanding and management of their child’s hearing loss (table 3.4). Seven of the families stated that the CAEP recording helped them to understand the potential benefit of hearing aid use. They commented that the CAEP test gave them increased understanding of the impact of the hearing loss and this encouraged them to agree to the trial of hearing aids. It also helped those families whose children demonstrated no aided CAEP benefit to consider the need for their child to be referred for cochlear implants. Some families discussed the initial difficulties they had encountered in recognising the presence of a hearing loss. At home, they had observed their baby responding to sounds and to their name being called. However, the CAEP test demonstrated their child’s inability to hear the quiet speech sounds, and because they were themselves able to hear the sounds played through a loud speaker they were able to understand their child’s level of hearing.



**Table 3.4** Parents' perspectives on the CAEP test.

The number of families who expressed similar views are shown in brackets with the denominator being the total number of families.

<b>CAEP affected their understanding</b>
Found the test easier to understand than the ABR (6/8)
Helped them to understand the degree of their child's hearing loss (6/8)
Helped them to understand the unaided hearing loss for speech (5/8)
Helped them to understand the aided benefit for hearing speech (5/8)
<b>CAEP influenced their management of the hearing loss</b>
Helped in the fine tuning of the hearing aids (3/8)
Helped them to engage with the decision to fit hearing aids (6/8)
Helped changed their attitude towards hearing aids use (7/8)
Helped them appreciate the need for future management (7/8)
Demonstrated to them the need for further assessments for cochlear implant (3/3)

Associated comments from the parents:

*"The speech test that our child underwent when she was about 4 months, we could see from the results that she required the aids, without this we would not have agreed to go ahead with the hearing aid fitting." (Parents of 6 month old girl)*

*"I also found the sticky pad test (identified as CAEP by the facilitator) useful in understanding what speech sounds my child can produce and being able to relate this to her hearing loss. As my child has a mild loss if we were not shown these test results I would not have believed the hearing loss, we would have not proceeded with the hearing aids." (Parents of 24 month old girl)*

Two of the 8 children had sloping high frequency hearing losses, and in both families the demonstration that their infants were unable to hear speech sounds with spectral emphasis in different frequency regions helped both their understanding and management. It also helped one parent appreciate the need to tune her child's hearing aid.

*“I was able to understand my child’s hearing loss when he had the test (the CAEP) without the aids. He could hear the other sounds but not the /t/. Now I could understand why he was turning to things at home and why we didn’t think he needed hearing aids.” (Parents of Male aged 12 months)*

*“I found it useful when hearing aids were tuned because he could not hear the m sound. I also found the way the results are set out on the screen understandable.” (Parent of male aged 15 months)*

Although all families offered positive comments about the CAEP tests, there were some additional unhelpful aspects of the test. The recordings achieved for the child with a moderate loss in one ear and a profound loss in the other, reflected the hearing in her better hearing ear, and this failed to help the parents understand the complexities of the asymmetrical PCHI. Two parents had additional negative observations. One remarked that the speech sounds used in the test were “very robotic”. Another family had found the CAEP recording process disappointing as no aided benefit had been shown. However, despite this reservation they agreed that the test had helped with their child’s subsequent cochlear implant referral. They said:

*“My child has profound hearing loss. I found the CAEP test to be disheartening as it showed up that my child could not hear any of the sounds even with the aids, it made us feel like ‘what was the purpose of using the aids.’” (Parents of 12 month old boy)*

### **3.4.3 Visual Reinforcement Audiometry**

A small number of the families participating in the group recalled and commented on the VRA test with speech sounds (Ling sounds) that had been conducted at around 7-8 months. Three had found that it clarified the hearing loss and the benefit of using the hearing aids. They also commented that it re-enforced the earlier results of the CAEP test with two families preferring the VRA test because they were able to observe their child’s behavioural responses to sound. Not all the comments were positive, and they are detailed in table 3.5.

**Table 3.5** Parents perspectives on the VRA test with speech sounds.

The number of families who expressed similar views are shown in brackets with the denominator being the total number of families.

Found test easy to understand and could see clear responses. (3/8)
It was reassuring because it gave same results as the CAEP test (3/8)
Was good that it wasn't computer based. The turns to sounds could be seen (2/8)
The observed responses to speech sounds helped encourage the use of aids (3/8)
The children found the puppets very scary (2/8)

Some of the comments were:

*"We both felt that we came to terms with her hearing loss after the puppet test which showed us what she could hear with and without the aids. We could hear the speech sounds from a loud speaker, which helped the process (the sounds parents were referring to were identified by the facilitator as the Ling 6 sounds). We could see when our child turned to speech sounds. We now know that the aids have been very helpful, and this has helped her speech. We use the sounds at home now to check the aids now"* (Parent of 20 month old girl)

*"My child hated the puppet test, every time they appeared she would freak out, need to think about using new puppets."* (Parents of 20 month old girl)

#### **3.4.4 Supporting the Audiology Tests**

A theme that was identified by all parents was that they needed to understand the results of the audiology tests to engage with the available management options. However, this understanding depended upon the additional support that was given *outside* the test session. Even though the CAEP test gave them greater understanding during the session, not all the parents had been present at this appointment and most of those who had been commented that they had found it too difficult to fully understand the results and all the implications in the clinic. The parents had therefore valued the explanations and input of the PSW in the home. The consensus of the group participants was that without the input

of the PSW the families would have “been lost” and they would have taken longer to decide on the aids and to make management choices. CAEP tests were used to make decisions about fitting hearing aids to two babies with a mild hearing loss. Both these parents revealed that they had only agreed to intervention after further discussions of the results in their home. The PSWs then acted as a bridge with the clinicians. The sub-themes relating to the importance of supporting the test results are detailed in table 3.6.

**Table 3.6** Parents perspectives on the need for support to understand the test results.

The number of families who expressed similar views are shown in brackets with the denominator being the total number of families.

Parents found it useful that the PSWs supported them with home visits (8/8)
The home visits more fully explained test results and empowered parents to make choices, especially when one of the parents hadn't been in the clinic when the test was done (8/8)
The home visits explained test results that were not always understood in the clinic because it was “too much” to take it all in at the appointments (6/8)
The home visit explained the CAEP results enabling parents to decide on aiding (4/8)
The home visits helped with the decision for referral for cochlear implant assessment (3/3)

A typical comment regarding the important role of the PSWs in understanding the test results was:

*“PSW helped me understand the test results such as the sticky pad one (identified as CAEP on further questioning by the facilitator) and was good in explaining to both of us how the test fits with my child’s hearing loss”. (Parents of 20 month old girl)*

### 3.5 Discussion

The focus group identified that both the audiological tests and the support available affected the families' understanding and in turn their management of the PCHI following confirmation by the NHSP. The test type and support influenced the adoption of hearing aids and when necessary the acceptance by parents that their child was a candidate for cochlear implants. The participants asserted that they needed to understand their baby's hearing impairment before hearing aids were accepted and before they were regularly used in the home. Although ABR is used in the diagnostic assessment of hearing thresholds and in the estimation of the prescription gain for fitting hearing aids in infants, nearly all the parents contributing to the focus group reported that they had not properly understood or appreciated the worth of this test. Whilst it is professionally informative to use ABR for confirming diagnosis, the discussions held revealed that it is not easy to communicate this information to parents. Although the ABR tests had all been undertaken by experienced and clinically registered audiologists, the results had not conveyed the information that these parents needed to understand the hearing loss and therefore their adoption of hearing aids depended almost entirely upon their acceptance of the professional advice offered to them. This was difficult because most of the parents were not receptive by the end of the ABR test session.

Conversely, the recording of CAEP responses to speech sounds followed the initial diagnostic electrophysiological tests in the infant audiology pathway and this test was found to be clearer. It had helped the parents to understand their child's hearing loss and the necessity for future audiological management. The demonstration of speech sounds that could and could not be heard was valued and it helped parents to realise the need for hearing aids. The CAEP results also demonstrated hearing aid benefit enabling parents to accept that their child was a candidate for cochlear implant assessment. Without the use of CAEP, those parents of children with a milder hearing loss agreed that they would not have accepted management with hearing aids. The test had therefore, helped them to make important management decisions with potentially long-term benefit to their child. However, once again the focus group participants highlighted the difficulties that parents had in assessing the outcome of the test during the clinical

appointment. They had not always been able to fully take on board the demonstration or the implications of their child's hearing of speech like sounds in the clinic. Explanations and guidance offered outside of the clinical environment and within the home were appreciated by all the participating parents. This home support had helped the families move forward and engage with intervention. These discussions were particularly important for partners who wanted to be involved in the decisions being made about their child, but who had not been able to be present during the demonstrative clinical tests. The understanding the family gained from the discussions and explanations in the home empowered them to make the necessary decisions about early hearing aid use and if necessary to consider referral of their child for cochlear implant assessment. A more recent study by Munro et al., (2019) showed that parents found CAEP testing to be a positive experience and recognised the benefit of having an assessment procedure that uses conversational level speech stimuli.

