Impact of conductive unilateral hearing loss in children- A qualitative investigation.

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Abbreviations

BAHA – Bone Anchored Hearing Aid

BHL - Bilateral hearing loss

CBCL - Child Behaviour Check List

CELF - Clinical Evaluation of Language Fundamentals

CHILD - Children's Home Inventory for Listening Difficulties

CHL - Conductive hearing loss

CMMS - Columbia Mental Maturity Scale

dB HL - decibels in hearing level

EAS - effort assessment scale

FAS - fatigue assessment scale

fMRI - Functional Magnetic Resonance Imaging

HEAR-QL - The Hearing Environments and Reflection on Quality of Life

HINT-C - hearing in noise test - children

HRA - Health Research Authority

HRQOL - health related quality of life

IQ - Intelligence Quotient

kHz - Kilohertz

MAIS - Meaningful Auditory Integration Scale

MRI - Magnetic Resonance Imaging

MUSS - Meaningful Use of Speech Scale

NH - Normal Hearing

NHSP - Newborn Hearing Screening Program

NICU - Neonatal Intensive Care Unit

NREC - National Research and Ethics Committee

OMI - Outcome Measurement Instruments

OWLS - Oral and Written Language Scales

PCHI - Permanent Childhood Hearing Impairment

PEACH - Parents' Evaluation of Aural/Oral Performance of Children

PedsQL - Pediatric Quality of Life Inventory

PICOS - Population, Intervention, Control, Outcomes, and Study design

PRISMA - Preferred Reporting Items for Systematic Reviews and Meta-Analyses

PROSPERO - Prospective Register of Systematic Review

QOL - quality of life

RCT - Randomised Controlled Trial

REC - Research Ethics Committee

SNHL - Sensorineural Hearing Loss

SSQ - Speech, Spatial and Qualities of Hearing Scale

SSQ-C - Speech, Spatial and Qualities of Hearing Scale for children

UHL - Unilateral hearing loss

UK - United Kingdom

WASI - Wechsler Abbreviated Scale of Intelligence

WHO - World Health Organization

WIAT-II-A - Wechsler Individual Achievement Test

WISC-IV - Wechsler Intelligence Scale for Child-Fourth edition

WISC - Wechsler Intelligence Scale for Children

Impact of conductive unilateral hearing loss in children-A qualitative investigation.

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Abstract

Unilateral hearing loss (UHL) has been defined based on pure tone averages, and refers to average air conduction thresholds (0.5, 1 and 2 kHz) worse than or equal to 20 dB HL in the impaired ear. The developmental, social and communication risks from having a unilateral hearing loss in a child have often been discounted or down-played, based on the belief that a contralateral normally hearing ear would provide sufficient access to sound. The aim of our study was to identify what is known about the impact of UHL through a systematic review and through a qualitative study to understand the perspectives of children on the impact of their conductive UHL and their parents' perspective of the conductive UHL.

The systematic review aimed to evaluate the literature to identify the impact of unilateral hearing loss on childhood development. 41 studies investigating the impact of unilateral hearing loss in children were included in this review. We identified seven main domains that have been studied in relation to the impact of UHL in children. The only outcome domain, where there was a consistent negative impact of UHL was hearing difficulties in real-life, such as hearing-in-noise and the ability to localise sound.

Our qualitative study explored the perceptions of the impact of a unilateral conductive hearing loss in children aged 11-17 years and their parents. The aim of the study focusing on the children's qualitative interviews was to understand the perspectives of children regarding the impact of unilateral conductive hearing loss on their lives. Our results indicate children with unilateral conductive hearing loss are keen to normalize their hearing loss and adapt to the resulting difficulties. There is a need to identify hidden difficulties, which become apparent on exploring their daily routine, to ensure appropriate treatment strategies can be advocated. This study highlights patient-specific outcome domains that are most relevant to children with unilateral conductive hearing loss

Our qualitative study also aimed to understand parental perception of the impact of unilateral conductive hearing loss in their children. Our study revealed three interlinked categories: (I) problems perceived by parents and acceptance, (ii) advice, monitoring and support to overcome these problems, and (iii) implications of active issues and parental concerns. Parental acceptance of relevant problems, and their concerns need to be considered when evaluating children with conductive unilateral hearing losses.

Our study highlights that the potential impact of UHL in childhood is not inconsequential. We identify the following areas of unmet research need, which either hinder evaluation of the problem or limit options for clinical management:

- Greater understanding of the impact of conductive UHL in children.
- Need for bespoke outcome measurement instruments (OMI) to quantify the true extent of problems in children with conductive UHL.
- Identification of interventions effective and acceptable in this patient group.

Declaration

No portion of the work referred to in the thesis has been submitted in support of an application for another degree or qualification of this or any other university or other institute of learning.

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Dedication

I would like to dedicate this thesis to my husband Raj and our children Neel and Taran. Without their patience, support, love and sacrifice, this research would not have been possible.

I would also like to dedicate this thesis to my father and father-in-law. My father has always encouraged me to believe in myself and do my best. My father-in-law enjoyed our conversations about this research, and during this research sadly passed away. You are missed.

Chapter 1: Introduction

Introduction

The World Health Organization[1, 2] estimates that 32 million children worldwide are affected by disabling hearing loss, which is defined' as a loss of more than 30 dB in the better hearing ear. The WHO classification system grades the severity of hearing loss based upon the hearing thresholds in the better hearing ear[3], *using a* 4-frquency pure tone average *(0.5, 1, 2 and 4 kHz). Normal hearing* is described as an average hearing threshold ≤ 25 dB HL, *mild hearing* loss from 26-40 dB HL, *moderate hearing loss* from 41-60 dB HL, *severe hearing loss* from 61-80 dB HL and *profound hearing* loss >80 dB HL. Although, these pure tone averages are useful to describe the degree of hearing loss, they do not fully characterize the nature of hearing deficit and the listening difficulties encountered by the affected individual, with the consequences of similar degrees of hearing loss varying between individuals.

Prevalence rates for hearing loss in children vary from 5 per 1000[4] to 1.7% depending on the age group. Congenital hearing loss of moderate or worse severity has a prevalence of 1.1 per 1000 live births[5]. Management strategies for children with hearing loss depend on the severity of the hearing loss, age at diagnosis, symmetry of hearing thresholds between ears (asymmetrical or symmetrical), and the nature (conductive or sensorineural) of the hearing loss. Early identification of children with bilateral permanent childhood hearing impairment (PCHI) of moderate or worse severity is particularly important in preventing the negative impact on childhood development, with the potential benefits of early intervention in preventing a delay in speech & language development a significant consideration. Early intervention in children with hearing loss can result in age-appropriate speech & language outcomes[6]. *Unilateral hearing loss* (UHL) has been variably defined based on pure tone averages. One of the earliest definitions of UHL by Bess et al[7] refers to average air conduction thresholds (0.5, 1 and 2 *kH*z) worse than or equal to 20 dB HL in the impaired ear and better than or equal to 15 dB *HL* in the better hearing ear. This loss may be congenital or acquired, sensorineural or conductive and of varying severity. Permanent hearing loss affects at least 1.65 live births per 1000 in the UK[8]. Following the introduction of the Newborn Hearing Screening Program (NHSP) (2006 in UK), the incidence of neonatal UHL has been reported to be 0.81/1000 [9]. The aim of the NHSP has been to detect bilateral permanent hearing impairment early, in the understanding that

intervention in the first 6 months of life[10] can improve outcomes in children with moderate or greater bilateral permanent hearing loss. Identification of permanent moderate hearing loss became possible within 4-5 weeks of birth, with the screening protocol set to refer babies who fail screening in one ear or both. This resulted in significant numbers of babies with UHL being identified at birth. The guidelines for early audiological assessment and management of babies referred from the NHSP recommended "monitoring and review of babies diagnosed with UHL, as evidence did not show benefits of active early intervention"[11]. Congenital UHL accounts for 42% to 47% of all children with UHL[12, 13]. In addition, children may acquire progressive UHL due to trauma, infections and other aetiologies, with the prevalence of UHL in children aged 6-19 years being reported to range from 3% to 6.3%[14].

The developmental, social and communication risks from having a UHL have traditionally been discounted or down-played, based on the belief that normal hearing in the contralateral ear would provide sufficient access to sound to develop unhindered[15]. The majority of studies and reviews have focused on investigating the impact of UHL on domains such as speech & language[16, 17], cognition[18] and educational consequences[19], with the choice of these outcomes being influenced by clinician opinion regarding the likely consequences and the nature of available outcome measurement instruments (OMI). There is a need to identify bespoke patient-specific outcome domains to help evaluate the impact of UHL across the stages of childhood, both within and between affected individuals, to enable greater understanding of the impact upon development and facilitate meaningful evaluation of the effectiveness of novel and existing therapeutic interventions.

One of the main factors influencing the measurement of impact from childhood hearing loss is the changing developmental ability in children. As a result, the most clinically relevant outcome domain for a two-year-old is likely to be different from that for a 12-year-old. When assessing the impact of UHL in children, consideration must be given to several key factors: 1. age at assessment, 2. severity of hearing loss, 3. type of hearing loss (sensorineural, conductive or mixed), and 4. comorbid factors (e.g. dual sensory or cognitive impairment).

Methodology

To capture any impacts experienced by children with a conductive UHL, and their parents, we chose to conduct a *qualitative descriptive exploratory study*. The methodology and rationale for

the qualitative study is described here, as the individual papers cannot contain this depth of detail due to journal restrictions.

There are two main interpretive approaches that may be adapted to study this area, namely *Phenomenology* and *Grounded theory*. Phenomenology is a qualitative method with its origin in European philosophy. Welman and Kruger[20] state that, "phenomenologists are concerned with understanding social and psychological phenomena from the perspective of people involved". This philosophical approach has evolved into a research methodology for qualitative research, but through my further reading I felt this approach did not suit my research question. Phenomenology helps understand the lived experience of a condition. A very important aspect of our research question was to understand, which aspects of any impact of UHL is important to children and their parents. Grounded theory with its sociology roots, allows us to do this by developing a theory inductively from the data that describes what is important to the studied individuals. Different philosophical perspectives have guided the conduct of grounded theory research. Early proponents of grounded theory Strauss and Corbin[21] advocated symbolic interactionism as a theoretical basis of grounded theory. Certain processes involved in generation of grounded theory, such as theoretical sampling, data collection & analysis and the use of memos support this theoretical basis. Kathy Charmaz's [22] *constructivist* approach, recognises the value of the interaction between the researcher and the participant. It values the researchers personal bias as it adds meaning to the theory generated. In my research, I have utilised certain aspects of each of these philosophical perspectives.

The aim of our study was to explore the impact of UHL perceived by children and parents on their day-to-day activities. We chose to specifically explore children with a conductive hearing loss, in order to gain an in-depth understanding of this subgroup of UHL. As we were keen to explore the perceptions of a specific group, we chose *theoretical sampling*. Theoretical sampling has been described as "the process of data collection for generating theory whereby the analyst jointly collects, codes and analyses [the] data and decides what data to collect next and where to find them in order to develop his theory as it emerges"[23].

When deciding on a sample size, it has to be appreciated that sampling in qualitative studies is unlike that in quantitative studies, wherein sample size is determined using statistical methods. Determining sample size in a qualitative study depends on multiple factors. The quality of data

gathered[24] is one of the important factors. This depends on the experience of the participant, how articulate, and how forthcoming they are to share their experience. This information is often not available at the time of study planning and estimating the sample size. The factors that may help estimate sample size are: 1. scope of the study, 2. nature of the topic, and 3. study design. In our study we planned to interview the parent and the child, and interview older children (11-17 years). This was helpful as we were able to gather information from adolescence and childhood developmental stages. Although not a rare phenomenon, there is significant controversy regarding the existence of negative consequences and the need for intervention for children and young people with unilateral UHL. We estimated a sample size of 15 children and their parents would yield data to generate the necessary information. Data collection was designed to be in the form of semi-structured interviews and specifically chosen questionnaires. There has been an on-going debate regarding the relative usefulness of qualitative and quantitative studies. Some authors have chosen an intermediate approach, combining the two modalities[25, 26]. The aim of our study was predominantly to understand the impact of conductive UHL in children. We used the questionnaires to get additional information on certain aspects of daily living and to identify the ability of these questionnaires to reflect participant experiences. For the qualitative data collection, we chose to conduct semi-structured interviews. There is debate in the literature over the advantage of unstructured interviews over semi-structured interviews. Corbin and Strauss[21] advocate the use of unstructured interviews as this allows the participant to talk about topics important to them. Glaser in his article on the use of interview guides says, “If the data is garnered through an interview guide, that forces and feeds interviewee responses then it is constructed to a degree by interviewer imposed interactive bias”. However, there are proponents of topic guides and semi- structured interviews. Open questions in the topic guide can focus the interview[22, 27] and allow all areas of interest to be explored. In addition, subjects still have the opportunity to add information important and relevant to them. The topic guide can be reactive to new information being gathered. Another feature of grounded theory methodology we adopted in our study was *constant comparative analysis*. As data collection progressed the data was analysed simultaneously and compared, in order to identify developing categories. Data analysis was conducted by breaking down the data and analyses. Data analysis occurred in three stages: 1. in-vivo coding, where we coded

sentiments described by parents and children using their own words, 2. secondary codes were formed from amalgamation of similar codes, and 3. meaningful categories emerged as we linked the secondary codes together.

At the end of parent and child interviews, the child completed three questionnaires, - the Speech, Spatial and Qualities of Hearing Scale (SSQ) for children, the effort assessment scale (EAS) and the fatigue assessment scale (FAS). This was done, so any influence of the questionnaire on the interview could be avoided. (Appendix-SSQ, EAS and FAS.). All three measures are designed to identify difficulties in the presence of suboptimal listening conditions[29]. The SSQ hearing scale for children questionnaire has been modified from the adult version with age appropriate language[30]. The effort assessment scale (EAS)[31] and the fatigue assessment scale (FAS) [28] have been validated in adult subjects. The EAS assesses the levels of listening effort experienced in different daily life situations in a noisy environment. The total score for the EAS is calculated by adding the score of each of the six questions to give a score between 0 and 60, with higher scores indicating more effort. The FAS is used to measure fatigue in an individual directly attributable to their hearing loss. The overall score for FAS is calculated by summing the responses obtained to each individual question. The total score of FAS ranges from 0 to 40, with higher scores indicating more fatigue.