Despite very early implementation of universal neonatal screening, the adoption of hearing aids for infants with mild-to-moderate losses has remained a continuing challenge (Mehta et al., 2017). Sjoblad et al. (2001) investigated similar substantial delays between diagnosis and hearing aid fitting across 45 American states and they identified 3 primary factors of concern for families in the early stages of their child's hearing aid use. These were uncertainty around the maintenance of hearing aids, the appearance of the device on their child and lastly not understanding the benefit for the child. In the early stages of the hearing device fitting, 65% of parents questioned the degree to which hearing aids were of benefit and 30% of parents requested better education on which sounds hearing aids could make audible.. Walker et al. (2015) in their more recent investigation of hearing aid use in 290 children concluded that inconsistent use in infancy and early childhood could be improved by an audiological demonstration of the benefit afforded by the hearing aids. Punch et al. (2016) found that their introduction of the recording of CAEP responses to speech sounds in Australia provided this information for parents and that this test was therefore an effective counselling tool when hearing aids were fitted after the neonatal hearing screen. This finding was corroborated by the parents who made up our focus group.

Walker et al. (2015) also concluded that parents needed individualised problem-solving strategies to help them in their use of hearing aids. This conclusion had also been reached by Muñoz et al. (2015) in their investigation of the parent reported challenges of hearing aid use in young children. They emphasised the parental need for emotional support and when they surveyed 349 paediatric audiologists in the US, the majority recognised the need for paediatric audiologists to receive further training in counselling skills so that they could meet these emotional needs (Meibos et al., 2016). In the present study further discussions of the results of tests and their implications were undertaken within the home by a peer-to-peer support worker. This enabled decisions to be made by family members who had been unable to be present at the clinical tests. It also offered emotional support for parents as well as individual pragmatic help for them in solving challenges they were facing with the hearing aid fitting. The parents participating in our focus group agreed that this input had been crucial to their understanding of the hearing loss, and to their management with hearing devices.

The reported study highlighted some aspects of the infant audiology pathway that can be developed to improve the parental understanding and management of their child's PCHI. The study had obvious limitations in that only 8 families participated in the focus group, and these families were those who were willing and able to attend. Walker et al. (2015) found that the level of maternal education was an important predictor of hearing aid use in children. Although the London district where the focus group was held is generally an area of deprivation, the educational levels of the focus group were not atypical of those found in most other parts of the UK and in this respect, although replication with larger studies is required, the difficulties expressed by the reported focus group may be generalised. An additional limitation of the study was that the facilitator was an audiologist from the clinical practice that the parents attended and parents could have wanted to provide positive feedback. However, the role of audiologist was not a central question for the group, so should not have influenced the openness of the responses.

### **3.6 Conclusion**

We had previously reported an earlier age of entry to intervention with hearing aids and cochlear implants following the inclusion of CAEP recording into the infant audiology pathway and hypothesised that this arose in part because the CAEP recordings enabled parents to be better informed about the effect of the hearing loss and the impact of hearing aids. We have analysed the discussions that took place in a focus group of parents whose infants had received the CAEP test following confirmation of their child's deafness from the NHSP. The participants confirmed that recording of CAEP responses to speech sounds had enabled them to understand the impact of their child's hearing loss and the benefit of hearing aids. With help and guidance available from professional support they were empowered to make the management decisions required for their child. The views of the parents now having been established, the next hypothesis was to investigate clinicians' views on the CAEPs in the following experimental study.



## **CHAPTER 4: Clinicians' views of using Cortical Auditory Evoked Potentials (CAEP) in the permanent childhood hearing impairment patient pathway.**

### **4.1 Abstract**

The aim of this research was to obtain clinicians' views on the use of CAEP in the infant audiology pathway. A questionnaire aimed at clinicians who use the HEARLab system with the Aided Cortical Assessment (ACA) Module. Results compared for Australians (where HEARLab produced) to other countries. The questionnaire was completed by 49 clinicians; 33 from Australia and 13 clinicians outside of Australia and 3 clinicians, destination unknown. The findings of this research demonstrated that clinicians using CAEPs found them valuable for clinical practice. CAEPs were used to verify or modify hearing aid fittings and were used for counselling parents to reinforce the need for hearing aids. With the use of speech token as the stimulus clinicians had more relevant information to increase confidence in decision-making on paediatric hearing management. The main benefit from the use of CAEPs (using speech token stimuli) was for infant hearing aid fitting programmes, to facilitate earlier decisions relating to hearing aid fitting, for fine-tuning the aids and as an additional measure for cochlear implant referrals.

### **4.2 Introduction**

Neonatal hearing screening programme (NHSP) is one of the most important clinical advances for identifying hearing loss in children, leading to early management with hearing aids. The objective assessment of hearing loss continues to be refined in the development of clinical protocols. Despite early identification, reports persist of deaf children's language levels being below that of their hearing peers (Ching et al., 2013). The reasons for this are complex but may partly be due to under-amplification through hearing devices over the early months. The evaluation of early hearing aid fitting or cochlear implant candidacy for infants is challenging for audiologists due to the lack of tools to support clinical decision making, particularly between 3 and 8 months developmental age.

Assessing hearing using behavioural measures can typically be carried out reliably from around 6 months of age, and these measures are necessary for the audiologist to confidently prescribe appropriate amplification for the individual child. Hearing aid fitting can be verified using real ear measurements (REMs) which takes into account the size and shape of the individual child's ear canal (British Society of Audiology, 2007). REM targets allow audiologists to see the spectrum of sound delivered to the child. However, all the prescribed gains are based on estimated thresholds, which are difficult measurements to obtain reliably thus resulting in uncertainty. The current UK hearing testing protocol during the first 6 months of life relies on estimating hearing thresholds based on objective measures. These are typically a mixture of auditory brainstem response (ABR) and/or auditory steady state responses (ASSR) measurements. Audiologists then make recommendations for and fit hearing aids based on these estimates. Prescribed gains using this approach can often be quite conservative because of uncertainty surrounding the results. In particular, when audiologists have insufficient information on audibility of speech sounds because the children are too young to respond.

The use of REM verifies the sound levels in the infant's ear, but does not evaluate audibility of the signal for the child or the transmission through the auditory pathway to the brain. In the standard infant audiological pathways in the UK, it is not until 6-8 months of age when Visual Reinforcement Audiometry (VRA) is possible, that behavioural responses can be measured to gain a more accurate assessment of the child's access to speech sounds through their hearing aids. The networks in the auditory system for processing speech sounds for normal hearing infants are formed early in life (Sininger et al., 1999). Thus, when sensory input to the auditory nervous system is interrupted during early development, the morphology and functional properties of neurons in the central auditory pathways can result in atypical anatomic and physiologic development (Sininger et al., 1999). Therefore, by the time a hearing-impaired child is able to demonstrate behavioural responses in clinic, important periods for linguistic development may already have passed, if the child did not have audible signals with their hearing aids.

An additional objective measure which has the potential to provide speech-related objective responses indicating detection of sounds in the brain, are the measurements of cortical auditory evoked potentials (CAEPs). These are electrophysiological responses which originate from neurons at the level of the primary auditory cortex, and from the auditory association areas in the temporal lobe. There are three major components in the recorded response referred to as the P1-N1-P2 complex. In the early months of life, P1 component varies as a function of age, occurring around 300 ms in newborns, decreasing rapidly over the first 2-3 years of life, decreasing until reaching a mature adulthood latency around 60 ms (Sharma et al., 2015).

CAEPs can be used to evaluate aided hearing by testing detection thresholds for speech sounds through hearing aids without requiring an active behavioural response. CAEP recording is conducted while the infant is awake, thus avoiding the need for sedation or natural sleep which is required for ABR and ASSR testing. Consequently, they can give supplementary information for evaluating the responses to speech at specific presentation levels in young infants or difficult-to-test children. Sharma et al. (2006) has shown that the latency of the P1 and morphology of the CAEP responses change with the development of the central auditory pathways and that there is a maximal period of cortical plasticity in the first 3.5 years. For children receiving cochlear implants, if they are implanted below this age they often achieve age-appropriate cortical responses within 3–6 months after stimulation. Cardon et al. (2012) has shown two major principles of neuroplasticity direct clinical outcomes: 1) adequate stimulation provided to the cortex and 2) appropriate timing of stimulation through hearing aids or cochlear implants. Early intervention with appropriate auditory input results in high likelihood of normal auditory cortical development in children with congenital deafness.

Munro et al. (2011) tested sound field CAEPs (using HEARLab) on 24 normal-hearing adults, with and without earplugs. The results showed that the CAEP response detection was good except for the lowest presentation level. They stated that this most likely occurs when the stimuli are within 10 dB of the behavioural threshold. Punch et al. (2016) reported a retrospective review of 83 infants with hearing loss in Australia. The infants were fitted with hearing aids and

underwent aided CAEP testing to confirm their hearing loss and to evaluate the initial hearing aid fittings. Their findings showed that CAEP testing facilitated rehabilitation and CAEP testing is now part of routine infant hearing aid fitting programmes in Australia. The introduction of CAEPs were easily implemented in Australia because there is a single organisation that takes care of hearing impaired children, this is Hearing Australia. National Acoustic Laboratories provided the scientific evidence (CAEP presence is correlated with audibility; CAEPs increase with hearing aid gain; the automatic detection works as well as an observer), and built a clinical device. The HEARLab was also used in Australia to test children with auditory neuropathy spectrum disorder (ANSO) where ABR responses or early latency electrophysiological responses were not observed and CAEPs are used to estimate audiograms (Pearce et al., 2007 & Rance et al., 2002).