The aim of this thesis was to evaluate the impact of conductive UHL in children, utilizing a systematic review of the literature and qualitative research methodology to gain understanding of what is important to children and their parents. We will present our findings in the form of three papers, which will form three chapters (Chapters Two, Three and Four) of the thesis.

Paper One-This chapter in the thesis provides a background from the literature. A Pubmed database search upto April 2018 was carried out to identify and review studies reporting on the impact of UHL in children. The reported outcomes are described under each domain affecting the child.

Objective: A systematic review to fully charecterise the impact of UHL in children by amalgamating understanding from the available literature and identifying gaps in knowledge.

Paper Two. The perspectives of children on the impact of conductive UHL on their lives: A qualitative study

We conducted a qualitative study of children with conductive UHL to understand their

experience and perspective of living with their hearing loss. This qualitative study explored the experience of children through semi-structured interviews using grounded theory methodology.

Objective**:** The aim of this study was to understand the perspectives of children regarding the impact of conductive UHL on their lives.

Paper Three. Parental perception of the impact of conductive UHL in their children – A qualitative study

This paper describes the parental perception of their child's conductive UHL on daily living and explores how parents see the impact. We compared parental and child perspectives to identify differences, and determine if parental report by proxy sufficiently captures the perspectives of children.

Objective: To understand parental perception of the impact of conductive UHL in their children.

Thesis format

The thesis will be presented in the "alternative format" used by the University of Manchester, as the research has resulted in findings that can be published. This format also demonstrates the candidates training, in skills required for dissemination of research outcomes to readers of academic journals. Pagination in these papers will follow the pagination in the thesis, as although the papers have been written for the journals, they have not yet been submitted. References in papers will be provided as a list at the end of each paper and references in the rest of the thesis will be provided in the thesis. Each paper will be listed as a chapter.

The candidate suggested the main aim of the study and supervisors Peter Callery and Kevin Munro advised on the design, data collection and analysis, interpretation of the study and revision of the manuscripts. The candidate and a research audiologist Helen Whiston conducted data collection in this study. As the candidate is a clinician involved in the clinical care of many of the children participating in this study, Helen Whiston conducted seven of the eleven interviews. For the systematic review the candidate and co-author Aleksandra Metryka carried out the literature search and extracted and synthesized the data.

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Chapter 2:Systematic review of the consequences of Unilateral Hearing loss in children

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Abstract

BACKGROUND: The developmental, social and communication risks from having a unilateral hearing loss have often been discounted or down-played, based on the belief that a contralateral normally hearing ear would provide sufficient access to sound to develop unhindered. In recent years, this traditional perspective has been challenged as evidence grows that whilst some children with unilateral hearing loss seem to develop without significant problems, other children encounter problems directly attributable to the presence of a hearing loss.

OBJECTIVE: This systematic review aimed to determine the impact of unilateral hearing loss on childhood development.

DESIGN: This review has been conducted in accordance with the statement on Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) and registered with the Prospective Register of Systematic Review (PROSPERO). Literature review to identify the impact of hearing loss in one year in children aged less than 18 years. We included peer reviewed experimental studies, cohort observational studies, longitudinal and cross section studies and qualitative studies.

RESULTS: 41 studies investigating the impact of unilateral hearing loss in children were included in this review. We identified seven main domains that have been studied in relation to the impact of UHL in children. These domains were: hearing difficulties in real-life situations, impact on education, effect on cognition, Speech and Language development, impact on Balance, central neurological changes, and psychosocial impact. The only outcome domain where there was a consistent negative impact of UHL was hearing difficulties in real-life, such as hearing-in-noise and the ability to localise sound.

CONCLUSION: The impact of UHL in children remains contentious, although the evidence for a potential negative impact upon various aspects of childhood development is increasing. As such, the traditional belief that the impact of UHL on childhood development is negligible, or insignificant, is no longer generalizable, necessitating an individualized approach based on a clear understanding of the potential negative consequences.

Keywords-Unilateral hearing loss, children, childhood development

1. Introduction

The World Health Organization estimates that 32 million children worldwide are affected by disabling hearing loss, defined as a loss of more than 30 dB in the better hearing ear[1,2]. The WHO classification system grades severity of hearing loss based on hearing thresholds in the better hearing ear[3], using a 4-frquency pure tone average (0.5, 1, 2 and 4 kHz). Normal hearing is described as an average hearing threshold ≤25 dB HL, mild hearing loss from 26-40 dB HL, moderate hearing loss from 41-60 dB HL, severe hearing loss from 61-80 dB HL and profound hearing loss >80 dB HL. Although these pure tone averages are useful to describe the degree of hearing loss, they do not fully characterize the nature of hearing deficit and the listening difficulties encountered by the affected individual, with the consequences of similar degrees of hearing loss varying between individuals.

Unilateral hearing loss (UHL) has been variably defined based on pure tone averages. One of the earliest definitions of UHL by Bess et al[4] refers to average air conduction thresholds (0.5, 1 and 2 kHz) ≥20 dB HL in the impaired ear and ≤15 dB HL in the better hearing ear. This loss may be congenital or acquired, sensorineural or conductive and of varying severity. Permanent hearing loss affects at least 1.65 live births per 1000 in the UK[5]. Following the introduction of the newborn hearing screening program (NHSP, 2006 in UK), the incidence of neonatal UHL has been reported to be 0.81/1000 [6]. The aim of the NHSP was to detect bilateral permanent hearing impairment early, in the understanding that intervention in the first 6 months of life[7] can improve outcomes in children with moderate or greater bilateral permanent hearing loss. Identification of permanent moderate hearing loss became possible within 4-5 weeks of birth. The screen protocol was set to refer babies who fail screening in one ear or both. This resulted in significant numbers of babies with UHL being identified at birth. The guidelines for early audiological assessment and management of babies referred from the NHSP, recommend monitoring and review of babies diagnosed with UHL as evidence did not show benefits of active early intervention[8]. Congenital UHL accounts for 42% to 47% of all children with UHL[9,10]. In addition, children may acquire progressive unilateral hearing loss due to trauma, infections and other aetiologies, with the prevalence of UHL in children aged 6-19 years being reported to range from 3% to 6.3%[11].

UHL was considered to cause minimum impairment[12]. Many studies and reviews have focused

on investigating the impact of UHL on domains such as speech and language[13,14], cognition[15] and educational consequences[16], with these reported outcomes reflecting clinician opinion of likely consequences and being influenced by available outcome measurement instruments. Most studies chose multiple outcomes and did not identify a primary outcome measure or domain. There is a need to identify bespoke patient-specific outcome domains to help evaluate the impact of UHL across the stages of childhood, both within and between affected individuals. This will enable greater understanding of the impact upon development and facilitate meaningful evaluation of the effectiveness of novel and existing therapeutic interventions.

When assessing the impact of UHL in children, consideration must be given to several key factors: age at assessment, severity of hearing loss, type of hearing loss (sensorineural, conductive or mixed), and co-morbid factors (e.g. dual sensory or cognitive impairment).

2. Aim

This systematic review was designed to identify and assimilate what is known about the impact of UHL in children (under 18 years of age).

3. Methods

The study protocol for this review has been registered with PROSPERO, the international Prospective Register of Systematic Reviews, with the protocol id CRD42019126330.

The review aimed to evaluate available literature and describe the impact of UHL in children on specific domains used in the literature and the global impact**.** A domain has been defined as a concept to be measured, a further specification of an aspect of health, categorized within a Core Area[18]. We took account of these selection considerations in defining the inclusion and exclusion strategy according to the Population, Intervention, Control, Outcomes, and Study design (PICOS) criteria for systematic reviews. The review has been conducted in accordance with the Preferred Reporting Items for Systematic Review and Meta-Analysis (PRISMA) statement. (http://www.prisma-statement.org/)

Search Strategy

We searched using terms “unilateral”, “hearing loss” and combined the output of these two searches. Different electronic databases were searched systematically (Pubmed, Medline, CINHAL. PsychINFO, EMBASE, BNI) and results combined. As multiple databases were selected duplicates required removal. The Pubmed database search identified the same

studies, however, this database does not have a functionality to apply limits (e.g. children, child or childhood)(Table 1). Hence titles and abstracts of identified studies were searched to include articles pertaining to children. The search was conducted on the identified databases in April 2018.

Inclusion and Exclusion Criteria

The PICOS criteria for this review are described in Table 2**.** Titles and abstracts of articles were screened to identify those relevant to the objective of our review. Articles referring to animal experiments, pertaining to adult populations, and review articles were excluded. Literature not available in English language was excluded. Conference abstracts were also excluded, as we did not have access to their full investigation. Papers reporting on mixed population of children and adults were included to investigate their findings in children under the age of 18 years. Many studies have included children with UHL and mild bilateral hearing loss together to understand the impact of minimal bilateral and unilateral hearing loss (MBUHS). We only included studies that described results in children with UHL as a separate entity. We found most studies have not described the type of hearing loss (congenital/acquired, conductive/sensorineural, mild/moderate/severe). However, as the findings could be relevant to our aim we included these studies, providing they specifically described the patient population as being children with UHL.

Data extraction

Data from each study was extracted separately by 2 authors (JN and AM), specifically the authors, year of publication, study design, patient characteristics (age, type and severity of hearing loss), number of participants, control groups, outcome domains and corresponding measurement instruments, and findings of the study.

Quality of evidence

The quality of selected articles was assessed using Downs and Black checklist[17]. This checklist has been designed to assess the quality of studies included in systematic reviews. The five sections have been designed to assess the overall quality of the study, external validity, study bias, confounding and selection bias and power of the study. Two authors JN and AM independently assessed each article against the items in the checklist. In the presence of differences in ratings by the two authors, discussions followed consensus ratings are reported.

4.Results

The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flow diagram (Figure 1) illustrates the flow, selection and exclusion of the studies included in this review, based on PICOS criteria. A total of 41 articles were identified through the database and manual search (Figure1).

Quality of evidence

The result of this methodological assessment is presented in Table 3. Our locally determined threshold of quality was set at 75%. The scores for individual studies ranged from 11% to 66%, with none of the studies (0/41) reaching our pre-determined threshold of high quality; due mainly to poor scores for internal validity, external validity and study power.

Data extraction

As we included a significant number of articles in this review, we have chosen not to include a single summary table, in an attempt to increase readability and ease of interpretation. Rather we have grouped articles based on the predominant domain reported on and in the order they appear in this review. Data analysis was carried out and extracted in respect to domains studied. . Through this process we identified seven main domains that have been studied in relation to the impact of UHL in children

These domains were:

- Hearing difficulties in real-life situations
- Impact on education
- Affect on cognition
- Speech and Language development
- Impact on Balance
- Central neurological changes
- Psychosocial impact

Many studies reported on more than one domain, often combining cognition and education, or hearing difficulties in real-life situation and education. None of the studies available in the literature report on the global impact of UHL in children. The level of evidence will be identified as per the description in table 4 [19]

Hearing difficulties in real-life situations

We classified the ability to localise sounds, hear in the presence of noise and understand speech-in-noise as hearing difficulties in real life situations. The impact of UHL on hearing difficulties in real life situations was reported in ten studies. Level of evidence in this domain was Level III (case control studies), except one study, which was level IV (observational study).

Nine studies measured hearing performance in quiet and noise, with the outcome measurement instruments used comprising speech recognition in quiet and noise, HINT-C, word recognition scores, nonsense syllable test, and the PEACH questionnaire. Localisation ability was measured using multiple speakers and sound arc, SSQ questionnaire and the H70 questionnaire. Listening difficulties were evaluated in younger children using the CHILD questionnaire. Two studies included children aged less than 3 years of age and 8 studies reported on older children aged 6 years and above. Two studies[20,21] reported poor hearing performance in noise in children aged 1-3 years. In children over the age of 6 years three studies [22-24] reported difficulties in localisation of sound. Seven studies [22,23,25-29] reported that children with UHL had significantly greater difficulty in understanding speech-in-noise, particularly when the noise was presented to the better hearing ear. Table 5 shows the summary of studies examining real-life hearing. There is uniform Level III evidence that older children aged 6 to 17 years with UHL have difficulty localising sounds and hearing in noise.

Education

Eleven studies were identified that investigated the impact of UHL on education. The level of evidence was grade III in 4 studies (case-control) and grade IV (cohort observational studies, surveys, and retrospective review) in the rest. The outcome measures evaluated included: educational performance measured using school achievement tests and standardised achievement tests (6 studies), educational progress measure by recording the need for additional resources, grade repetition and parent-teacher reports (5 studies). Two studies[30,31] reported no evidence of persistent negative impact of UHL on education. Difficulties related to academic achievement in children with UHL were reported in 3 studies[32-34]. The need for additional educational resources in children with UHL was identified in five studies[34-38]. Another identified consequence of UHL in children was the need to repeat a school grade[33,34,37-40], as

reported by six studies. Children with sensorineural hearing loss were more likely to repeat a grade than children with conductive hearing loss[34]. Table 6 summarises the characteristics of studies investigating impact of UHL on education. Recognising the potential for educational difficulties and regular monitoring are necessary if children encountering difficulties at school are to be identified expediently and interventions introduced.

Cognition

Twelve studies investigated the impact of UHL on cognition in children. The level of evidence was grade III in eight studies and grade IV in the rest. We identified one meta-analysis, which reported on combined results of 4 of the 8 case control studies.

Intelligence Quotient (IQ) in children has been the most commonly used instrument to measure cognition. Subsets of the IQ test which measure verbal reasoning (use words for problem solving) and non-verbal reasoning (use visual information for problem solving) have been used to ascertain if differences in cognitive ability may be due to the deleterious effect of UHL on a child's ability to access words (impact on verbal reasoning). Word processing and learning aptitude tests were also used to assess impact on cognition.