The current work has expanded on the Punch et al. (2016) study by exploring the viewpoints and attitudes of clinicians towards HEARLab. In addition, attitudes outside of Australia were evaluated, in regions where the use of cortical measurements is not part of the clinical routine and it was determined how the equipment is used in the infant hearing pathways.

Whilst children with severe-to-profound hearing loss are at risk for speech, language, social and emotional difficulties, there is uncertainty for those with a milder degree of hearing loss and are often placed on a programme to monitor and record hearing and speech development (Bagatto et al., 2013). Audiologists face additional challenges when recommending hearing aids for milder degrees of hearing loss because of the uncertainty around the benefits of amplification (Bagatto et al., 2013; Fitzpatrick et al., 2014). Mehta et al. (2017) showed that there can be delays in hearing device provision for mild-to-moderate hearing loss due to parental and potentially audiologist uncertainty about the benefits of, or requirements for, hearing aids. They demonstrated that with the introduction of CAEPs the median age of hearing aid fitting was reduced from 9.2 to 3.9 months for all extents of hearing loss, but for cases with the mild-to-moderate hearing loss, the median age for hearing aid fitting reduced from 19 to 5 months. It was suggested that the CAEPs offered an educative process for the parents and

audiologists to support decision-making for hearing aid fitting, particularly for those infants with a milder degree of hearing loss.

Subsequently, Mehta et al. (2019; also chapter 3) explored the idea that the CAEPs may have influenced parental decisions. The findings from that study demonstrated that the use of CAEPs was a major factor in helping families appreciate that hearing aids could be beneficial for their child because of interpreting the presence of a response. Parents found the overall procedure, stimuli and visibility of results valuable.

In order to understand the views of clinicians using CAEP testing as part of hearing management for children referred from the NHSP, a questionnaire was developed. This questionnaire was specifically focussed on understanding the benefits of a commercially available system, HEARLab but it is believed that these findings are generally relevant to all clinically available cortical response measurement systems. The study reported the opinions of audiologists on the use of CAEPs, in particular for those using the HEARLab system with the ACA Module. One of the features of the HEARLab equipment is that it uses speech tokens /m/,/g/ and /t/ which were extracted from running speech. These stimuli are used to assess aided and unaided responses at different stimuli presentation levels to determine the level at which the speech token(s) are detected by the child. HEARLab uses an automated statistical analysis of the waveforms, therefore clinicians do not rely on their visual analysis to indicate if a response is absent or present. The findings may therefore be relevant for any cortical response system using speech-like tokens to assess aided responses.

The intention of this current study was to evaluate the effectiveness of the equipment in countries outside of Australia where routine infant audiology pathways do not include CAEP measurements, as well as updating the Australian viewpoint. The research questions aimed to determine the following:

How does the use of CAEP impact on confidence for patient management decisions (RQ1),

What are the stages in the patient pathway where CAEPs are most helpful in the clinical management of infants with hearing loss (RQ2)?

Which patient groups benefit most from the use of CAEPs (RQ3),  
In the clinicians' view, what is the impact of having CAEP results for parents of children with hearing loss (RQ4),  
What are the overall clinical benefits that may be derived (RQ5).

### **4.3 Methods**

A questionnaire had been developed for clinicians who use the HEARLab system with the ACA Module (Punch et al., 2016). This was adapted for the current purpose following feedback from audiologists and expert reviewers. The original questionnaire contained statements that the respondents agreed/disagreed with, choosing their responses from a Likert scale.

Two audiologists initially reviewed the original Punch et al. (2016) questionnaire and revised the questionnaire adding a section on demographics and removing some sections from the original that were not relevant outside Australia. The excluded statements related to training and installation of equipment, additional features such as impedance checking, electrode problems, adverse reaction issues and report writing.

#### **4.3.1 Expert review of the questionnaire**

A first draft of the questionnaire was sent to three additional experts for review of content and clarity. The panel included one clinical scientist, one speech and language therapist and one research physiologist specialising in auditory evoked potentials. All three had backgrounds in audiology and deafness. The feedback suggested minor amendments to the wording and grammar, which were revised for the new version of the questionnaire. Some additional statements were added to the adapted questionnaire as advised by the experts. These statements were:

- The results of HEARLab do not change my approach to rehabilitation
- The HEARLab is important to my clinical practice

- The information HEARLab provides in achieving optimal habitation outcomes for children has been useful in clinical practice

The questions in the final version of the questionnaire can be seen in Appendix 1. An online version of the questionnaire was created in UCL OPINIO (Object Planet, Inc., n.d.) survey platform. An e-mail was sent to potential participants informing them of the study and the link to the questionnaire. Ethical approval to conduct the study was given by UCL research ethics department: project ID: 9781/001.

The British Society of Audiology and the British Academy of Audiology administrative teams circulated the online link to the questionnaire to their members. All audiology departments using a HEARLab system in the UK, Australia and worldwide were sent a link to the questionnaire from the National Acoustic Laboratories in Sydney, Australia (developers of HEARLab). The online questionnaire included a section to indicate consent prior to participating in the on-line questionnaire. All audiology departments using a HEARLab system in the UK, Australia and worldwide received the link which was forwarded by the National Acoustic Laboratories in Sydney, Australia (developers of HEARLab). The online questionnaire contained a section on consent to participate before respondents could complete the online questionnaire.

Part 1 of the questionnaire (Q1-Q3), included demographic questions about the clinicians and how much experience they had with HEARLab and with whom they used the equipment. Part 2 of the questionnaire (Q4-Q11), aimed to find out the management pathways that were most appropriate for including the CAEPs; Q12-Q22 addressed clinicians' views on the equipment. The response format used the Likert five point scales. For the analysis the 'strongly agree' and 'agree' options responses were collapsed together as were the 'strongly disagree' and 'disagree'. The responses were compared between Australia vs other-countries because the equipment has been available in Australia for longer with better support and training.

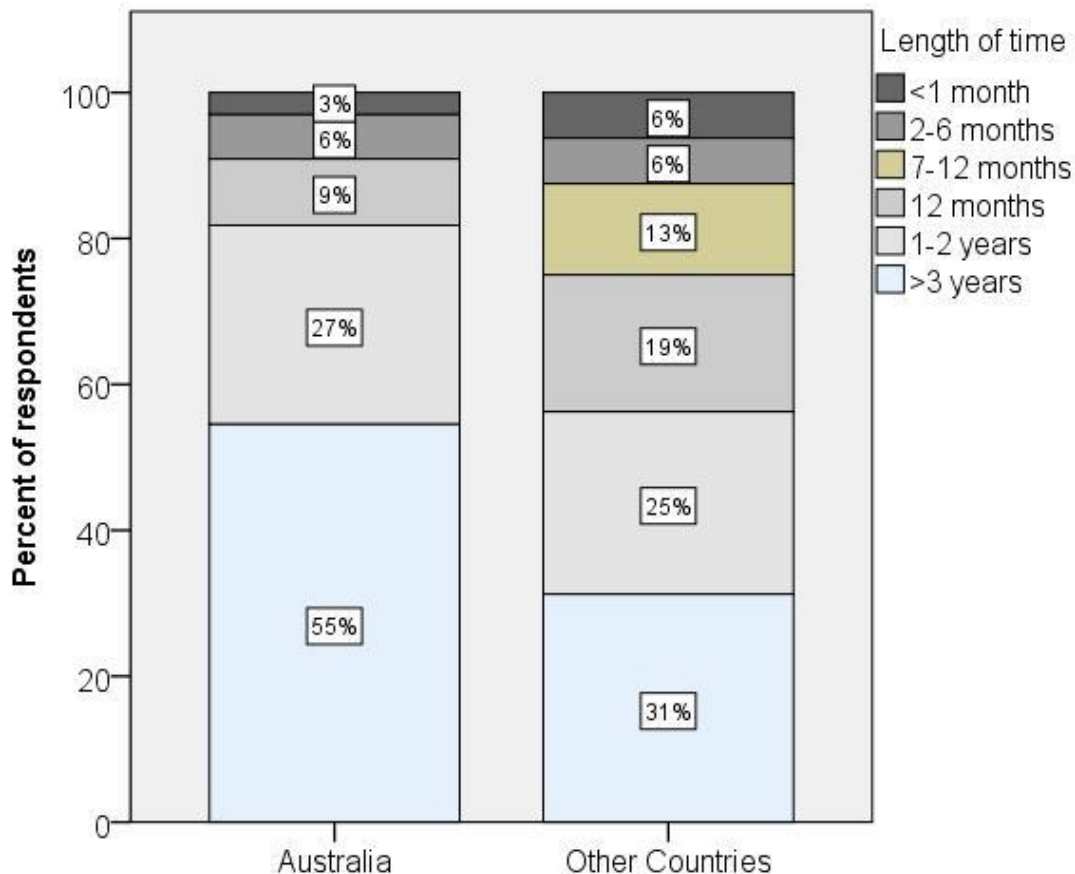
## **4.4 Results**

### **4.4.1 Respondents**

The online questionnaire was completed by 48 audiologists and 1 clinical university professor. The responses were collected from Australia (33), 16 clinicians outside of Australia (Turkey (2), UK (7), New Zealand (2), Taiwan (1), United States of America (1)) and 3 which were completed from unknown countries outside Australia.

The results showed that 47% of clinicians that completed this survey had >3 years' experience in using the HEARLab equipment and 49% had completed between 11-50 assessments. When the data was categorised into Australians versus other-countries 55% of clinics from Australia have >3 years' experience, compared to 31% of other-countries with >3 years' experience see figure 4.1.





**Figure 4.1** Distribution of the length of HEARLab experience of the respondents as percentage of Australians (N=33) and other countries (N=16).

#### 4.4.2 Research Q1. Does the use of CAEPs make audiologists more confident about patient management?

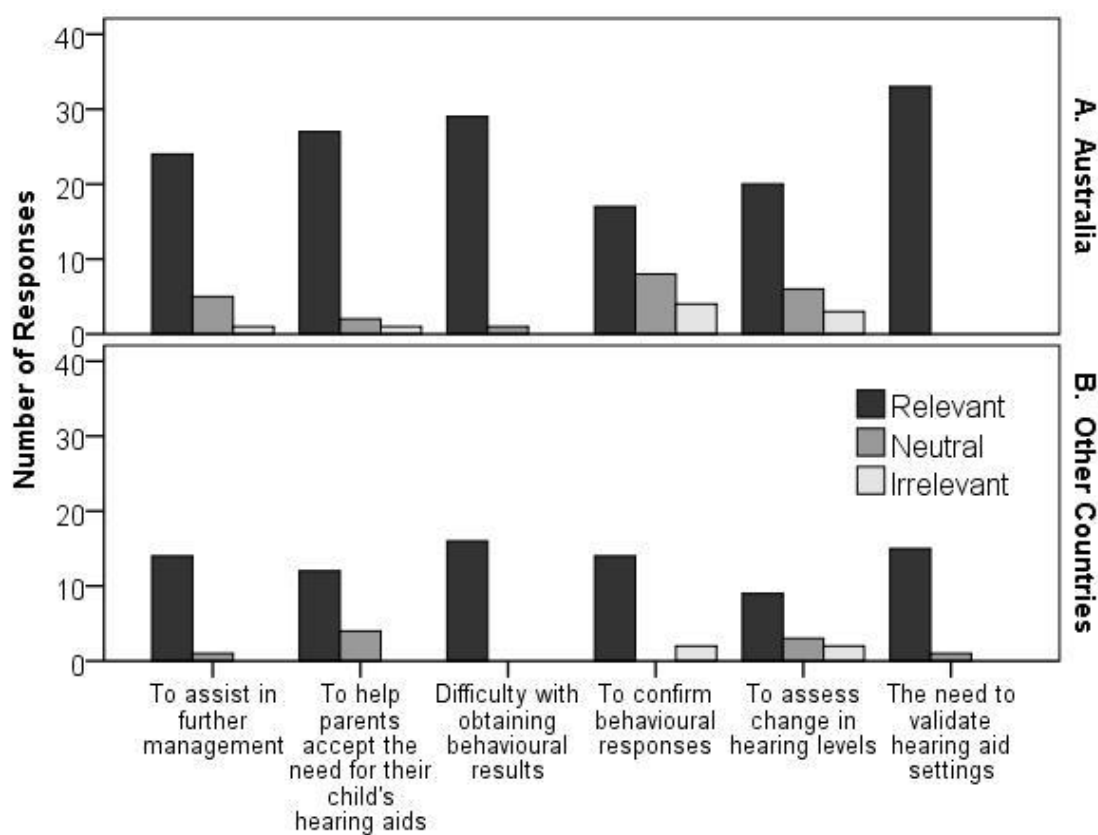
The primary aim of the research was to assess clinicians' views of CAEP use in the management of infant hearing loss. Answers to questions 4-8 were averaged. These questions in the survey were specifically looking at patient management. There was high agreement that CAEP made audiologists more confident in patient management. When responses were separated into Australia and other-countries, the findings were that 73% of responses from Australia agreed that CAEPs made clinicians more confident and 27% clinicians disagreed. This is compared to other-countries where 63% of clinicians agreed and 37% disagreed. Overall, there is a strong agreement that CAEPs made audiologists more

confident in their management process. Clinicians commented that they used the equipment *'for children with mild/moderate hearing loss to determine whether to fit hearing aids.'*

Across countries, 91% of clinicians use the HEARLab to validate hearing aid settings. When data was compared by region it showed that 100% of Australians and 93% of other-countries use the CAEPs to validate hearing aid settings. With respect to provision of hearing aids 87% of clinicians' reported basing a decision to provide hearing aids on aided cortical responses (across regions) and 52% used the cortical responses to help adjust the hearing aid setting. A comment on the survey by one of the clinicians stated that: *'we have had excellent success with improvement in performance following hearing aid settings based on cortical assessment'*. However, there were clinicians that also commented *'that they do not feel confident that the HEARLab provides useful and reliable information'*. The questionnaire responses indicated that 45% of clinicians had something negative to say about the HEARLab and 55% of clinicians had something positive to say about the equipment. Some of the negative comments about the equipment were generally referring to the HEARLab equipment itself. Examples of the negative comments about the equipment were that it was *'very temperamental, have problems with the system very often'*, *'sometimes it is difficult to identify which peak is the real P1'*, *'the traffic light button takes a long time before it goes green while testing'*, *'a smaller unit would be more practical'* and *'poor connection between electrodes and pre-amplifier'*.

The survey also demonstrated that approximately 67% of clinicians use cortical responses to assess changes in hearing levels. The survey has shown that 85% of clinicians used the visualisation of presence/absence of responses to help parents understand and accept the need for their child to wear hearing aids with 77% of professionals agreeing that parents were positive about the use of the equipment. When broken down by region the data demonstrated that 81% of clinicians from Australia and 75% of other-countries used the CAEP results to help parents. The results of the CAEPs allowed audiologists to discuss the potential management options with parents, and show aided benefits, where they could not in the past. One of the clinicians commented that the *'ability to discuss with families the results from the HEARLab has helped guide management*

decisions.’ However, some clinicians commented ‘we found it difficult to discuss the results when no reliable responses were found due to movement or an unsettled child.’ This experience would most likely be the same for all objective tests but some clinicians found it frustrating that they were not always able to comment on the aided results, and in those cases that this disengaged families from the management discussions. When separating the results into Australian clinicians versus other-countries the consensus regarding CAEP showed similar proportions agreeing on relevance (figure 4.2).



**Figure 4.2** The proportion of positive responses from clinicians, in areas that the HEARLab is used in clinical practice and in patients’ pathways. Categorized A) responses from Australia and B) responses from other countries.

#### 4.4.3 Clinical benefits of the CAEPs

##### Research Q2. Stages in the patient pathway in which CAEPs are most helpful?

As can be seen in figure 4.3 (results of Q10), the results showed that 26% from Australia and 29% of professionals from other-countries use HEARLab at the follow-up after the initial hearing aid fitting and 20% from Australia and 25% from other-countries use the equipment with older children who are difficult to condition behaviourally. One clinician supported this finding by saying that they used the equipment for *'children diagnosed via ABR with slight or mild hearing loss to determine amplification recommendation.'* *The survey has shown that 18% from both groups showed that clinicians used the HEARLab for cochlear implant candidacy. Clinicians use the results from CAEP to refer children for cochlear implant assessment. Where aided CAEP responses are absent, the results may help parents and audiologists move towards referral for cochlear implants. A clinician's comment was that CAEPs 'can provide information about whether infants/toddlers/young children/difficult to test clients are getting sufficient benefit from their hearing aids, whether they require adjustments or referral for cochlear implant candidacy assessment.'*