Five studies[33,39,41-43] did not report evidence of significant difference in cognitive ability when compared with NH children or with standardised scores. Five studies[27,44,45 38,46] and one meta-analysis[47] combining 4 of these studies report a negative impact of UHL on cognitive ability. Two studies investigated the impact on cognition of the laterality of UHL, one of which[46] describes no significant difference in IQ scores in children with right or left sided UHL, as compared to another[48] which reported lower verbal IQ scores in children with right sided UHL and lower non-verbal IQ scores in children with left sided UHL. The impact of laterality of hearing loss and its significance is described in the section reporting on Central Neurological sequalae. One study[36] reported improvement in Verbal IQ scores and Full scores over time and it was suggested that improvement in the scores could be an indicator for improvement in language scores.

Table 7 summarises the studies investigating the impact of UHL on cognition. Although, evidence exists that some children with UHL have comparable cognitive ability to children with

NH, this equivalence is not universal and requires further investigation to identify factors influencing the achievement of intellectual and educational potential.

Speech & Language

We identified 12 studies that investigated the impact of UHL on speech & language development in children. Four of these studies were Level IV studies as they were observational studies or retrospective reviews and eight were Level III (case-control) studies. Most studies used a battery of speech & language development tests to assess production of speech, understandability of speech and language development.

Three studies[21,30,36] investigated the impact longitudinally. Fitzpatrick et al [21] did not find a difference in the language skills in children up to 4 years of age, whilst the other two [30,36] describe improvement in language scores in children with UHL over time. Five studies[41,43,27,35,38] identified a negative impact of UHL on speech & language development. No difference in speech & language outcome in children with UHL was reported in 4 studies[20,39,49,50].

Table 8 describes the studies investigating the impact of UHL on speech & language development. Although there appears to be some impact of UHL on speech & language development in children, it is not clear from the available literature why some children are more susceptible to delay or under-achievement.

Balance

The impact of UHL on balance skills in children has not been reported extensively. Although a large proportion of children with conductive UHL are likely to have normal inner ear anatomy, there is a high incidence of structural abnormalities of the labyrinth in children with congenital sensorineural hearing loss (SNHL).[51] The prevalence of these abnormalities has been reported to vary from 29% to 67% [52]. Structural abnormalities of the cochlea may be associated with abnormality of the balance organ due to the common embryonic origin. Additionally, both static and dynamic posture control depends on sensory-motor input from the internal and external environment and the ability to localise sound contributes to spatial awareness [53]. These factors may contribute to impaired balance skills in children with UHL.

Two studies investigated the impact of UHL on balance. One case control study[54] reported that compared to NH children, children with UHL performed worse on challenging tests that investigated static and dynamic balance. Another observational longitudinal study, commenting on motor coordination in children[30] described that a higher proportion of children with UHL were reported to be 'clumsy' by their teachers at seven years of age, improving by age 11.

Although the literature alludes to impairment of balance skills in children with UHL, the extent and significance of these difficulties is not clear and children with UHL, particularly those with congenital SNHL, may be prone to subtle difficulties in difficult balance situations where visual input is removed.

Central neurological changes

We identified research investigating the central neurological effects resulting from monoaural auditory input, including understanding of the impact of UHL on central auditory processing. Eight studies have reported on the central neurological changes secondary to UHL. Seven of these studies provide Level III evidence (case control studies) and one was Level IV (observational study). These descriptive studies elaborate on the central neurological changes resulting from the monaural input.

One study[55] used evoked magnetic fields to investigate these changes, but with evolving technology functional (3 studies) and resting state (4 studies) MRI has become the instrument of choice. One study[55] looking at cortical responses to auditory stimuli demonstrated differences in hemispheric response when exposed to auditory stimuli on the side with normal hearing and the side with UHL. Jung et al [56] studied resting state functional connectivity using MRI scans in children with UHL and their siblings with NH, and reported various changes in the central cortical networks with increased activity in certain areas of the brain and decreased or aberrant activity in others; areas known to have a role in cognition and decision-making. These adaptive/maladaptive changes have been postulated to account for some of the difficulties children with UHL encounter during their education.

Report of reduced deactivation of the resting state network[45] on functional MRI imaging has been hypothesised to aid in performing demanding cognitive tasks, and in children with UHL

this reduced deactivation may be contributing to the difficulty in performing demanding cognitive tasks.

Studies have reported specific effects on the primary auditory pathway, with reduced stimulation of this area in response to sound being suggestive of degeneration or demyelination of the auditory pathway due to monoaural auditory deprivation.[57,58] Changes in the secondary auditory area; auditory processing area and visual processing areas have also been reported in response to narrow band stimulus, speech-in-noise and in the resting state. [45,59]

Central neurological changes secondary to the laterality of hearing loss have also been investigated in an attempt to explain the right ear advantage theory. Several differences have been reported in the activation and deactivation of auditory and visual processing areas in children with right UHL, left UHL and normal hearing [45,58,60,61]. Attention areas were activated in children with normal hearing and left UHL, but not in right UHL. These findings suggest the secondary auditory association area is affected in UHL and attention networks are not activated in children with right UHL. This may explain some of the variability seen in the impact perceived by children with UHL. The relevance of these changes is not entirely clear and the evidence available is not conclusive. Review of available literature suggests differences in activation and deactivation of the central cortical network, but the relevance of this phenomenon needs further clarification. It is likely that some of these changes are consequent to the UHL and some may contribute to the difficulties experienced by children with UHL.

Psychosocial development

Unilateral hearing loss in children may impact on psychosocial development, as suggested in a few studies evaluating psychosocial development along with other consequences. A major difficulty in measuring the impact on psycho- social development is the lack of an appropriate measurement instrument. We identified three main domains in this area: impact on behaviour and social skills, quality of life, and accounts from teachers, parents and children themselves.

Impact on behaviour

Six studies[33, 34, 35,36,40,50] have reported on the impact of UHL on behaviour. All the studies provide Level IV evidence, with surveys, reports and behaviour rating scales being used. The

measurement instruments used included parent and teacher reports, child self report and the Child Behaviour Checklist (considered the gold standard for measuring behaviour issues).

All studies detailed behaviour issues as reported by parents and teachers, with reports suggesting that the left atresia group[40] (conductive UHL) had significantly higher prevalence of behaviour problems and the presence of lower social skills in children with atresia[50] compared to children with sensorineural UHL. However, when children reported using the self-rating behaviour scale[33] there was no significant difference between children with normal hearing and those with UHL in contrast to the teacher report which described behaviour problems in children with UHL.

Impact on quality of life

Studies to determine the quality of life (QoL) in children with hearing loss have tended to combine UHL and BHL, for comparison with NH children[62]. As such, it is difficult to establish from these studies the specific impact of UHL on QoL. Umansky et al[63] examined QoL in children with NH (n=35), UHL(n=35) and BHL(n=45) using the validated PedsQL questionnaire, but did not identify any significant differences in QOL in these 3 groups of participants.

Qualitative studies

Qualitative studies aimed at capturing the personal perspective of UHL have reported some interesting findings. A focus group of parents and children[62] with lived experience of UHL (4 mothers of children with UHL and 3 children with UHL) identified that children with UHL experienced difficulties related to psychosocial development, but despite experiencing these barriers they reported being “normal”. Unilateral hearing loss affected the way they interacted with friends and they described difficulties in social situations, e.g. when in a cafeteria with friends or when friends talked to them on the side of their UHL. The main concern from parents was regarding “difficulties in school and misunderstanding conversations”. In another qualitative study[64], academic and social experiences of children with UHL were studied through semi-structured interviews, with most participants reporting positive academic and social experience. Despite this positivity, these children reported facing difficulties listening in noisy conditions, in group discussions and when they did not have eye contact with their speakers, leading to

feelings of frustration, nervousness and embarrassment. The available evidence suggests that UHL can have a potentially negative impact on psychosocial development in childhood.

5. Discussion

This systematic review identified 41 studies investigating the impact of UHL on various outcome domains in children with UHL, utilising a variety of study designs: 29 case control studies, 3 cohort observational studies, 2 surveys, 3 longitudinal observational studies and one retrospective review.

The age group evaluated to study the impact of UHL ranged from infants to adolescents. Although seven studies included a mixed group of life-stages (adults and children) we did not exclude them in the belief that the findings were still relevant to at least the adolescent period. Most case control studies focused on children aged 6-16 years, as developmentally, children in this age group are believed most likely to exhibit a negative impact from hearing loss, should this be present. There were only two studies which investigated the impact of UHL in children younger than 4 years[20,21]. This could be either because there is a paucity of instruments to detect the impact of UHL in this age group or researchers believe that UHL affects older children more than infants.

Most studies were limited by the small sample sizes available to the researchers, with the potential to meaningfully combine data from different studies being further inhibited by a failure to adequately describe patient groups and heterogeneity in outcome reporting. By way of example, of the 41 studies included in this review, 16 studies did not describe the type of hearing loss (sensorineural, conductive or mixed). In addition, significant heterogeneity was found in the outcome measurement instruments used, understandable to an extent due to the necessity of matching instruments to life-stage across childhood. The only domain with homogeneity with regards to the instrument used was cognition, although the limitations relating to patient characterization applied to this domain as well. A meta-analysis[47] combining the results from four studies investigating the impact on cognition reports negative impact of UHL on cognition. However, this meta-analysis combines low risk and high-risk (of hearing loss due to stay in NICU) children, which may affect the generalisability and applicability of the findings of this meta-analysis

When evaluating the impact of UHL, many studies investigated more than one domain. 18 studies investigated more than one domain. Although it is helpful to understand how the impact of UHL on one domain may contribute to an impact on another domain, findings on individual domains do not fully charecterise the significance of the impact of UHL on a child's development. Specific quality of life measures relating to hearing loss in children are likely to aid in detecting the global impact. Although the HEAR-QL[63] has been designed to measure specific QOL it requires further validation.

The only outcome domain where there was a consistent negative impact of UHL was hearing difficulties in real-life, such as hearing-in-noise and the ability to localise sound. The extent to which UHL impacts on education, cognition and speech & language remains contentious. The existence of multiple factors recognised to potentially affect speech & language development, cognition and educational attainment, means that it is difficult to reliably establish the true impacts of UHL by controlling for all of the possible confounding factors. Interventions that have been shown to be beneficial are ideal positioning in classroom[29], use of educational resources[36] and support from parents and teachers[65]. Although benefit from existing interventions in UHL may limited[66,67,68], there remains a clear need to manage parental anxiety regarding the impact of their child's UHL[65].

6. Conclusion

The impact of UHL in children remains contentious, although the evidence for a potential negative impact upon various aspects of childhood development is increasing. As such, the traditional belief that the impact of UHL on childhood development is negligible, or insignificant, is no longer generalizable, necessitating an individualized approach based on a clear understanding of the potential negative consequences.

7. Legend for tables and Figures

Figure 1.2 - PRISMA flow chart of search for systematic review

Table 1.2 - Search Strategy Pubmed- UHL-Children

Table 2.2 - PICOS criteria for inclusion and exclusion of studies

Table 3.2 - Quality of Studies

Table 4.2 - Levels of evidence

Table 5.2 - Summary of studies examining real life hearing.

Figure 1.2 - PRISMA flow chart of search for systematic review

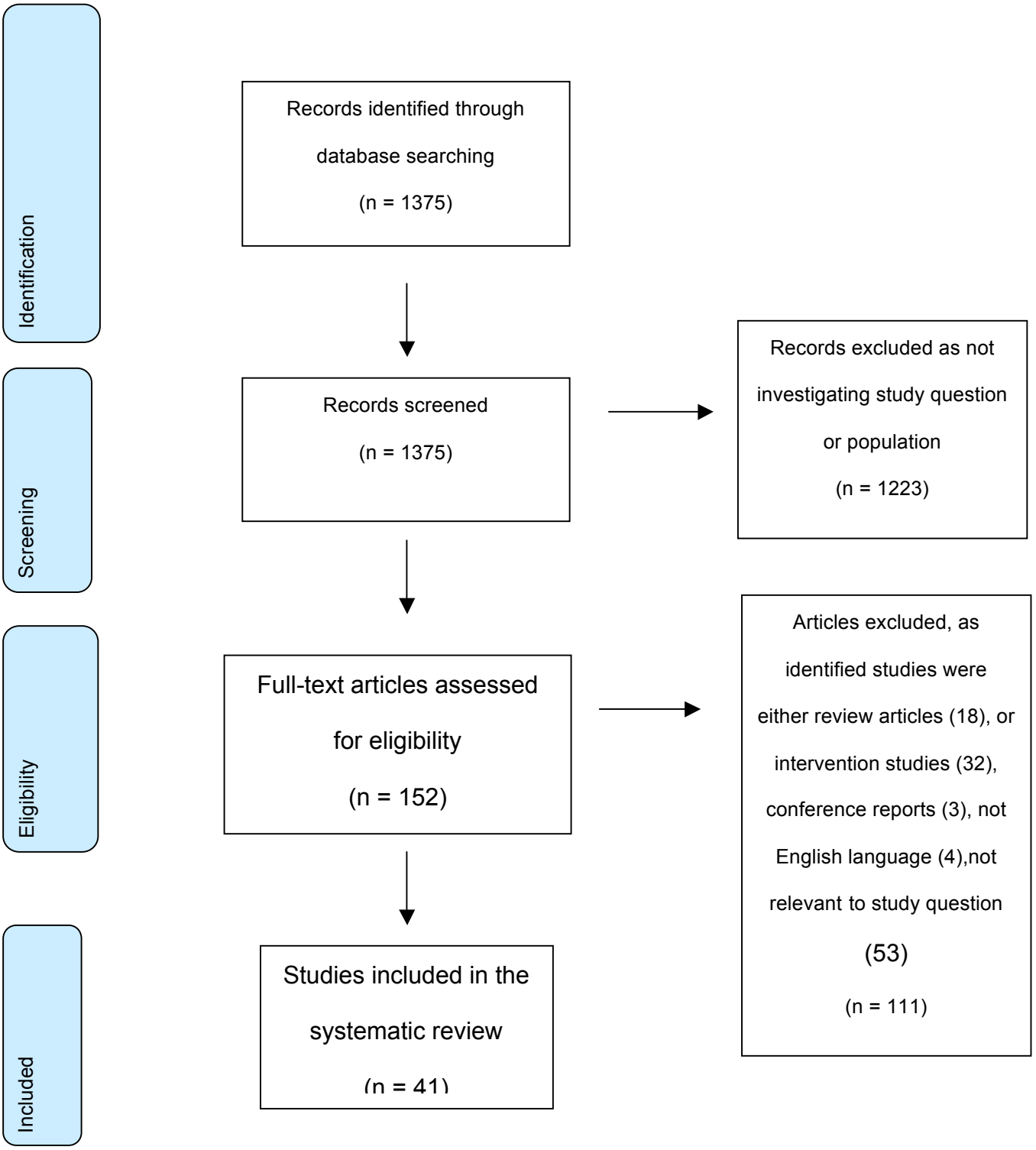

Table 1.2- Strategy 410858 16 Apr 18 - 17:27

# Database	Search term	Results
1. PubMed	(Unilateral). ti, ab	123786
2. PubMed	(hearing loss).ti,ab	84336
3. PubMed	(1 AND 2)	4272
4. PubMed	(children).ti,ab	2356641
5. PubMed	(3 AND 4)	1463