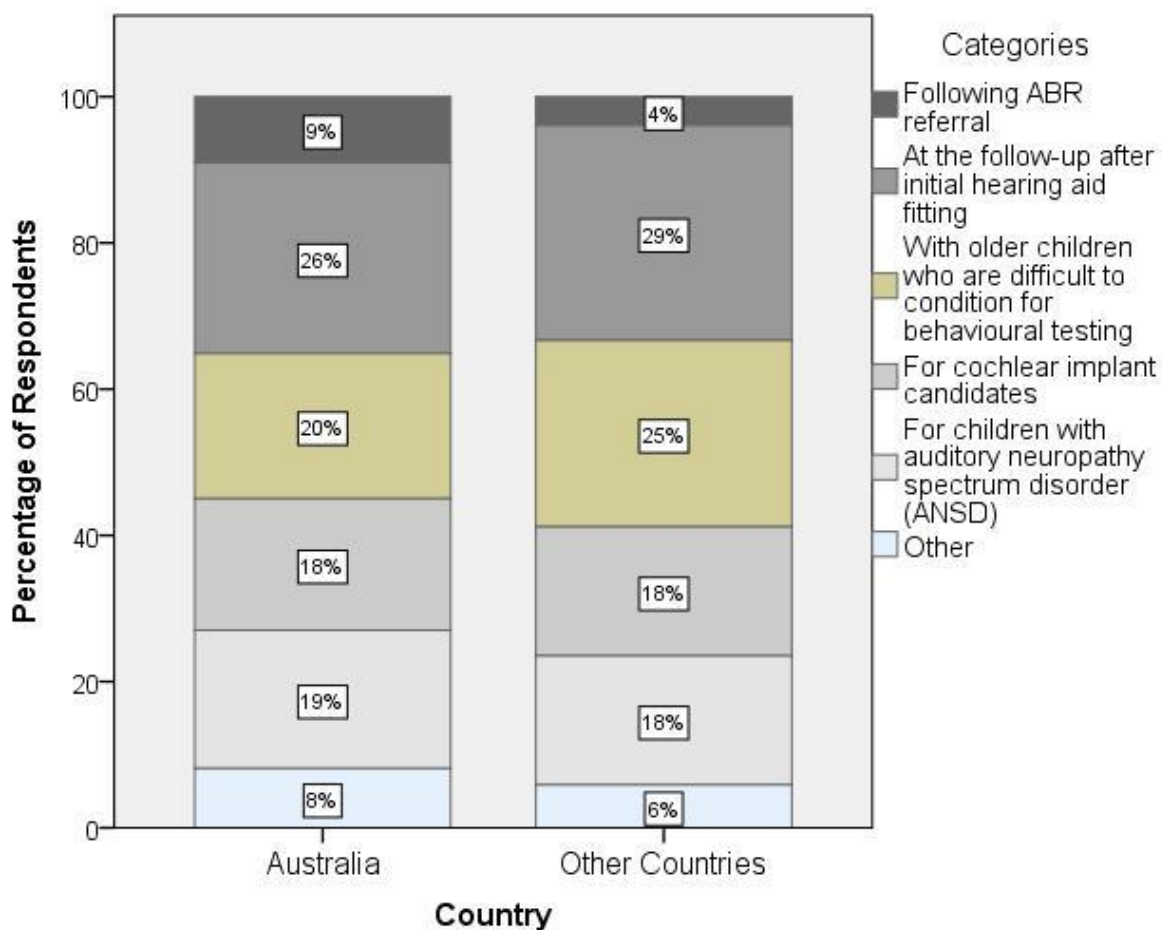
The results have also shown that the percentage of clinicians using the CAEPs following ABRs are low (9% in Australia and 4% from other-countries), showing that clinicians predominantly rely on ABRs as their preferred method for assessing hearing acuity. The CAEPs are typically used either before hearing aid fitting for clinicians to decide whether to aid a child or after the fitting to show aided responses.

The CAEPs were mainly used for determining appropriateness of amplification with 84% of clinicians' claiming that they used CAEPs to determine potential benefit of hearing aids. Clinician comments included the following:

*'Yes - the information (in combination with other sources of information such as parent questionnaires) are used to determine if hearing aids are adequate, if hearing aid settings need to be adjusted, and if cochlear implantation should be considered.'*

*'I use the results to make adjustments to hearing aids before behavioural testing is possible/reliable (7-8 months of age).'*

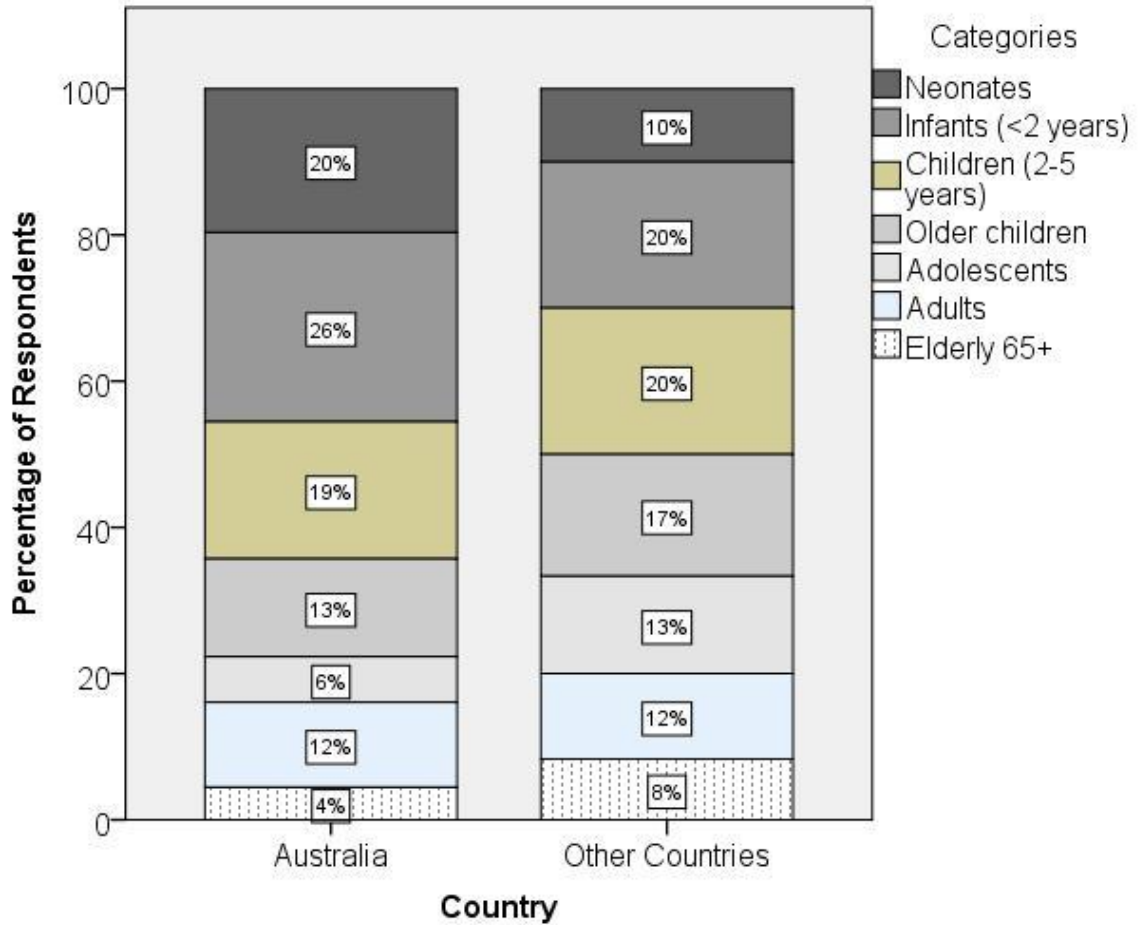
Findings from question 5 from the survey showed that 92% of clinicians used aided CAEPs to fine-tune the aids once fitted. In the Australian group, 94% of clinicians agreed that CAEPs helped them to adjust the aids and 88% of clinicians from other-countries reported the same. One clinician reported that *'they have found excellent success with improvement in performance following hearing aid changes based on the cortical assessments.'*



**Figure 4.3** Distribution of patient pathways that clinicians use the HEARLab system for and responses shown in relative percentages grouped by Australia and other-countries.

#### **4.4.4 Research Q3. Which patient groups benefit most from CAEPs?**

Results from question 11 from the survey showed that CAEPs were most helpful for infants at < 6 months and under 2 years of age. It showed that 20% of the Australian clinicians use the CAEPs as supplementary to the ABR results, on neonates however, a much smaller proportion (10%) of clinicians from other-countries do this. Twenty percent of clinicians from other-countries used the CAEPs on infants under 2 years of age and with children aged between 2-5 years but it was not so common for very young children. In general, CAEPs are used for children where no reliable responses can be obtained. Figure 4.4 shows that 46% of clinicians from Australia and 30% from other-countries use the equipment on children under the age of 2 years and 19% from Australia and 17% from other-countries use it on older children to assess their hearing levels. Only 12% from both groups of respondents used CAEPs with adults and the elderly population.



**Figure 4.4** Patient populations for which the HEARLab system is used comparing Australia to other countries.

#### 4.4.5 Research Q4. What are the clinicians' opinions of parental views of the CAEPs?

Question 12 from the questionnaire was used to answer this question. The results of the questionnaire show that 80% of clinicians from both groups believe that the results from CAEPs are positively received by parents. When the groups were separated into Australians and other-countries there is a similar pattern of results in both groups, with 84% of clinicians from the Australian group and 88% of clinicians from other-countries observing that the HEARLab results are positively received by parents. Clinicians and parents both find the results easy to interpret. One clinician said that *'parents understand the analysis easier as they are able to relate to the sounds'* however, not everyone was positive. A different

clinician reported that *'parents mentioned that they found the sounds were cumbersome and seemed artificial, not like /m/, /g/ and /t.'*

#### **4.4.6 Research Q5. The overall views on the clinical measurement of CAEPs with the HEARLab equipment**

##### **4.4.6.1 Clinical viability**

In general, the results from question 18 showed that the HEARLab equipment was specifically well received by the respondents with 89% of clinicians stating that they would recommend it to other audiologists. Question 19 revealed that 92% of clinicians found the HEARLab to be important to their clinical practice. One clinician responded and said *'it is essential to clinical practice for infants fitted at under 6 months and even for those under 12 months and certain patient groups.'* Furthermore, results from question 20 shown that 86% of professionals feel that the information that HEARLab provides optimal habilitation outcomes and 67% of clinicians do change their rehabilitation approach depending on the results. A clinician said *'I always use HEARLab as part of a test battery and management/rehabilitation decisions are made once all information is looked at correctly.'* Question 13 revealed that 26% of clinicians take 15-20 minutes to complete the measurements using the three speech sounds at one intensity and 28% of clinicians said that it took 21-30 minutes. So the majority suggested that 30 minutes was sufficient to complete the testing at one level. However, this time could vary due to a child's arousal state and restlessness, with 61% reporting that one of the major factors preventing test completion was the restlessness of the children.