Table 2.2 PICOS criteria for inclusion and exclusion of studies

Participants	Children aged <18
Intervention	Hearing loss in one ear
Control	Normal hearing
Outcomes	Impact of intervention in identified children.
Study designs	Peer-reviewed experimental studies; randomized controlled trials (RCT), cohort observational studies, longitudinal and cross section studies and qualitative studies

Table 3.2-Quality of Studies

Studies	**Quality of Reporting Q1-Q10**	**External ValidityQ11-Q13**	**Internal Validity-Bias Q14-Q20**	**Internal Validity-Confounding Q21-Q26**	**Power-Q27**	**Total**
Fitzpatrick et al. (2019)	8	1	2	2	0	13
Fitzpatrick et al. (2015)	6	1	3	1	0	11
Priwin (2007)	6	2	4	1	1	14
Reeder et al. (2015)	6	0	4	1	0	11
Bess et al. (1986)	7	2	3	2	0	14
Lewis et al (2016)	5	2	4	1	0	12
Priwin et al (2007)	4	0	4	0	0	8
Noh & Park (2012)	4	0	4	0	0	8
Welsh et al (2012)	2	0	2	0	0	4
Ruscetta et al (2005)	5	2	2	1	1	11
Lieu et al (2013)	9	0	3	1	1	14
Peckham &Sheridan (1976)	2	0	0	3	0	5
Keller & Bundy (1980)	8	1	4	4	0	14
Brookhouser et al (1991)	2	3	0	2	0	7
Culbertson & Gilbert (1986)	7	3	3	2	0	15
Kesser (2013)	6	0	0	1	0	7
Jensen et al (2013)	7	0	2	0	0	9

Lieu et al (2012)	8	2	4	3	0	17
Bess & Tharpe (1986)	2	0	0	1	0	3
Reed (2016)	8	1	2	1	0	12
Klee & Davis-Dansky (1986)	7	2	6	3	0	18
Emmett & Francis (2014)	8	2	2	2	0	14
Kiese -Himmel (2002)	8	2	2	1	0	13
Martinez-Cruz et al (2009)	7	2	3	1	0	13
Ead et al (2013)	8	0	3	1	0	12
Schmithorst et al (2014)	7	2	3	2	0	14
Fischer & Lieu (2014)	8	2	1	2	0	13
Niedzielski et al (2006)	6	2	2	1	0	11
Lieu et al (2010)	8	2	2	2	0	14
Borg et al (2007)	8	2	3	2	0	15
Laugen et al (2017)	8	2	2	1	0	13
Wolter (2016)	8	0	3	1	0	12
Vasama and Makela (1997)	4	1	1	1	0	7
Jung et al (2017)	10	2	4	1	0	17
Wu et al (2009)	6	0	4	1	0	11
Propst et al (2010)	8	2	4	1	0	15
Tibbetts et al (2011)	8	2	4	1	0	15
Schmithorst et al (2005)	9	2	4	1	0	16
Zhang et al (2016)	8	1	4	1	0	14

Table 4.2 Levels of evidence

Level	Type of evidence
I	Large RCTs with clear cut results
II	Small RCTs with unclear results
III	Cohort and case-control studies
IV	Historical cohort or case-control studies
V	Case series, studies with no controls

Table 5.2 - Summary of studies examining real life hearing

Study	No. of groups	Type of Study	Groups	N	Type of hearing loss	Severity of hearing loss	Age in years	Measures	Instrument
Fitzpatrick et al. (2019)	3	Cross-sectional case-control study	Unilateral Hearing Loss	38	24/14- (Sensorineural/Conductive)	Mild-Profound	1-3	Hearing performance in quiet & noise	PEACH-
			Mild Bilateral Hearing Loss	31	28/3- (Sensorineural/Conductive)	Mild-Moderately severe		Listening difficulties	CHILD
			Normal Hearing	51					
Fitzpatrick et al. (2015)	3	Longitudinal case-control study	Unilateral Hearing Loss	31	Not described	Mild-Profound	1-3	Hearing performance	PEACH
			Mild Bilateral Hearing Loss	24		Mild-Moderately severe		Listening difficulties Listening difficulties	Early listening function CHILD
			Normal Hearing	45					
Priwin (2007)	3	Case-control study	Unilateral Hearing Loss	13	Conductive hearing loss	Moderate-Severe	6-18	Speech recognition in noise	Speech audiometry
			Bilateral Hearing Loss	9				Localisation	5 loudspeaker set-up
			Normal Hearing	15				Questionnaires	MAIS, MUSS
Reeder et al. (2015)	2	Case-control study	Unilateral Hearing Loss	20	Not described	Profound	6-18	Speech in noise	HINT
			Normal Hearing	20				Localisation	140^{o} arc-15 speakers
								Parent reported questionnaire	SSQ
Bess et al. (1986)	2	Case-control study	Unilateral Hearing Loss	25	Not described	Moderate-Severe	6-13	Localisation	180^{o} arc-13 speakers
			Normal Hearing	25				Speech recognition in noise	Nonsense syllable test
Pirwin et al (2007)	1	Observational study	Unilateral Hearing Loss	25	Congenital conductive loss	Moderate-Severe	3-20	Speech recognition in noise	Swedish PB list
								Questionnaire	H-70 questionnaire

Noh & Park (2012)	3	Case-control study	Unilateral Hearing Loss	25	Not described	Severe - Profound	10-19	Speech recognition in noise	Nonsense syllable test
			Normal Hearing-Children	25			10-19		
			Normal Hearing-Adults	25			22-32		
Welsh et al. (2004)	3	Case-control study	Normal Hearing	19	Sensorineural hearing loss	Severe - Profound	9-73	Speech in noise	Speech in noise test
			Unilateral Hearing Loss	16			7-73		
			Bilateral Hearing loss	20			54-84		
Ruscetta et al (2005)	2	Case-control study	Unilateral Hearing Loss	20	Not described	Severe - Profound	6-14	Speech in noise	HINT Nonsense syllable test
			Normal Hearing	17					
Lieu et al (2013)	2	Case-control study	Unilateral Hearing Loss	107	Sensorineural hearing loss	Mild - Profound	6-12	Speech in noise	Word recognition scores
			Normal Hearing	94					

Table 6.2- Summary of studies - Impact on education

Study	No. of groups	Type of Study	Groups	N	Type of hearing loss	Severity of hearing loss	Age in years	Measures	Instrument
Peckham & Sheridan (1976)	1	Longitudinal observational	Unilateral hearing loss	46	Not described	Moderate	7-11	Parental report Medical report Educational report	Parental interview Medical exam Educational scores
Keller & Bundy (1980)	2	Case control	Unilateral hearing loss Normal hearing siblings	63 23	Not described	Not described	9-14	Scholastic standardized scores	Stanford achievement test Metropolital achievement test
Brookhouser et al (1991)	1	Longitudinal observational	Unilateral hearing loss	172	Sensorineural	Mild to profound	< 19 years	School performance	Teacher reports
Culbertson & Gilbert (1986)	2	Case control	Unilateral hearing loss Normal hearing	25 25	Not described	Moderate to severe	6-13	Cognitive performance Academic performance Behavior Self-concept	WISC –R, Hiskey-Nebraska School achievement tests Behavior rating scale Piers-Harris Children's self-concept scale
Kesser et al (2013)	2	Survey	Unilateral hearing loss Unilateral hearing loss	40 12	Conductive Sensorineural	Not described	>5 years	Academic performance Behavior	Use of individualized educational plan Repeat a grade Use of speech therapy Behavior problems
Jensen et al (2013)	3	Retrospective review	Right aural atresia Left aural atresia Bilateral atresia	48 19 7	Conductive	Moderate to severe	>2 years	Speech & Language Education	Speech pathologists report Parental report
Lieu et al (2012)	1	Prospective longitudinal cohort	Unilateral hearing loss	46	Sensorineural/ Mixed/Conductive	Mild to severe	6-8	Cognitive Achievement Language Behavior Academic performance	WASI WIAT-II-A OWLS CBCL Educational plan
Bess & Tharpe (1986)	1	Case-note review	Unilateral hearing loss	25	Sensorineural	Moderate to severe	6-18	Academic performance	Teachers report

Reed et al (2016)	2	Survey	Right aural atresia Left aural atresia	94 46	Conductive	Moderate to severe	1-30	Academic performance Resource utilization Behavior	Survey
Klee & Davis-Dansky (1986)	2	Case control	Unilateral hearing loss Normal hearing	25 25	Sensorineural	Mild to profound	6-13	Language measures Cognition Educational progress	Battery of tests WISC- R Grade repetition
Fischer & Lieu (2014)	2	Case control	Unilateral hearing loss Normal hearing siblings	20 13	Sensorineural	Severe to profound	12-17	Education progress	Grade repetition

Table 7.2- Summary of studies - Impact on cognition

Study	No. of groups	Type of Study	Groups	N	Type of hearing loss	Severity of hearing loss	Age in years	Measures	Instrument
Emmett & Francis (2014)	2	Cross-sectional	Unilateral hearing loss Bilateral hearing loss Normal hearing	162 46 4615	Not described	Moderate to severe	6-19 years	Cognition	WISC-R Wide range achievement test
Klee & Davis-Dansky (1986)	2	Case control	Unilateral hearing loss Normal hearing	25 25	Sensorineural	Mild to profound	6-13 years	Language measures Cognition Educational progress	Battery of tests WISC- R Grade repetition
Kiese - Himmel (2002)	1	Observational study	Unilateral hearing loss	31	Sensorineural	Mild to profound	1-10 years	Cognition Language development	CMMS Colored progressive matrices Multiple tests
Culbertson & Gilbert (1986)	2	Case control	Unilateral hearing loss Normal hearing	25 25	Not described	Moderate to severe	6-13 years	Cognitive performance Academic performance Behavior Self-concept	WISC –R, Hiskey-Nebraska School achievement tests Behavior rating scale Piers-Harris Children's self-concept scale
Lieu et al (2013)	2	Case-control study	Unilateral Hearing Loss Normal Hearing	107 94	Sensorineural	Mild - Profound	6-12 years	Speech in noise Cognition	Word recognition scores WASI
Martinez-Cruz et al (2009)	2	Case-control study	Unilateral Hearing Loss Normal Hearing	21 60	Sensorineural	Moderate to profound	7 years	Cognition	Stanford- Binet intelligence scale
Ead et al (2013)	2	Case-control study	Unilateral Hearing Loss Normal Hearing	7 7	Sensorineural	Severe to profound	9-14 years	Cognition	Multiple tests
Schmithorst et al (2014)	2	Case-control study	Unilateral Hearing Loss Normal Hearing	21 23	Sensorineural	Moderate to profound	7-12 years	Cognition Central processing	WISC-IV FMRI studies
Fischer & Lieu (2014)	2	Case-control study	Unilateral Hearing Loss Normal Hearing	20 13	Sensorineural	Severe to profound	12-17 years	Language Cognition	OWLS CELF WASI
Niedzielski et al (2006)	1	Observational study	Unilateral hearing loss	64	Sensorineural	Profound	6-16 years	Cognition	WISC- R

Lieu et al (2010)	2	Case-control study	Unilateral Hearing Loss Normal Hearing	74 74	Sensorineural	Profound	6-12 years	Cognition Language	WASI OWLS
Lieu et al (2012)	1	Prospective longitudinal cohort	Unilateral hearing loss	46	Sensorineural/Mixed/Conductive	Mild to severe	6-8	Cognitive Achievement Language Behavior Academic performance	WASI WIAT-II-A OWLS CBCL Educational plan

Table 8.2 - Summary of studies - Impact on speech & language

Study	No. of groups	Type of Study	Groups	N	Type of hearing loss	Severity of hearing loss	Age in years	Measures	Instrument
Fitzpatrick et al (2015)	3	Longitudinal case-control study	Unilateral Hearing Loss Mild Bilateral Hearing Loss Normal Hearing	31 24 45	Not described	Mild- Profound Mild-Moderately severe	1-3	Hearing performance Listening difficulties Listening difficulties	PEACH Early listening function CHILD
Peckham & Sheridan (1976)	1	Longitudinal observational	Unilateral hearing loss	46	Not described	Moderate	7-11	Parental report Medical report Educational report	Parental interview Medical exam Educational scores
Lieu et al (2012)	1	Prospective longitudinal cohort	Unilateral hearing loss	46	Sensorineural/Mixed/Conductive	Mild to severe	6-8	Cognitive Achievement Language Behavior Academic performance	WASI WIAT-II-A OWLS CBCL Educational plan
Kiese-Himmel (2002)	1	Observational study	Unilateral hearing loss	31	Sensorineural	Mild to profound	1-10 years	Cognition Language development	CMMS Colored progressive matrices Multiple tests
Fitzpatrick et al (2019)	3	Cross-sectional case-control study	Unilateral Hearing Loss Mild Bilateral Hearing Loss Normal Hearing	38 31 51	24/14-(Sensorineural/Conductive) 28/3-(Sensorineural/Conductive)	Mild- Profound Mild-Moderately severe	1-3	Hearing performance Hearing performance Listening difficulties Receptive & Expressive language	PEACH-Quiet PEACH-Noise CHILD Battery of tests
Lieu et al (2010)	2	Case-control study	Unilateral Hearing Loss Normal Hearing	74 74	Sensorineural	Profound	6-12 years	Language	OWLS
Lieu et al (2013)	2	Case-control study	Unilateral Hearing Loss Normal Hearing	107 94	Sensorineural hearing loss	Mild - Profound	6-12	Speech in noise Cognition Achievement	Word recognition scores WASI Wechsler Individual Achievement Test OWLS

								Oral language skills	
Fischer & Lieu (2014)	2	Case-control study	Unilateral Hearing Loss Normal Hearing	20 13	Sensorineural	Severe to profound	12-17 years	Language Cognition	OWLS CELF WASI
Borg et al (2007)	11	Case-control study	Unilateral hearing loss Normal hearing	156 97	Sensorineural & conductive	Mild to profound	4-6 years	Language skills	Multiple tests
Klee & Davis-Dansky (1986)	2	Case control	Unilateral hearing loss Normal hearing	25 25	Sensorineural	Mild to profound	6-13	Language measures Cognition Educational progress	Battery of tests WISC- R Grade repetition
Laugen et al (2017)	3	Case control	Normal hearing Unilateral and mild bilateral hearing loss Moderate to severe hearing loss	123 14 21	Sensorineural	Mild to profound	4-5 years	Language Social skills	Receptive vocabulary Social skills rating syster report

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Chapter 3:The perspectives of children on the impact of unilateral conductive hearing loss on their lives – A qualitative study

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Abstract

Objective: To understand the perspectives of children regarding the impact of unilateral conductive hearing loss on their lives.

Methods: Semi-structured interviews with 11 children with a known diagnosis of unilateral conductive hearing loss, aged 11-17 year, recruited from a tertiary paediatric hospital. A qualitative descriptive exploratory approach was used with data coded and categorized using NVIVO data analysis software.

Results: Six categories were identified: (i) self-perception of hearing ability, (ii) adaptations and impact of adaptations (behaviour issues and fatigue), (iii) support from friends, families and teachers, (iv) dealing with being different, (v) concerns about safety, and (vi) implications on future life. Children described their hearing as "normal'; however, exploring their daily routine,

revealed hidden problems with hearing in noise, localisation, and hearing the teacher in the classroom. Children described how they adapted for example positioning themselves, in an attempt to overcome their hearing issues. They talked about some of the difficulties they encounter as a result of adaptations. Families and teachers supported them through offering additional help in class, and at home. Friends helped with bullying at school and supported them in the classroom. Not only were children dealing and adapting to the physical impact of hearing loss, but were also having to deal with being different from their peers. At the same time children had considered the impact of their hearing loss on safety on roads, and took extra care to rely more on looking for traffic prior to crossing roads. They were aware of possible implications of their hearing loss on their future careers and general well being.

Conclusions: Children with unilateral conductive hearing loss are keen to normalize their hearing loss and adapt to the resulting difficulties. There are, however, hidden problems, which need to be identified, by parents, teachers and caregivers to ensure appropriate treatment strategies can be advocated. This study highlights patient-specific outcome domains that are most relevant to children with unilateral conductive hearing loss.

1. Introduction

Unilateral hearing loss (UHL) is defined as average air conduction thresholds at frequencies 0.5, 1 and 2 KHz ≥ 20 dB HL in the impaired ear and ≤ 15 dB HL in the better ear [7]. Congenital UHL is reported to affect 0.81/1000 live births [32] and these children are diagnosed within 4-5 weeks of birth due to the provision of universal newborn hearing screening programs. In addition, progressive hearing loss and many acquired conditions can also result in UHL. The prevalence of UHL has been reported to range from 3% to 6.3% in children aged 6-19 years [14]. With early identification of UHL, there is an increasing need to understand the full impact of different types and severity of UHL. Multiple studies have investigated the impact of UHL in childhood focusing on hearing in noise [33-39], cognition [39-41], speech and language development [39, 42, 43] and education [44-46]. Studies measuring quality of life specific to children (PedsQL) have been used to assess the impact of UHL in children [47, 48]. This measure, however, is not specific to UHL. Specific instruments to assess the impact of a permanent unilateral hearing loss in children are lacking. In order to develop such an

instrument, there is a need to first identify how this condition impacts an affected child. Qualitative studies have an important role in understanding the impact of health issues on affected individuals [49].

We identified two qualitative studies, which explored the impact on education and psychosocial impact of UHL[47, 50] however; there were no studies that investigated the global impact of UHL as perceived by the affected child. Other limitations are:

- Most studies do not describe the type (congenital/acquired), severity (mild/moderate severe) or nature (conductive /sensorineural) of hearing loss affecting their participant group
- Children with UHL are often investigated along with mild bilateral hearing loss and grouped under minimal hearing loss
- Results of studies are generalized to all types of hearing loss

Therefore, there is a need to understand the child's perspectives of the impact of UHL. As management options will often depend on the nature of the hearing loss, whether sensorineural or conductive, it is useful to investigate these two subsets separately. Children with a conductive hearing loss will have normal inner ear function. The presence of a normally functioning cochlea allows options for treatment of the unilateral hearing loss, with the use of hearing aids and improves their natural hearing. Hence, we chose to focus our attention, in this study, on understanding the impact of unilateral conductive hearing loss.

2. Aim

To understand the perspectives of children regarding the impact of unilateral conductive hearing loss on their lives.

3. Method

A qualitative, descriptive exploratory approach was used to understand the impact experienced by the affected child. The study methods were guided by grounded theory methodology, including the use of semi-structured interviews, purposive sampling, constant comparative analysis, and coding [51, 52]. We chose this approach as it facilitates understanding of the issues from the perspective of participants, encouraging them to be open and avoids introducing assumptions about what is important from a professional's perspective. The topic guide was adapted as new themes developed, as is standard practice.

3.1 Participants

Participants for this study were recruited from the audiology database of a tertiary Children's Hospital. We chose to study children from 11 years of age, as they would be able to participate in the interviews and describe their experiences. Additionally, we considered that they would be able to provide perspective on their experience of UHL during earlier childhood. The education system in the UK is structured such that 11 years of age represents a significant development stage, with transition from primary to secondary level education, characterized by a more demanding listening and educational environment. National Research and Ethics Committee (NREC) approval and Health Research Authority (HRA) approval was obtained prior to recruitment (REC reference 16/LO/1080). Written informed consent was obtained from the parent/legal guardian and assent from the child, prior to performing any study-related procedures.

3.1.1 Participant selection criteria

Inclusion Criteria:

1. Aged 11 to 17 years.
2. Conductive hearing loss (CHL) with normal bone conduction thresholds and air conduction thresholds worse than 25 dB HL (as an average of measured thresholds at 0.5, 1, 2 and 4 kHz*)* in the worse hearing ear.
3. Normal hearing in the contralateral, unaffected, ear (hearing thresholds ≤ 25 dB HL)
4. English speaking child and parent.
5. Congenital and acquired unilateral CHL, with purposive sampling to ensure both groups represented.

Exclusion Criteria:

1. Children with learning difficulties as reported by parents.
2. Dual sensory deficit, confirmed by parental report of visual impairment.
3. Families with difficult social circumstances identified by the clinical staff (e.g. recent bereavement in family, child in the care of local authority).

Thirty-four children with conductive hearing loss meeting the inclusion criteria were identified and contacted by letter. Although 14 parents agreed for their child to take part in the study, three parents were unable to attend their appointment due to unforeseen circumstances and did

not want to rearrange the appointment, meaning 11 children completed the interview (32% response rate). Those declining to participate in the study either said that their hearing was not an issue, they were not keen to take their child out of school, time constraints precluded participation, or they were not contactable by phone. All children completed their interviews. Participant characteristics are presented in Table 1.

3.2 Data collection

3.2.1 Semi-structured interviews

Semi-structured interviews were conducted at the parents' choice of venue, either in an interview room at the hospital or in the participant's home. Only one participant's parent chose that the interview be conducted at home. Two interviewers, one of the authors JN and a research audiologist, conducted the interviews. As some of the recruited children and parents were well known to one of the authors (JN), due to their clinical condition, where possible the research audiologist to ensure independence from the clinical relationship in the interviews conducted the interviews. The research audiologist conducted all but four interviews. Interviews with children were conducted with reference to a topic guide, in the presence of their parents and lasted approximately 30 minutes. The topic guide (Appendix 1.3) contained a set of open-ended questions used as prompts for the interviews and was adapted to include emerging ideas and to be responsive to the agendas of importance to participants. In addition to the topic guide, we used a slideshow of neutral prompts about the pattern of a typical day (e.g. getting up, going to school, time with friends and family etc.) as a prompt to encourage participants to talk about how UHL affected their daily living.

Data analysis was concurrent with data collection, in order to respond to emergent themes. The interviews were audio recorded, transcribed, and anonymized. Data was analyzed using computerized qualitative data analysis software NVIVO for Mac© (http://www.qsrinternational.com). Data was analyzed in three stages. The first stage was in-vivo coding, where we coded sentiments described by children using their own words. Codes with similar sentiments were amalgamated into secondary codes. Further analysis of secondary codes allowed us to link the codes together to meaningful categories. Results of the interviews were analyzed and emerging categories are presented with quotations from participants. Names in the quotations are pseudonyms to maintain participants' anonymity.

4. Results

Table 2 summarises the categories and subcategories developed from combining meaningful data. We identified six categories. Some of the categories had subcategories and are described in detail.

4.1. Self-perception of hearing

4.1.1 Normalizing the hearing loss

All our participants had experience of long-standing unilateral conductive hearing loss. Ten children said that they could hear well at the start of the interview and when certain aspects of hearing in daily life were explored. As their hearing in one ear was normal, they said they did not perceive difficulties hearing in their day-to-day life. One child went on to say that his hearing on one side was "more powerful than others." Participants appeared keen to emphasise the normality of their hearing and lack of problems with their hearing. They described situations in their daily routine for example at school and during other activities, where hearing loss in one ear has not contributed to any difficulties.

> "Yeah, nothing's affected by my hearing, I feel like it's just like normal". (Sophie)

> "No, because usually it's just like one or two or three of us like messing about in the morning playing football. So, then what we do is just pass it around or play wallie, or something. So, it doesn't make much difference, my hearing. (Gerrard)

> "Like it didn't really bother me 'cause I still can hear perfect" (Ellie).

> "Well sometimes they said that we can choose (where I sit in class), at that point I normally do go to the back and then…so I normally go to the back with my friends and I can still hear fine. (Luke)

> "No because when I play my drums it doesn't do anything to my hearing. Like if someone was standing next to me and I was playing drums, and even if I had my ear

defenders on, then I could still hear them and the drums, and even if I had both hearing, it'd still be the same with the drums playing. (Liam)

4.1.2 Hearing difficulty in certain places or settings

As interviews progressed details of their everyday lives were discussed, and problems with hearing difficulties were identified. The areas of hearing difficulties that children highlighted were, hearing in certain places, such as in a noisy room or a large hall, certain situations, when there were many individuals around them, talking at the same time or certain settings, such as a social gathering or a discussion. Some participants also talked about the impact on their education, where they would sometimes struggle to hear the teacher. This was particularly a problem if the lesson was interactive and there were discussions taking place.

"Hearing's good in school, it's not really bad. The dinner hall is bad because most people are talking, and the sports hall is also bad. They're the two main places." (Edward)

"Yeah, I feel quiet comfortable, but if there's a lot of people and it's a crowded place and your mate's trying to talk to you, or family, it kind of gets a bit difficult to hear, and I always have to look at them, and then look where I'm going." (Leo)

"Just if everyone's talking and the teacher's trying to talk I can't listen to the teacher." (Ellie)

"Mostly science just because it's all discussions and stuff, and then miss will always speak during those discussions, so I can never hear her. " (Nathan)

4.1.3 Hearing difficulty when further away from source of sound

When we explored issues at school or out of school, children mentioned that hearing when further away from the source of sound could sometimes be difficult. Having identified this difficulty, children described the steps they took to overcome the difficulty. However this was not

always possible, for example when the seating plan in the class was based on ability of the child, children described struggle hearing the teacher.

> "I mean, if there is a lot of children and I can't hear, I just get closer and that's it." (Luke)

> "PE, I've got one that has quite a lot of noise in. He'll have a whistle, and that's when everyone should come in to speak to him, and I always get to the front, so I can always hear". (Nathan)

> 'Yeah. And if you were at the back, what way would you struggle?' 'Maybe not catching something if the teacher's said it quietly, but if she's, like, normally saying it and saying it to the whole class then probably I'd most likely hear it.' (Luke)

> "Sometimes 'cause I'm quite a shy person to ask for help 'cause in my English especially I sit quite far away from the teacher so it's harder to ask and hear people" (Ellie)

4.1.4 Hearing difficulty when not facing the speaker

Another difficulty that came to light was hearing when not facing the speaker, particularly in a classroom setting. This was an issue when they were in a group and having a conversation and struggled to hear their friends/family not directly facing them. Although the participants did not talk about these issues directly, these difficulties with their hearing become apparent through the adaptations they had made. Children pointed out that if they were having conversations with friends or family, and the source of sound was on the side of their hearing loss, they would struggle hearing. They often positioned themselves such that they would have their normal hearing ear towards the speaker. This occurred frequently at home, with friends and in school.

> "Like if a teacher does come over to me I have to look at them, I have to look at what they say to be able to fully concentrate and hear them." (Ellie)

> "Depending where I'm sat, if it were to be at the back, I'd have a little bit of a struggle, but if it were at the front I'd probably…I mean, if I was facing the right way I'd probably hear it." (Luke)

"When I am out with my sisters, if one of my sisters says, something, I know they are talking to me, but I have to look at them to get what they are saying" (Eva)

"No, the only time it's affecting, is if it's, like sat in the classroom, I can hear the teachers, I can hear everything, it's just like if someone's here, on the side of that ear, that I can't hear, and that's the only time it affects me. (Sophie)

4.1.5 Localising sound

Identifying the direction sound came from was difficult for the participants. Although this was not an issue highlighted directly, we were able to identify this difficulty though the adaptation they had made. Children often said they looked around and figured out where the sound was coming from. This suggests children with UHL are possibly making changes and reacting in a manner that allowed them to locate the source of sound and not see this as a difficulty. Only one participant predicted their inability to localise sound as a potential problem for the future.