Overall, audiologists have found the HEARLab equipment valuable to both themselves and families and discussion of the results provided a way to fill the gap between diagnosis of the hearing loss and the point at which behavioural testing could take place. As mentioned by one of the clinicians, *'the HEARLab is one more tool which helps to verify intervention.'* Another commented that *'every clinic dealing with infants' hearing aid fittings should use CAEPs before behavioural testing is possible.'*



#### **4.4.6.2 Clinical views of the speech tokens (Q14)**

This revealed that clinicians found the results easy to interpret and that the detection results for the /g/ stimulus in particular are consistent with the behavioural results. This was found in responses to question 14: 80% of clinicians answered that the /g/ token was similar to the behavioural results and 67% found the /t/ sound to be similar to behavioural results. Clinicians commented that /m/ is most affected by the child's activity level and middle ear status and one of the clinicians commented that *"/m/ is more likely to be absent even if the child is hearing the sound."* Clinicians reported that 39% of /m/ token correlated to the child's behavioural responses. This shows that clinicians mainly use the /g/ and /t/ stimulus for testing. The survey to question 6 demonstrated that 81% of clinicians feel that the results from CAEPs correlated with behavioural thresholds.

#### **4.4.6.3 Difficulties with clinical equipment (Q16)**

Some clinicians reported technical difficulties e.g. with the size of the HEARLab equipment and with poor connections between electrodes and the pre-amplifier. The equipment also requires ongoing repairs and can be 'temperamental'. About 44% percent of clinicians have reported that excessive epoch rejection has meant that clinicians are unable to carry on testing, which can be time consuming. One clinician commented that *'I don't like it, it takes so long for each speech sound, 5-7 minutes on average – it is very hard for many little children to be kept still.'* Around 25% of clinicians who question the reliability of the results, also the size of the equipment and one of the clinicians commented, *'it is too black and white and the equipment is very inflexible.'*

### **4.5 Discussion**

The aim of this study was to gather clinicians' viewpoints of the CAEPs using the HEARLab in the infant audiology pathway. This research specifically focussed on the HEARLab equipment but the findings are generalizable to other CAEP systems which use speech tokens.

Punch et al. (2016) developed a questionnaire to evaluate the use of HEARLab equipment with Australian clinicians. One of our goals was to compare the experiences in other countries with those of Australian clinicians. The findings from this study were similar to those of Punch et al. (2016), the majority of clinicians who routinely used the HEARLab system reported that it was a valuable component in their clinical test battery. The results have shown that clinicians feel confident about using the HEARLab equipment and found it helpful for adjusting and fine-tuning hearing aids. The measures allow clinicians to ensure that adequate amplification is provided and appropriate speech input levels can be achieved, thus increasing the chances that the child will hear spoken language and this will contribute to/enhance their speech and language development. The clinicians also revealed that the HEARLab was inflexible and time consuming.

Additionally to the above, the current study has shown that clinicians from other countries and Australia who completed the questionnaire felt more confident about patient management, especially for the age groups between 4 and 12 months when other behavioural testing is limited and hearing aids have to be fitted based on estimated audiograms. The results from the HEARLab were easy to interpret especially for parents. Furthermore, this study found the results of CAEPs helpful in decision making and were used for counselling parents to reinforce the need for hearing aids. These findings were different to the Punch et al. (2016) which showed that parents were against the use of the test and preferred to wait until behavioural responses and they reported that parents found it hard to relate CAEP results. The findings of this research demonstrate that even though CAEPs have not routinely been used in general clinical practice outside Australia, those clinicians using CAEPs find them valuable for clinical practice. The numbers of non-Australia respondents is small because it is an emerging technique. A review of the literature to determine the proportion of articles outside of Australia using CAEPs to speech sounds identified 31 articles that discussed using CAEPs and of those 11 did not have any Australian authors. This reflects that CAEPs are emerging as an area of clinical importance outside of Australia. The non-Australian viewpoints represented one third of the respondents in this research and the comparison with the Australian respondents is interesting. Outside of Australia the support network is not as strong and there

are different healthcare systems influencing behaviour with different pressures on audiological services due to funding and service provision approaches. The smaller number of respondents outside of Australia was expected and has been raised in the study limitations.

Overall, CAEPs have shown their value in the clinical environment as audiologists are able to show parents aided benefit, especially for those with mild hearing losses and also for children with complex need's. It has been reported to be a great counselling tool for parents. In addition, it helped the audiologists themselves understand the need for aiding and provided a tool for fine-tuning the hearing aids. The results indicated that approximately 50% of clinicians use the CAEPs for children <5 years of age. The use of CAEPs have helped clinicians confirm the child's hearing loss and reduce the ambiguity regarding appropriate clinical interventions for children with mild hearing loss. With the use of speech token CAEPs clinicians are able to make a confident decision on the child's further management. The major difference between the two regions is that Australian clinicians are using CAEPs more routinely in clinics and are a part of their clinical protocol, which is not the same for other countries.

The current results showed CAEPs to be useful at different stages in the infant audiology pathway. The questionnaire demonstrated that CAEPs were particularly useful after the initial hearing aid fitting. Clinicians reported a positive response from parents because it was easy for them to observe the aided benefit because of the simple visual display of the CAEP measurements when a response was present. It was reported in the questionnaire how CAEPs were helpful with the management of babies who had mild hearing loss. Audiologists around the world face difficulties when deciding whether to fit hearing aids on children who have mild-to-moderate hearing loss. It has been demonstrated by Fitzpatrick et al. (2010) that there is ambiguity regarding appropriate clinical interventions for children with this level of hearing loss particularly involving the need for audiological management. In the cases where the stimulus presentation did not elicit a response the test helped parents and professionals to know when to consider moving towards cochlear implant referral. The CAEPs have allowed clinicians and parents to see if responses are present to speech tokens, opening

up opportunities for clinicians to speak to parents using meaningful results about further management.

Sjoblad et al. (2001) demonstrated that 65% of parents in the early stages of the hearing device fitting questioned aided benefit and 30% of parents requested better education around the benefit that the aids provided to their child. The current study showed that clinicians found that the use of CAEPs were positively received by families. It has highlighted that clinicians from Australia and other countries felt that families valued the demonstration of perception of speech sounds and used this tool as a discussion point for management. Mehta et al. (2017) have demonstrated the importance for families to be educated on hearing loss and hearing aids and CAEPs were a key element for intervention. Their study showed that parents who defer hearing aid fittings fell from 40% to 19% for children who had a mild or moderate hearing loss when CAEPs were used to demonstrate access to sound to parents. They were also able to show 82% of cases with aided benefit through the CAEPs, which motivated the parents to support their child's consistent use of hearing aids.

The data have shown that fewer than 10% of clinicians in both Australia and other countries use it immediately after an auditory brainstem response (ABR) referral. An ABR referral occurs in some countries after failing the neonatal hearing screen as part of the post-screen diagnostic test protocol. Our survey has shown that ABRs are the standard procedure used in NHSP to verify hearing acuity. ABRs are the initial stages of diagnosis and are useful in obtaining thresholds in babies. CAEPs are used to supplement initial ABR results when the child needs to be asleep. Thus, not many clinicians are using CAEPs at this initial stage in the infant audiology pathway.

Fortnum et al. (1996), showed that 1/1000 in the UK are born deaf, with up to 40% of deaf children showing additional health, social or educational needs ranging from dyslexia to more severe disabilities such as cerebral palsy and Down's syndrome. In Australia 3.3/1000, children are affected by hearing loss (Australian Hearing, 2017) and Ching et al. (2013) has shown that 27% of hearing-impaired children have additional disabilities. Research conducted by McCracken & Pettitt (2011) indicated that patients with complex needs such as

autism or cerebral palsy tend to have a later diagnosis of hearing loss than those without. Their research revealed that 14 parents out of 51 identified significant delays in both achieving detailed assessment and their child being fitted with amplification. Under a third of children in their study were diagnosed by 6 months and similar numbers were not diagnosed until they were at least a year old. Such delays meant that these children were not appropriately aided and crucial time was lost in the early months of language acquisition. CAEPs are an objective measure and are therefore, not reliant upon a behavioural response which makes it an important tool for assessing children with complex needs to avoid delays in the provision of amplification. The data presented in this study have shown that around 20% of all respondents use CAEPs for assessment of children with additional complex needs and it is considered by those using it to be a valuable measure; it provides a systematic and objective means of indicating some detection of frequency information at low, mid and high levels.

As with any equipment there are drawbacks and the results revealed that the specific HEARLab equipment has some disadvantages and some clinicians expressed how restrictive the equipment is. Clinicians found the equipment bulky and 'temperamental'. In addition, clinicians indicated that they would like to be able to change the stimulus. Another drawback of the equipment is how restless children sometimes can prevent results from being obtained. Even with this disadvantage, 69% of clinicians reported that the assessment takes around 30 minutes to complete.

One of the limitations of this study is that only 13 clinicians from other countries took part in this study, compared to 49 from Australia. This reflects the widespread use of HEARLab in Australia and that the equipment is used less routinely outside Australia. Clinical experience from Australia shows that they view CAEPs as an important part of their clinical audiology diagnostic toolbox. Between the two groups it shows that 55% of the Australian group have >3 years of experience with the use of the CAEPs compared to 31% of clinicians from other countries. This shows that clinicians from Australia are more experienced and confident in using CAEPs than clinicians from other countries. This may also result from the fact that the other countries are not supported by the National Acoustic Laboratories as closely as the Australian clinics and thus, this should be

considered when interpreting the results. The questionnaire showed that there are peer-to-peer working support groups established in Australia, which is not currently routinely available in other countries. Australian clinicians have more training opportunities on the CAEPs. The lack of support and no requirement or guidance about routine use of CAEPs means clinicians in other countries may not be so aware of how to interpret the results. HEARLab was the only system at the time of research that used speech tokens and equipment that was easy to use in a clinical setting but other similar CAEP equipment can be used to help assess patients in the infant audiology pathway. This questionnaire demonstrates that the equipment is useful for supporting clinical decisions and that the use of speech stimuli makes the measurements meaningful for understanding access to speech sounds. It is recommended that more manufacturers should consider using speech tokens in their CAEP equipment.

#### **4.6 Conclusion**

CAEPs can be effectively used at many stages in the infant audiology pathway providing valuable information where behavioural testing is unreliable or impossible for babies and infants and is a useful counselling tool for parents to be able to observe impact of hearing loss on speech and benefits of hearing aids. A major area where CAEPs that use speech tokens are beneficial is with infant hearing aid fitting programmes, fine-tuning the aids and providing an additional measure to facilitate earlier patient management decisions relating to hearing aid fitting or cochlear implant referrals. The CAEPs were reported to be important in the complex needs population where routine assessment is not always possible. Specifically in our research, we questioned clinicians using the HEARLab equipment of the ACA module to determine the usage outside of Australia where the equipment was developed and is supported. The general pattern of response was similar when comparing Australia to other countries with general trends suggesting that it was a crucial part of the infant audiology pathway.

## **CHAPTER 5: Discussion**

### **5.1 Overview of findings**

The research in this thesis has reported on the findings from a clinic in the UK which implemented the measurement of CAEPs using HEARLab in the infant diagnostic pathways following the NHSP. The HEARLab system is an efficient diagnostic measurement system because of the automated statistical analysis approach for determining whether a response is present or absent. This makes the system a supportive tool for clinicians and allows non-expert clinicians to use the technology appropriately (Golding et al., 2009; Van Dun et al. 2015; Chang et al. 2015) to evaluate audibility of sounds for infants with SNHL. Their research demonstrated that the presence or absence of CAEP responses, defined by the automatic statistical criteria, were effective in showing whether increased sensation levels provided by amplification was sufficient for sounds to be detected in the cortex. Thus, this has meant that clinicians can see whether hearing aids are likely to be beneficial.

In Australia, the HEARLab was introduced routinely into the Australian paediatric clinical pathway (King et al., 2014 & Punch et al., 2016). Results from the Punch et al. (2016) found that CAEPs measurements are an extremely useful addition to the paediatric hearing aid evaluation test battery. The overall aim of the work in this thesis was to determine if it was worth introducing the CAEPs into the infant audiology pathway in the UK following NHSP and whether this implementation was acceptable to clinicians and parents and if it had an effect of the clinical interventions. The CAEPs were measured soon after the ABRs were recorded in both unaided and aided conditions. The age at hearing device intervention was assessed, taking into account the degree of hearing impairment. We found that the introduction of CAEP measurements facilitated earlier hearing aid fitting for children with mild-moderate impairments. Those with profound hearing loss were also referred for cochlear implant assessment at a significantly earlier age following the introduction of CAEPs, however this was in line with a national trend for earlier implant referral (Wood et al. 2015). We found that compliance with early hearing aid use and referral for cochlear implants depended upon parents' understanding of their infant's hearing loss. Their understanding was enhanced

by the demonstration hearing test results and the benefit of amplification using the CAEPs. Clinicians reported that the main beneficial uses for CAEPs (using speech token stimuli) were to facilitate earlier decisions relating to hearing aid fitting, fine-tuning the aids and as an additional measure to support cochlear implant referrals. CAEP measurements provided additional information to support more traditional assessment techniques, such as Behavioural Observation Audiometry, as a means of confirming that sounds with spectral energy predominantly in the low, mid and high-frequency regions are all audible to the child using hearing aids. When CAEP responses were repeatedly absent, even after adjusting the hearing aid gain it helped parents and professionals to make the decision to move forwards for cochlear implant evaluation.

Being able to use CAEP measurements at a time point between diagnosis and the stage where the infants were able to be assessed behaviourally at 6-8 months of age, makes it an important resource for clinicians. One of the most dramatic areas that the CAEP measurement was able to influence was the detection of mild-moderate losses and the associated hearing aid provision. Currently in the U.K., there is a great deal of uncertainty about the best treatment approach for infants with mild to moderate hearing losses. Some follow a “watchful waiting” approach, only providing hearing aids if the child does not progress well and others choose to try out hearing aids, However, there is concern that they could be over amplifying; there is little guidance to support such clinical decision making. A Canadian study of 337 children identified with mild bilateral or unilateral losses between 1990 and 2010 found that 87% of children were recommended to receive amplification, and 50% of the 337 had a considerable delay of 12 months or more for the fitting of hearing aids. They reported that of 164 children that had mild bilateral hearing loss who were not initially recommended for hearing aids, 73% (121) eventually received aids. One of the limitations of this study is that it did not measure any language outcomes. (Fitzpatrick et al., 2013).

Wood et al. (2015) have shown that with the introduction of NHSP, hearing aids are now being fitted on average within 82 days. The question now arises, ‘are these aids been consistently worn?’ Previous research has shown that poor compliance rates in the use of amplification may be due to parental belief that



their child hears sufficiently well without hearing aids (Walker et al., 2015). The current research has shown that CAEPs were used to demonstrate aided benefit to parents, which helped encourage consistent hearing aid use. We obtained parental perspectives on the introduction of CAEPs in the infant audiology pathway to find out if the results from the CAEPs were a contributing factor for earlier intervention. The results from this study showed that parents of children with a milder and moderate hearing loss agreed that they would not have accepted management with hearing aids if it was not for the use of CAEPs. Parents may reject hearing aid fitting unless they recognise their child's deafness and the impact that it will have on development (Watkin & Baldwin 1999). These results showed that parents valued the indicators on the CAEP machine that demonstrated if speech sounds could or could not be heard and it helped parents appreciate the need for hearing aids. Thus, the CAEPs were able to help parents understand what speech information their child is potentially missing, and recognise the difficulties facing their child. The results showed that parents reported that CAEPs had helped them to understand their child's hearing loss and need for intervention. A more recent study by Munro et al. (2019) reported on the feasibility, and parent acceptability, of recording infant CAEPs. They constructed an 8-item questionnaire (7-point scale, 1 being best) and results showed a mean score on the acceptability questionnaire ranged from 1.1 to 2.6. Their interviews with parents found CAEP testing to be a positive experience and recognised the benefit of having an assessment procedure that uses conversational level speech stimuli. This shows that there is a role for CAEPs in the infant audiology pathway to help reassure parents and supplement existing practice.

With the use of CAEPs, we were able to give more information to parents about the benefits of amplification. This gave us an insight into how we can better approach management plans. It is important that all information is shared with the families and they are guided appropriately. In addition to understanding, parents might also experience difficulties coping with the diagnosis and may not be receptive to the information given to them about diagnosis and hearing aids (Munoz et al., 2014). The CAEPs provided reassurance to parents that the hearing aids would enable speech sounds to be detected and this in turn means that parents will persevere with them. Prior to the use of CAEPs and the use of

speech tokens, parents had not always been able to fully comprehend the demonstration or the implications of their child's hearing of speech. CAEPs were used to demonstrate access to sound to parents.

Parents also reported finding it useful to have Parent Support Workers involved because they offered emotional support and introduced families to a local parent-to-parent support programme, which had evolved to meet their needs. Mehta et al. (2018) conducted a study on 35 families who received parent-to-parent support and reported that 63% felt that a parent who had a shared experience was the person best placed to offer help and advice immediately following the diagnosis. An overwhelming 97% would recommend peer support as being useful. Furthermore, 12 professionals, replied saying that PSWs are a beneficial addition to the existing programme and none wanted the service to be withdrawn. The research found that the use of PSWs enabled better links with the home environment and meant greater shared experience in caring for a deaf child.