"Yeah, I do but I don't really in football but I do in other things, like if someone shouts my name I'll probably wouldn't know where they're coming from" (Leo)

Interviewer: you always know where they are and where they are shouting from?
Dan: Yeah. Because then you're looking with your eyes. (Dan)

Interviewer: so what would you do, if someone shouts for you would you just... Leo-yeah, just look around. (Leo)

"Getting a car and being able to drive, and go to places, but with hearing that might be a bit of a difficulty, for traffic and cars and stuff, and ambulances et cetera. So, I'll have to properly look into it, and see what I can do to find something out about that." (Nathan)

4.2 Adaptations and impact of adaptations

4.2.1 Adaptations

Children adapted their routine on a daily basis to overcome some of the issues highlighted above. Some of the changes they made were deliberate, while others were not. All participants interviewed said they positioned themselves in a certain manner to hear well. They would either face the speaker, or position themselves with their normal-hearing ear facing the speaker. They moved to get themselves into this position. Sometimes their friends and family would change their position to help. In school, however, some preferred to do this discreetly without attracting attention to their need to be facing the teacher, whilst others were open about it.

> "In every lesson I always sit with my right ear facing the teacher." (Edward)

> "Ask if I could sit at the front" (Luke)

> And when I walk with people, if I'm on this side and they're on the other side this my bad side, I always make sure I tell them change just so I could hear them properly." (Ellie)

.

> "They (brothers) always come right up in front of me, so I can always see and hear them". (Nathan)

> "Yeah, I mean that's always just been what I do. If it's someone with me, just like one on one, then I always walk on that side of them, or sit on that side of them." (Sophie)

4.2.2 Impact of adaptations-behaviour issues and fatigue

When children struggled to hear in a classroom setting, some asked the teachers to repeat themselves, but they felt that teachers sometimes got frustrated with. Others were concerned about the disruption this would cause, and chose to get help at the end of the lesson. Many relied on friends to fill them in. This did result in children and their friends getting detentions as they were perceived to be disrupting the class and raised issues about behaviour in classroom.
Children talked about their hearing loss being "stressful", caused tiredness, affected their concentration and caused distraction. This in turn affected their performance at school and ability to fully engage.

"My friends don't really need to help. If I haven't heard the teacher, I ask them to repeat what they have said" (Eva)

"I've not been paying attention, but it's not, it's just because I physically couldn't hear them, and then when you put your hand up to tell him, he usually shouts at you for it and stuff. So, yeah, it isn't the best thing in the world to have that constantly. But, if you go to them after lesson to speak to them, some of them are understanding about it, but some aren't." (Nathan)

"I don't know whether it's my hearing or whether I just block out the outside world. I kind of just get a bit of distracted. So, if I'm actually trying to hear, I can hear absolutely fine. So, I don't know if that's just that I don't listen." (Sophie)

"A fair amount, to the point where it was getting quite annoying, just having the constant stress of being told off by teachers when you haven't done anything wrong. Probably when I go home, I usually end up being really down about it. And I never really say to my mum and dad, I never really speak about it. I always end up getting stressed out, so I probably take it out on my brother and sister, which obviously isn't nice for them, to have to come home to their brother, and I'll just be nasty to them." (Nathan)

4.3 Support from others

4.3.1 Support from teachers

Children talked about the extra resources and lessons needed in school and at home to catch-up to their peers. Some of them missed school due to the need to attend hospital appointments, whilst others found extra help useful to catch up on information they may have missed at school. Although they found this extra help beneficial, they also saw this as an additional burden on top of their lessons. Some children also talked about the additional help they got during exams.

"So I usually find a room that's quite quiet, so I can just sit and get it all done. Then mum will go over it to see what parts I've got right, or parts I haven't, so I've got it all done, and then I can just do and do what I want." (Nathan)

"I've like fallen back with my education. So, I get an extra lesson with my Maths like to boost me up, and then English to boost me up, an extra hour, and then the next hour's like just talking about general life, and stuff, yeah. And then in class…yeah, and then in quite a few of my lessons it's like a couple of them, there's always like a support teacher and like once or twice he always comes over to me, ask me if I got most of it like written down or if I've heard it." (Gerard)

"Personally, I hated having the tutors. And it's not going to mean anything, it didn't change a thing." Luke

"Good to have the tutor, because every time I have the tutor, the next Maths lesson I have the next day, I know it." (Edward)

4.3.2 Support from friends and family

Children described the help and support from their parents and siblings from being woken up in the morning to help with their schoolwork. They also talked about their siblings looking out for them. Two children talked about how their siblings had helped them maneuver the traffic. They also benefitted from emotional support. In addition friends and siblings helped them overcome unpleasantness and bullying in school.

"Nearly got run over as I was not looking when crossing the road, but my sister shouted out and I luckily escaped." (Eva)

"When I'm outside of the house, if I'm on my bike I hear cars coming but if I don't they (my sisters) normally just say, there's a car coming." (Edward)

"If people ever do call me my friends would stick up and say it's not right, there's nothing wrong with her, she's just different to everybody else". (Ellie)

"It isn't always the nicest thing because I might get picked on by Year 11s, more older people than me, and get pushed about. Which would then put a downer on me and then I end up taking it out on other people and that isn't the best thing to do. Luckily enough I have friends who always stick up for me and will always be there, but if it weren't for them I don't know what I'd do." (Nathan)

4.4 Dealing with being different

Although most children were not worried about their hearing, they had to deal with being different to their peers. Children said they had been bullied or picked upon for being different. This happened at different ages. Some children talked about appearing ignorant when they had not heard a question fully and their response to the question was inappropriate. Children were keen to avoid attracting attention, particularly when asked about the use of hearing aids, they said that the benefit was limited and it attracted attention to their hearing loss. Participants who had not trialed a hearing aid, were keen to try one, but also said they wanted to avoid anything 'big and visible' This concern for hearing aids however was not shared by all. Some of the children were keen to try a hearing aid, so they could "hear well".

"If it's a friend, it's fine, 'cause they know. If it's someone I don't know that well, then it's a bit embarrassing, in a funny way, 'cause they're like, why, can't you hear anything? Yeah, it's the hearing. It's just the funny side of having to say, what, to everything. Like, I don't know what they're laughing at. It bothers me in the way that I have to say, what, every time, but not in the way that I'm embarrassed. 'Cause I used to be, but I'm fine now." (Sophie).

"They'll always pick on me, call me big ears, and stuff like that, and push me about, and try and grab my ear and stuff. Did get picked on because of my weight and stuff, but you kind of try and don't let it get to you." (Nathan)

"I didn't like it, it made me look weird, it made people fuss about me even more than they did beforehand, so, yeah, I didn't like it." (Luke)

"If you ever get a chance of hearing on the other side, I'd say take it because it'll be much better. (Liam)

4.4.1 Dealing with the cosmetic appearance

In the congenital hearing loss group, children talked about the impact of the cosmetic appearance of their microtia. This was of a significant concern and children found different ways of dealing with it. Some children talked about it being due to an injury, whereas others tried to not talk about it, so as not to make a big deal of it. This desire to be normal, however, made them more aware of the cosmetic issue and reduced their concern about the hearing loss itself.

"I'd just like to look a normal boy, because I don't need to hear out of this ear." (Dan)

"No, I don't really tell them about it, because in primary it was quite annoying the fact that everyone was watching out for me, which made me feel different in a way that was bad, but now I just tell them an excuse about an accident, but that normally works.
No, I like it, like, I'd like myself to be from now, nothing else really, just normal." (Luke)

"It was just mainly when I started, everyone was a bit like oh what's different like, why is it like that and stuff and people questioned but it got to me 'cause like I'd never had it from primary so going to another school and then getting people to say things, it just got me. Like they'd ask me, they'd be polite and just ask me like how come that's happened, what's happened or were you born like that, and I'd still get upset about it". (Ellie)

"Yeah. My friends that have come from primary, they don't ask me any more. But my new friends, they ask me like, how did it happen, how long have you had it. And when I used to go to my primary, the little kids, they used to come up to me and they used to

go, what happened to your ear? And I said I was born with it, and they used to say oh, I thought a cat bit it off!" (Liam)

4.4.2 Dealing with recurrent ear infections

In children with an acquired hearing loss, children who suffered from discharge from their ear, found it embarrassing to deal with. They were aware that it was foul smelling and may give a perception that they were not hygienic. They tried to discretely clean their ear in order to avoid questions about the discharge. They also had to deal with certain restrictions the recurrent discharge imposed on their day-to-day activities.

"Discharge annoys me now and again because it like gets on to my pillow case, and that stuff, and then like after like through classrooms I have to like ask if you can go to toilet and like clean it out. It like dribbles down a bit, so yeah." (Gerrard)

"Yeah, on to school grounds, if you wear a coat, you'll get shouted at by the teachers. So I have to do different things to try and stop stuff getting into my ear, for example, like cotton wool, to try and make it, but that draws quite a lot of attention to me and stuff." (Nathan)

4.5 Concerns about safety

When discussing the impact of hearing loss on safety, children described mixed feelings. They were generally confident of gaining independence. Children appreciated that they would not hear cars when the traffic was towards their affected ear. They learnt to rely on looking for traffic and information boards. Two children talked about incidents where they needed some help when they were on roads. They also talked about the need to look more carefully to stay safe.

"I can hear and usually I just look. I want to make sure they're far away and walk." (Dan)

"Yeah. It's just normal because I'll look right, and then I'll look left, and then I'll look right again, and then I'll cross. And then normally I remember what to do, and I just cross the road and nothing bad happens, and yeah, but it's easy." (Liam)

"Yeah, I turn my head a bit more so that I can hear a bit more with that ear." (Leo)

"Mmm, and then…well when I'm like putting bales out I've got to be careful because dad drives around loading, and then I'll like pull sheets off bales and then I've got to be careful where you're driving, don't I?" (Gerard)

"I wasn't looking, when I was nearly knocked down by the car, now I am more careful. I look before I cross:" (Eva)

4.6 Implications on future careers and general well being

With regards to any future plans, most children had an idea of what they wanted to do when they grew up. Children did not feel their hearing loss would have a major impact on their choice of careers/jobs they would like. Some children were keen to explore certain restrictions their hearing loss may impose on a select career options. In light of their concern about difficulties they may encounter due to their UHL, some children had made subtle changes to their choice of future career.

"This is not really got to do with my ear but if I join the army, if I don't get a five, it won't be the end of the world because if I have to do a really bad job, like bricklaying, which I really don't want to do, if I am doing that I'm doing it with the best friends I'll ever have in my life. So that's kind of the upside to it."(Edward)

"I want to be a history teacher ". (Eva)

"Yeah. Well no, he makes concrete panels, but I want to be a wagon driver, so I'll work for my dad delivering panels to places." (Liam)

"I have an idea of being a policewoman but you have to have full hearing to have it thingy so it could affect me to not get the job 'cause of the way it is. Cause you have to have full hearing, you have to listen and everything. So that might be a concern to not have it, not get it." (Ellie)

5. Discussion

The aim of this qualitative study was to gain an insight into the impact of hearing loss, as perceived by children with a conductive UHL. An important finding was how children "normalized" their hearing loss. Other researchers[53] investigating common long-term conditions affecting children and adolescents, such as asthma, have reported this normalizing behavior, where children adapt/restrict their activity to their health, such that the impact on activity is not detected (as the activity is being avoided). But normalizing behaviour in children with UHL has not been reported before. This finding where children normalize their hearing loss by changing their position and overcome their difficulty is an important consideration for parents and health-care.

Only on exploring different areas of daily living, were we able to identify the impact perceived by children. Our qualitative study was able to identify hidden problems of UHL in children and adolescents. This study highlights the hidden impact of UHL and shows how children normalize their hearing loss and adapt to their difficulties. However, hidden problems become apparent when exploring their adaptations. This is an important consideration, not only when investigating the impact, but also when considering treatment and management strategies. Many instruments have been designed by health-care providers to assess the impact of UHL. Most of these instruments test the impact of UHL either on testing children in an audiology booth or based on questionnaires. Some of these questionnaires are directly enquiring children, parents or teachers [29, 47, 48]. Testing in audiology booths to assess some of the difficulties highlighted is not routinely utilized, as it is often time consuming and does not often reflect real life difficulties. Available questionnaires may not be able to detect the hidden problems in children with conductive UHL as the initial response to these questionnaires is likely to be "normalizing" which is a common strategy adopted by the developmental stage of adolescence. As such questionnaires asking about hearing loss may not identify a problem, which is not a criticism of the questionnaire, rather the use in this age group. Simple addition of questions that explore certain hearing situations may highlight the difficulties these children encounter, leading to acceptance of treatment options. This finding may prove invaluable when designing new or adapting available patient reported outcome measures.

As we explored hearing in different aspects of daily routine, we identified areas of difficulty. Hearing in a noisy environment and when many speakers were talking at the same time was one of the aspects of hearing difficulty described by children. This is an area that has been studied by other investigators [33-39, 54] and our findings align with the results of these quantitative studies. Our participants also noted difficulty hearing when they were further away from the source of sound. Research into optimal seating position for a child with UHL has shown that speech discrimination scores of children decreased more as they moved further away from the source of sound when compared to children with normal hearing, with ideal position being seated within 4.35 m from the source of sound [54].

Our participants also described difficulty in localising sound. Children, only becoming apparent when they talk about the adaptations they have made to overcome difficulty in localising sounds such as when crossing roads or playing a sport, do not describe this as a difficulty. Impact on localisation ability in UHL has been investigated by other researchers. Three studies [33, 34, 55] reporting on localisation ability of children with UHL, compared to age-matched normal hearing children, report significant difficulty in localisation ability in children with UHL.

Adaptations children make, appear to contribute to concerns about their behaviour. Children described occasions when they had to seek help from friends and teachers. They checked with their friends in the classroom setting, when they missed part of the instruction. Teachers sometimes considered this chatter in the classroom as disruptive behaviour. Impact of UHL on behaviour has been reported by other researchers [56, 57]. Participants themselves did not see this as disruptive behaviour, but as their attempt at overcoming the difficulty they were facing at school. This difference of opinion on behaviour, where children report differently, to the teacher's perception has been reported in another observational study [58].

Another consequence of adaptation was effort, fatigue and stress. Children described becoming distracted, and tired. This is another important consequence that is best detected through reports from children. Evidence from available literature of this consequence of UHL is limited[59].