Luterman et al. (1999) conducted a retrospective five-question survey of 75 parents whose children had hearing loss. The questionnaire asked for parents' perspectives to identify key elements of a comprehensive management programme. These parents indicated that their predominant need during the identification process was contact with other parents. Both these results have shown that parents find it more comfortable asking another parent questions about their child's hearing loss and hearing loss and future. The Parent Support Worker is a good way of providing this parental provision in a peer support model. This was beneficial because of the emotional support that was provided, engaging in self-directed learning, parent-to-parent networking and assistance in developing a better understanding of the tests involved in the infant audiology pathway. In other districts the main mode of support for families are the TOD's. The provision of peer support has been valued as an additional support by the parents in this area, especially as it entailed the discussion in the home environment which followed the HEARLab unaided and aided appointment.

The questionnaire aimed to evaluate the use of CAEPs in a clinical setting and in countries where CAEPs are less commonly used. The aim was to find out if audiologists found the use of CAEPs beneficial, particularly with children with

milder and moderate losses because of the uncertainty associated with understanding if aiding would be beneficial. Overall, the results from the questionnaire showed that with the addition of CAEPs to the test battery, clinicians felt more confident about patient management and were able to advise families according to the results. Results from this experimental study showed that the use of CAEPs helped clinicians confirm the child's hearing loss and reduced the ambiguity regarding appropriate clinical interventions for children with mild and moderate hearing loss. In line with the findings from other experimental chapters in this thesis, clinicians reported that using CAEPs is important for helping with families' education on hearing loss and hearing aids.

Currently hearing aid fitting is based on predicted thresholds from the ABR measurements which in turn leads to hearing aid targets being implemented based on these results. Fitting hearing aids to prescriptive targets in infancy can result in aids not being correctly amplified (McCreery et al., 2013). The current research found that clinicians found it useful to validate the hearing aid fitting. The CAEPs allow a way to ensure that the aids are providing sufficient amplification for speech sounds, which was particularly important during the period of time following diagnosis and when behavioural assessments can be conducted at 6-8 months. Currently in the UK, once the child has been aided there is no formal validation of hearing aids prior to behavioural assessment. Parents have to wait until their child is developmentally ready for the next hearing test to find out how much of an impact the hearing aid is having, and this can often result in months of waiting.

Overall, the responses from the questionnaire indicated that CAEPs were beneficial with children where behavioural assessment was not possible and provided another testing method for audiologists during the early critical months in the infant audiology pathway. Children with complex needs tend to have a later diagnosis of hearing loss than those who do not (McCracken & Pettitt 2011). These delays and the difficulties obtaining reliable hearing thresholds from this population means that appropriate aiding can be late and that crucial time is lost in the early months of language acquisition.

The findings indicated that CAEPs are useful at different points in the infant audiology pathway. Aided CAEPs have the potential to supplement existing clinical procedures especially before reliable aided behavioural assessment is possible. Research by Van Dun et al. (2016) demonstrated that CAEPs can potentially be used to assess hearing aid gain in hearing-impaired users. It has demonstrated that currently there is a gap in the infant audiology pathway and CAEPs close this gap from diagnosis to validation. The results indicated that approximately 50% of clinicians use the CAEPs for children <5 years of age and have used the equipment for counselling and fine-tuning hearing aids. CAEPs can be effectively used at many stages in the infant audiology pathway providing valuable information. CAEPs can supplement existing practice for children around 4-6 month olds.

## **5.2 Thesis limitations**

One of the limitations of the HEARLab is that the parameters of the equipment are rigid and thus, testing at other levels or with different stimuli was not possible. The stimulus choice for the HEARLab was limited to /m/, /g/ and /t/ and thus, we were unable to compare responses to other speech tokens. The clinicians highlighted limitations of the HEARLab equipment. The test itself is limited by the speech stimuli that can be used. Since this research was conducted, NAL launched a new module that incorporated the /j/ speech token (Van Dun. 2017). In addition, HEARLab® ACA™ cannot be used for threshold estimation, because presentation levels are fixed and stimuli are presented in the free field. Furthermore, the HEARLab does not provide testing in free-field to levels lower than 55dBA which is a limitation when trying to test mild losses. It is evident from the current research that milder losses require monitoring and in some cases aiding. However, in spite of these limitations, use of CAEPs have been positively received by clinicians. This research established that the use of this equipment, or equivalent equipment should be explored further in the clinical setting. A positive suggestion arising from this research is that clinicians would appreciate peer-to-peer working and training opportunities on the use of CAEPs. This is particularly the case in countries outside of Australia where support for the product is not available.

### 5.3 Future directions

A larger multi-centre study to explore outcomes and predictors should take place with larger numbers. To further explore the contribution of the inclusion of CAEP to improved language outcomes, a multiple regression model could be used to take account of all the factors that are known to affect language outcomes. The inclusion of CAEP in the infant audiology pathway needs to be one of the variables included in the research model. This is to measure what impact CAEPs have on speech and language outcomes. Future studies should aim to explore both patient/caregiver and practitioner perspectives on service provision, as such data can help inform evidence-based practices when providing services to cultural minorities.

Furthermore, it would be helpful to employ the same speech tokens used in the CAEPs to determine minimum responses' levels when the child is able to undertake behavioural assessment, using visual reinforcement audiometry so that responses can be compared. Also, future studies should include greater numbers of mild hearing loss cases so that better comparisons can be made with this level of hearing loss. It would also be beneficial to have had some language outcome measurements in this research so that speech and language development could have been assessed. Future studies should consider additional components of language, including comprehension, syntax and pragmatics. Also, further research is required to see if the HEARLab can be used to fine-tune hearing aids, how accurate it is at optimising hearing aid fittings and does this facilitate better outcome measures for these children?

This current research has shown that children with milder levels of hearing loss are performing as poorly as those with moderate to severe hearing losses. In many developed countries the NHSP have begun to address mild hearing losses by identifying them in infancy. However, in the United Kingdom (UK), this programme is not designed to detect mild levels of hearing loss (Bamford, Uus, & Davis, 2005), meaning that many children born with mild congenital SNHL will not be detected until later in childhood (Watkin & Baldwin, 2011). This raises the question of whether the NHSP should be also targeting those children that have milder losses. Should this be something that the UK should introduce into the

assessment pathway allowing for the challenges of separating high numbers of conductive from sensorineural cases? This is something that needs to be investigated further and future research would benefit from a greater number of cases of mild hearing loss.

#### **5.4 Conclusion**

This research has shown clinical evidence that CAEP testing allows hearing aids to be optimally fitted earlier and infants to be referred to cochlear implantation sooner. Just like Australia, the use of CAEPs should be introduced into the UK to all infant audiological pathways. Furthermore, the current research has shown that parents look to their child's primary care providers for advice. Professionals now have the ability to motivate families with the capability to show aided and unaided responses and to enrol them into the intervention programme in a timely manner. It would be interesting to see if the early aiding and referral for cochlear implants has had any cost implications on the NHS long term, for example, has engaging families reduced the number of follow-ups required? The use of CAEPs has aided management decisions and allowed for honest, informative discussions with families. For families themselves, increased awareness of their child's condition coupled with good quality, evidence based intervention can help their child towards reaching their full potential. This has been an important study as it has demonstrated the progress of children diagnosed with a hearing loss and shown better ways to improve process in the UK.

## CHAPTER 6: References

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## **Appendix 1**

Please see below the list of questions that were asked on the questionnaire.  
(The questions which are underlined are the questions which were added to the Punch et al. 2016 questionnaire)

Demographic questions

### **Clinician's experience with the use of HEARLab:**

How long have you been using the HEARLab system for?

Approximately how many assessments have you made using the HEARLab system?

### **Management Pathway:**

"The results of HEARLab influence my approach to rehabilitation."

"The results of HEARLab 'help me' adjust the hearing aid setting."

The results of HEARLab are clinically consistent with behavioural results.

The results of HEARLab are easy to interpret.

I feel confident with the results I get from HEARLab.

What are the relevant factors when making a decision to book a client for HEARLab assessment?

At what point along the clinical pathway have you used HEARLab for?

What client groups/patients have you assessed using the HEARLab system?

### **Clinicians' views:**

The results of HEARLab are positively received by parents.

On average how many minutes does it take you to complete a test run of 3 speech sounds at one intensity?

Which stimulus or stimuli from /m/, /g/ and /t/ do you find is consistent with behavioural results?

The calibration function for free field testing is efficient.

How often do the following issues prevent you from completing HEARLab assessment?

The results of HEARLab do not change my approach to rehabilitation.

“I would recommend HEARLab to other Audiologists who do not have the HEARLab system.”

“The HEARLab is important to my clinical practice.”

The information HEARLab provides in achieving optimal habilitation outcomes for children has been useful in clinical practice.

Please list the things you like LEAST about the HEARLab Aided Cortical Assessment (ACA) Module.

Please list the things you like MOST about the HEARLab Aided Cortical Assessment (ACA) Module.