A significant proportion of children in our study talked about the need for extra help to catch up at school. Some children needed additional help/tutoring at home or in school to catch up to their peers. Children found this one-to one help teaching useful. This observation has also been reported by other studies [45, 56, 57, 60].

Many of our participants had to cope with the stigma of being different to their peers. They described being “bullied” or “picked on” as a direct result of their hearing loss. This has also been documented in studies investigating the impact of other chronic conditions on wellbeing in childhood [61, 62]. This concern about being bullied was not reported by all of our participants. Many of our participants talked about their friends being supportive. However, some of our participants talked about “not making a big deal of it”. They were keen to avoid talking about their hearing loss as “friends made a fuss about it”. This is in keeping with adolescent behavior associated with chronic conditions [61, 63]. These findings highlight the importance of taking the adolescent behavior patterns and beliefs into consideration when managing children with UHL

6. Strengths and Limitations

In depth interviews with children to identify impact perceived by children with UHL is one of the main strengths of our study. We also focused our study to evaluate a very specific group of children with UHL. In addition multi-disciplinary analysis was instrumental in understanding the issues that children were describing.

Our study had some of the following limitations: Our response rate for recruitment was 32%. The factors impacting on the recruitment rate were

- Additional visit for taking part, time consideration;
- Maybe those who did not respond did not see UHL as a significant factor.

Children talked about their experiences in the presence of their parents. It is possible, that this may be considered a limitation as they may have been inhibited to disclose certain difficulties. In addition, the number of children in the congenital and acquired hearing loss groups may be considered as small.

7. Conclusion

This study highlights many issues faced by children with UHL. We identified six categories from our interviews. Adolescents and children with conductive UHL are keen to normalize their hearing loss and adapt to the resulting difficulties. Hearing difficulties in children with conductive UHL were identified indirectly by exploring the adaptations children made to their daily routine. Our qualitative study was able to identify hidden problems of UHL in children and adolescents such as behaviour problems, association with stress and fatigue through exploring the impact of adaptations on daily living and difficulties with hearing in challenging situations and localisation. This study emphasizes the care needs and responsibilities of parents, teachers and care providers to support these children to reach their full potential and highlights patient-specific outcome categories that are most relevant to children with conductive UHL, that should be developed and reported in effectiveness studies in this patient group.

Conflict of interest: None

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Legend for Tables

Table 1.3. Demographic details of participants.

Table 2.3 Summary of categories and subcategories.

Table 1.3. Demographic details of participants.

Pseudonym	Age (years)	Sex	Type of hearing loss	Duration of hearing loss (age at onset of hearing loss)
Dan	11	M	Congenital	11 years (Birth)
Edward	13	M	Acquired	11 years (2 years)
Ellie	14	F	Congenital	14 years (Birth)
Eva	13	F	Acquired	6 years (7 years)
Gerrard	13	M	Acquired	9 years (4 years)
John	17	F	Acquired	7 years (10 years)

Leo	12	M	Acquired	5 years (7 years)
Liam	11	M	Congenital	11 years (Birth)
Luke	12	M	Congenital	12 years (Birth)
Nathan	13	M	Acquired	5 years (8 years)
Sophie	17	F	Acquired	12 years (5 years)

Table 2.3 Summary of categories and subcategories.

Categories	**Subcategories**
Self-perception of hearing	Normalizing the hearing loss
	Hearing difficulty in certain places or settings
	Hearing difficulty when further away from source of sound
	Hearing difficulty when not facing the speaker
	Localising sound
Adaptations and impact of adaptations	Adaptations
	Impact of adaptations - behaviour issues and fatigue
Support from friends, families and teachers	Support from teachers
	Support from friends and family
Dealing with being different	Dealing with the cosmetic appearance
	Dealing with recurrent ear infections
Concerns about safety	
Implications on future	

Appendix 1.3 The Impact of Unilateral Hear Loss: Topic Guide for Child / Young Person interviews

The interviews will aim to investigate the impact of the child's hearing loss by looking at various aspects such as the effect on education, behaviour and development.

N.B: The interview will take the form of a conversation. Interviewees will have the opportunity to raise issues of importance to them. This topic guide may therefore change as data collection progresses.

At the start of the interview

- Thank the child / young person for agreeing to be interviewed.

- Briefly tell them about the research, why you are doing it and what you are aiming to find out.
- Check it is still OK to record the conversation (even though written consent will have been received). Let them know that a written copy of the interview will be made and that it will not have their name on it. It will be anonymous.
- Reassurance them that anything they tell you will be kept in c anonymous (their name will not appear any of the written copies of the interview or on any of the questionnaires they fill in).
- Ask about any worries about the interview before starting.
- Have they got any questions?
- Make clear it is OK to stop at any point or refuse to answer questions during interview.

Questions

- Lets talk about your daily routine- Weekday and weekend
- What wakes you up in the morning?
- Getting out and about, are there any aspects you do differently?
- Moving around in school and with friends- any preferences?
- Any preference about classroom seating?
- Other activities such as hanging out with friends, music, sports, computer games.
- Have you tried hearing aids, what do you think about them?
- Are there things you like to do but find them difficult?
- What about the future?
- Any questions or concerns?

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Chapter 4: Parental perception of the impact of unilateral conductive hearing loss in their children – A qualitative study

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Abstract

Objective: To understand parental perception of the impact of unilateral conductive hearing loss in their children.

Methods: Semi-structured interviews with parents of eleven children aged 11-17 years, with a known diagnosis of congenital or acquired, unilateral conductive hearing loss. A *qualitative descriptive exploratory approach* was used, with data coded and categorized using NVivo software.

Results: Three categories were identified:

(i) Problems perceived by parents and acceptance.

The main problem reported by parents was related to perceived hearing difficulties and acceptance of resultant limitations. They described difficulties hearing in challenging listening situations and localizing sounds. They also described adaptations that children made to

overcome localizing and listening difficulties. Parents talked about the acceptance of the highlighted hearing difficulties. Most parents described little impact from the conductive UHL on speech & language development, the only concern being their child's inability to modulate their speech loudness.

(ii) Monitoring, advice and support.

Parents described their experience of advice given at the time of the diagnosis of conductive UHL and the role it played in the acceptance of their child's hearing difficulties. They described mixed feelings about the advice given by healthcare providers. Monitoring of hearing was a reassuring exercise and parents were keen for surveillance of their child's hearing levels. They also described the support their child received and their perception of its positive impact on their child's education, although sometimes children found this annoying.

(iii) Implications of active issues and parental concerns.

The main subcategory was *trial of hearing aids*. Parents in the *acquired hearing loss* group were keen for their child to try this intervention, whilst parents in the *congenital hearing loss group* appeared to accept their child's UHL and described the impact as limited, mainly as a result of adaptations. However, parents in both groups described the dislike expressed by children in relation to hearing aids. Parents in the acquired hearing loss group also described their concerns about the risk of ongoing ear infections, reflecting the underlying aetiologies in this group. In addition, parental concerns focused on the *impact on education* and their children's future prospects. Parents in the congenital hearing loss group talked about their worry regarding deterioration in their child's normal hearing ear, as compared to the acquired hearing loss group in whom the anxiety was about deterioration of hearing in both ears. Crossing roads safely was another concern expressed by parents in both groups.

Conclusions: Three interlinked categories were identified: (i) problems perceived by parents and acceptance of hearing loss, (ii) advice, monitoring and support to overcome these problems, and (iii) implications of active issues and parental concerns. This study highlights parental perceptions of impact of unilateral conductive hearing loss on childhood, and acceptance of these problems, that should be considered when evaluating children with conductive unilateral hearing losses. As parents accept their child's hearing loss, they may not perceive it as a health

issue or seek intervention. Considerations of parental perceptions are also important when reporting outcomes of therapeutic interventions. Omitting the perspectives of parents' risks underestimating the impact of the hearing loss or therapeutic interventions. There is an unmet need for a bespoke measurement instrument designed specifically for this patient group and incorporating the perspectives of parents, if evaluation of novel and existing interventions or conservative management strategies is to be clinically meaningful.

1. Introduction

With the introduction of universal newborn hearing screening programs (UHSP), increasing number of infants with hearing loss are being diagnosed early in life [10]. *Congenital* unilateral hearing loss (UHL) is said to affect 0.81/1000 live births, and these children are often diagnosed within 4-5 weeks of birth [32]. Unilateral hearing loss has been defined as average air conduction thresholds at frequencies 0.5, 1 and 2 KHz of ≥ 20 dB HL in the impaired ear and ≤15 dB HL in the better hearing ear [7]. Congenital UHL accounts for 42% to 47% of all children in this group [12, 13]. However, there exists an ongoing dilemma regarding the management of infants identified early in life [11]. The guideline for assessment and management of infants identified by the Newborn Hearing Screening Program (NHSP, UK) recommends, "monitoring and review of babies with unilateral hearing loss as current evidence does not support early intervention". In addition, there exist progressive and acquired forms of UHL, with the prevalence of UHL in children being reported to range from 3% to 6.3% in children aged 6-19 years[14].

Although children with a diagnosis of UHL are now detected early, our understanding of the impact of UHL is not entirely clear. Multiple studies have investigated the impact of UHL in childhood focusing on different outcome domains. A consistent negative impact of UHL on hearing in noise [33, 34, 36-39, 54] and localising ability [33, 34, 55] is described. However, evidence on other outcome domains is not so clear. Some studies report no evidence of negative impact on speech & language development [46, 64-66], education [67] and cognition [46, 58, 68, 69], while others describe a negative impact on speech & language [39, 69, 70], cognition [39, 40, 71], education [40, 71] and behavior [57]. There are also reports that these negative effects persist for a long time [43]. Impact on quality of life (QoL) has also been reported in some studies[47, 48]. Borton et al [47] describe parental perceptions of the impact

on health related quality of life (HRQOL) of their child's congenital UHL . The qualitative arm of their study comprised a focus group with participation from four parents, who expressed concerns about difficulties in school and misunderstanding conversations.

Many of these studies do not clearly describe their participant group, including the type of unilateral hearing loss (congenital/acquired), severity (mild/moderate/severe), or the nature of hearing loss (congenital/acquired). The resultant uncertainty in evidence to support best and individualised practice is likely to cause concern for parents and clinicians. To address this unmet need there is a requirement to explore the impact of the differing types, onset and severities of UHL.

Conductive UHL in children (congenital or acquired) by its nature is typically mild to moderate in severity, and often associated with normal underlying cochlea function. A normally functioning cochlea means that it can be possible to restore their *natural hearing*. Therapeutic interventions may be non-surgical (hearing aids) or surgical (bone conduction implants utilising the normally functioning cochlea, or ossicular reconstruction), with the chosen management strategy influenced by the nature of the difficulties experienced by the child and parental acceptance of surgical risk. In our companion paper, reporting on the perspectives of children regarding the impact of unilateral CHL on their lives, we found that children normalize their hearing loss and tend to adapt to the resulting difficulties. Hidden problems of UHL in children and adolescents were identified through exploring the child's daily routine. However, parents have direct knowledge and experience of the impact on their child (e.g. through daily observation in different situations and/or comparison to siblings), such that failing to incorporate parental perceptions into therapeutic decisions and evaluations risks omitting important negative consequences of the hearing loss or misinterpreting actual benefit from the chose therapeutic intervention. Greater understanding of the experiences of parents of children with conductive UHL will help to identify and structure appropriate services to support children to reach their educational and social potential, irrespective of whether *non-surgical*, *surgical* or *no treatment* management strategies are chosen.

2. Aim

The aim of our study was to understand the impact perceived by parents of their child's unilateral conductive hearing loss.

3. Method

A qualitative study was used to understand the parental perception of the impact of conductive UHL in their children. We adopted a descriptive exploratory approach guided by *grounded theory methodology* including use of purposive sampling, semi-structured interviews, coding and constant comparative analysis (31, 32). We used this approach in order to understand the issues parents felt were important and to encourage parents to describe their perceptions. We used a topic guide that was adapted as new categories developed, as is standard practice in qualitative research methodology.

3.1 Participants

National Research and Ethics Committee (NREC) approval and Health Research Authority (HRA) approval was obtained prior to recruitment (REC reference 16/LO/1080). Written informed consent was obtained from the parent of the child, prior to performing any study-related procedures.

Participants for this study were recruited from a tertiary Children's Hospital. Our centre receives referrals for assessment and management of children with UHL, has a multidisciplinary clinic for management of children with microtia, and manages children with chronic otitis media (COM). Potential participants were identified from our database containing the records of children with conductive hearing loss and we contacted parents by letter. We chose to study parents of children from 11 years of age as the education system in the UK is structured such that this age represents a significant developmental stage, with transition from primary- to secondary-level education that is characterized by a more demanding listening and educational environment. Additionally, this is a life-stage when children are beginning to seek and gain independence, in terms of using public transport and travelling to school, making it a milestone for parents in relinquishing some of their control over the safety of their children.

3.1.1 Participant selection criteria

Inclusion Criteria:

1. Parents of children aged 11 to 17 years with

2. Conductive hearing loss with normal bone conduction threshold and air conduction thresholds worse than 25 dB HL (as an average of measured thresholds at 0.5, 1, 2 and 4 kHz) in the worse hearing ear.

3. Normal hearing in the contralateral other ear (Hearing thresholds ≤ 25 dB HL)

4. English speaking parent

5. Congenital and acquired unilateral CHL, with purposive sampling to ensure both groups represented.

Exclusion Criteria:

1. Children with learning difficulties as reported by parents
2. Dual sensory deficit, confirmed by parental report of visual impairment.
3. Families with difficult social circumstances identified by the clinical staff (e.g. recent bereavement in family, child in social care/living separately to parents).

3.2. Data collection - semi-structured interviews

Semi-structured interviews were conducted at the parents' choice of venue, either in an interview room at the hospital or in the participant's home. Only one parent chose to have their interview at their own home. Two interviewers conducted the interviews, the first author and a research audiologist. The first author is a clinician and involved in the clinical care of many of the majority of these children with conductive UHL, meaning that the research audiologists conducted most (7/11) interviews. The interviews lasted approximately 30 minutes. We used a topic guide. (Appendix 1.4- topic guide) that contained a set of open-ended questions, which were used as prompts for the interviews. This topic guide was adapted during the study to explore emerging categories and perspectives. Interviews with parents were conducted in the presence of their child with the conductive UHL. Children were interviewed following the parental interviews and the findings of these interviews are reported in a companion paper. (The perspectives of children on the impact of unilateral conductive hearing loss on their lives: A qualitative study). The interviews were audio recorded, transcribed, and anonymised. Data analysis was concurrent with data collection in order to respond to emergent categories. Data analysis occurred in three stages: i. in-vivo coding, where we coded sentiments described by parents using their own words, ii. Secondary codes were formed from amalgamation of similar codes, and iii. Meaningful categories emerged as we linked the secondary codes. A

computerised qualitative data analysis software, NVIVO for Mac© (http://www.qsrinternational.com) was used. Names in the quotations are pseudonyms to maintain anonymity.

4. Results

34 parents of children with unilateral conductive hearing loss were identified and contacted by letter. 11 parents (32% participation rate) completed the interview (14 participants agreed to take part in the study, but three parents were unable to attend their appointment due to unforeseen circumstances and did not want to rearrange the appointment). Those declining to participate in the study either said that their child did not have a hearing problem, time constraints prevented participation, or they were not contactable by phone. Participant characteristics are presented in Table 1. The categories that emerged are presented with illustrative quotations from participants.

We identified three interlinked categories, namely; (i) problems perceived by parents and their acceptance, (ii) support, advice and monitoring of their child's conductive UHL, and (iii) implications of hearing issues and concerns. These categories and the related subcategories are summarised in Table 2.

4.1 Problems perceived by parents and their acceptance

Parents described the problems they noticed in their child, that they attributed to their child's conductive UHL, constituting a proxy perspective. We identified two main subcategories, namely *perceived hearing problems* and *speech & language issues*, with problems directly linked to hearing loss considered the most significant. .

4.1.1 Perceived hearing problems and acceptance

Parents were able to describe the problems that their child experienced as a result of their conductive UHL. They described hearing problems in certain places (e.g. noisy rooms), certain situations, (e.g. multiple talkers) and difficulties when the normally hearing ear was covered or away from the source of sound. We merged these difficulties into a subcategory of *hearing in challenging situations*. Other subcategories were *localisation of sounds* and *adaptations to the hearing loss*. Another identified subcategory was *acceptance* of their child's hearing loss. In contrast to the parental perception of hearing problems and acceptance, our companion paper

on children perspectives describes how children normalise their conductive UHL, with hidden problems only emerging through exploration of their daily routine.

Parents of children in both the congenital and acquired conductive UHL groups described their children struggling to hear if they were not facing the speaker, when there were multiple speakers involved in a conversation, or in the presence of competing television noise. They also described their child struggling if they spoke to their child whilst being on the side of the poorer hearing ear. Parents talked about having to raise their voice to be heard. Similarly, difficulty was encountered when parents spoke to their children from another room. Parents described their frustration at having to go to their child when they needed their child's attention or signposting them to where they were. Parents in both the congenital and acquired hearing loss groups raised concerns regarding these difficulties, but parents of children with a congenital loss described these hearing issues as less of a problem. Most parents describe difficulties in day-to-day life due to their hearing loss

> "Yeah, it's just if…you know, if we're all sat in like a semicircle round the telly and Gerard's in the middle, mum, you know, sits in a chair that side, she says something, he doesn't always pick it up, you know, and then like, Gerard. Gerard. What, you know, and then ask you again" (Gerrard's dad)

> "If you sort of…if you're in the picture, so if you're sitting watching a television programme, if I was sitting on Liam's left and I wanted to say something to him, then there's a possibility he's not going to hear you, because (a) he's concentrating on what's on the television, (b) there's background noise, and (c) I'm sitting on his left hand side. Whereas if you were sitting on his right, the probability is that he would hear what you are saying." (Liam's mum)

> "Thinking back to the classroom situation, I think it does…if there's lots of background noise, I think he sometimes says it can be a bit of a struggle, but, again, just seems to get on and…" (Luke's dad)

> “I suppose even now sometimes you can tell that he can't hear what you're saying but I don't know whether it's just certain times or maybe where he is in the room or if something else is going on.” (Leo’s mum)

Parents described their child having difficulty in localising sound that posed practical challenges for them and their children. Most families found ways of dealing with this, for example, by signposting their children to where they were. Most families spoke about their concern with sound localisation, particularly in the context of crossing roads. They mention that they remind their children regularly about the need to look repeatedly to locate sound sources.

> “If I’m in a different room and Dan is in a different room, if I was to call his name, would he know where I was? Well, he doesn’t. He tends to look round every room until he actually finds me. I think he does struggle to locate where sounds come from.” (Dan’s mum)

> “Obviously we’ve taught her again from a young age that she has to look that bit more, than rely on just the listening because obviously traffic comes both ways.” (Ellie’s mum)

Parents talked about the *adaptations* made by them, their child, friends and families and school, to overcome some of the hearing difficulties. Parents described how their child turns their head slightly so that sound is directed towards their normal hearing ear with parents choosing to position themselves on the side of their child’s better hearing ear. They used the phone more often, for example text messaging their child, even when they were in the same room, as they talked about this measure overcoming the need to get their attention and also not having to worry about repeating themselves. In addition, families made significant changes to their daily lives to adapt to their child’s hearing loss, including changing job arrangements to ensure school pick-up, changing holiday plans to avoid water activities (in the acquired hearing loss group). Parents said that through these adaptations children were able to overcome most of the hearing difficulties helping them cope with their hearing loss.

> “I’d say, he’s probably with friends. Is that right, Dan, when you’re crossing the road? Yeah, but I do worry actually and I’ve even been looking at part-time jobs, just to try and adapt to that time when Dan’s coming home. (Dan’s mum)

> “But she’s done it despite her hearing. So perhaps her hearing impairment is not so significant, and maybe she’s been able to tiptoe around it, and hide it, by just making sure she’s not necessarily on the front, but maybe she’s on the right side. If she can’t get to the front she’s on the right side. And when she’s at the back, she just has to lip read better.”(.Sophie’s mum)

> “It is, but not to the extent that I would think it would be, but that’s because I think Liam has learnt to adapt, and Liam has learnt to cope with it. If Liam is talking to you and having a conversation with you, he will slightly turn his head so that his right ear is facing you, more so he can pick up what you are saying more. That’s something that I notice. (Liam’s mum)

Although parents described hearing difficulties, they appeared to have accepted their child’s hearing loss and the need for adaptations in everyday life. They talked about how many of these changes had become a part of their daily routines. Parents described how the changes they had made helped their children live with hearing limitations and they talked about everything being ‘normal’. The process of becoming aware of the problems and learning to deal with and adapt to the problems became a way of overcoming the difficulties. They did not see these difficulties as health issues, but rather as another challenge parents face as a part of their child growing-up. Parents talked about the hearing loss being a limitation that needed to be identified and dealt with, rather than a specific problem. Parents in both the congenital and acquired groups echoed these sentiments. Their acceptance of the limitations of their child’s hearing and the need for adjustments has not been reported in previous studies. The qualitative methods that we used enabled exploration of the meaning of experiences to participants,

“I think we have been compensating for so long. It’s sort of a second nature now to say something and if there is no response just to say it again louder. And then if there is still no response, do it a third time even louder until he does hear it, and to us it’s normal to do that. It’s not abnormal, that has become normal. And I don’t think we even think about it anymore.” (Edward’s mum)

“I think Dan’s very good at coping with, living with hearing from one ear. I think probably a lot of children are very similar or there’s a lot of people, in general, that to Dan, it’s almost normal, he probably doesn’t even think that there is any issue because he’s not necessarily aware of what it would be like because he was born that way. I mean perhaps it would be completely different if he’d lost his hearing at this age, for example. So, I think, for Dan hearing, as he does, it’s completely normal.” (Dan’s mum)

“But I think it’s just become normal that that, like you say, sometimes she doesn’t and you just either repeat yourself or, you know...it’s just what we do. It’s not very often; we’re not sat repeating ourselves a lot. We just have a normal relationship but there’s just sometimes where the telly is a bit loud or there’s a film with lots of music and different things on that it might, but other than that, no.” (Elli’s mum)

“No, it's just mainly as long as people know, you know, he can't hear on one side and once they get to know he can't they can adapt, put him in the right place to hear so his learning’s better. He's learning himself to listen out more, you know, by moving his head around and to watch, to look more and it's just, you know, as time goes on you do…you know, if he gets better at understanding what his problem is and how…you know, how to react to it.” (Gerrard’s dad)

“We don’t make a conscious effort to do something…we don’t make a conscious effort to, well we don’t feel as if we need to make a conscious effort to accommodate his not having hearing on the left side, because it’s something that if he hasn’t heard, it’s not the end of the world because I’ll just touch his leg, and he’ll look…you get his attention.

And it's not…as his mum, speaking from my perspective, I don't think it's something to be made a huge issue out of." (Liam's mum)

"So, from a personal perspective, I just think people are just a bit, it's an era of crash helmets on bicycles, and elbow pads on skateboards, and I think that actually, people can take quite a lot of responsibility for themselves, and so on, and work to their own limitations." (Sophie's dad)

4.1.2 Impact on speech & language development

Parents of children with conductive UHL generally felt that the effect on speech & language development was not significant. In the acquired hearing loss group, most children acquired their hearing loss over the age of 4 years (except for one patient), with the 3-4 years of age period being considered a critical age for speech & language development.

A parent of one child in the congenital hearing loss group and one parent in the acquired hearing loss group raised some concern about pronunciation of words. The child in the acquired hearing loss group developed a hearing loss at 2 years of age and there were subsequent concerns regarding his speech. Despite both children having received help from a speech therapist at a younger age (primary school age, 5-10 years of age), their parents still felt that some of these issues persisted into adolescence.

"Certain sounds he still can't pronounce very well. He still sometimes says letters incorrectly or something. So in that respect I think that was a little to do with his hearing." (Dan's mum)

"More long term, he had to have speech therapy for many years, as a child. I suppose, it's just a process, isn't it. He can't hear sounds; therefore, he can't pronounce sounds, so he had that input." (Edward's mum)

Parents talked about loudness and clarity of speech in both groups. Some parents attributed loud speech to not hearing their own voice well and hence not being able to control the volume. Two parents (one in each group) noticed some issues with clarity of speech and thought this may be due to the hearing loss in one ear.

> “She is so loud, we tell her to lower her voice, she says, she can’t help it. The only time, she was quiet was when she tried the hearing aid for a week. (Eva’s mum)

> “Whether he can hear himself louder than we can. A lot of the time we have to say, you’re mumbling we can’t hear what you’re saying, say it again. Haven’t we? That’s been over the last two or three years, I think, hasn’t it? (Nathan’s mum)

> “I think sometimes…I wonder if sometimes he mumbles a little bit more actually now I think about it and that’s one thing, but, again, yeah, sometimes he doesn’t speak quite as clearly as his brothers do.” (Luke’s dad)

No comments were made about language development in either of the groups. This may be as children recruited into the study were aged 11-17, when parental concerns regarding language development may have resolved as their children acquired language in-line with their peers. As we explored this aspect in older children, and asked parents to recollect any impact on language development, they said their child’s language was not something they had worried about.

> “We have never had any issues with her speech. She started talking very early and always has been chatty, No, we haven’t had any concerns regarding her language development” (Eva’s mum)

> “And we are not a Welsh speaking family, but we are Welsh but it’s something that we felt was really, really important, and that was one of the things that we thought about a little bit longer when Liam was going to school, because the hearing side of things, they’re taught totally in the medium of Welsh. So that’s one of the things that we did sort of wonder, whether there would be a problem there, but it’s never been a problem.” (Liam’s mum)

4.2 Monitoring, support and advice

Another category we identified was the impact of advice given about the diagnosis and impact of their child's UHL, monitoring of their child's conductive UHL and support received through various services to overcome difficulties associated with the UHL.

Parents described the *advice* given at the time of diagnosis of their child's conductive UHL. Parents of children with congenital hearing loss (of children with a congenital abnormality of the outer ear) described being told, that their child had no hearing at all in one ear. They also described being told that as their child had normal hearing in one ear, it would have little or no impact on what their child could do. Parents described this advice as reassuring and appear to have been instrumental in the acceptance of their child's UHL. There were mixed feelings about advice given by healthcare providers regarding the management of their child's hearing loss. Some parents described that they would have preferred to try an intervention if there was any benefit at all for their child. Many parents were glad they had a choice and an intervention was not essential, although some parents found this choice difficult, as they felt pressurised to make a decision about their child. Parents felt it was the responsibility of healthcare providers to make decisions rather than offer choices.

> "We had a follow up appointment with an ENT consultant when he was six weeks old and at that point, we were told that he would never have hearing on that side, and that's how it would always be, and there would never be an improvement. (Liam's mum)

> "He has this condition which obviously impacts on his hearing. It's only on one side. So, that has been seen birth. When he was born I was told that it shouldn't cause any problems throughout his life because he can ear sufficiently with one." (Dan's mum)

> " At one stage we were advised about our options, but it wasn't advice as such it was, that's one of the options, what do you think. And that is the kind of approach that I struggle with. I am not a specialist in ENT. So, although I understand why doctors do that, I also disagree that that's the way to be presented to a parent when a parent is not a specialist in the area." (Edward's mum)

Most parents approved of *monitoring* of their child's hearing. Parents of children with congenital hearing loss were aware of the need to monitor the normal hearing ear and parents of children with the acquired hearing loss were more focused about new or on-going problems with the affected ear. Most parents were glad they were being monitored. However, some parents were not keen on regular assessment, as they did not find this particularly helpful.

> "Since birth and every year, practically, sometimes more often he's been for hearing tests. It's been really good; we've always kept a close eye on his hearing levels" (Dan's mum)

> "No, no, and that's what we were told, that we always had to look out for if he had any sort of discomfort in that right side, then jump on it straight away basically, to sort of make sure…because obviously that was his only source of hearing." (Liam's mum)

Parents were reassured by involvement of audiology services and additional *support* arranged at school, as they believed that schools were more likely to act upon information and guidance from healthcare professionals. Visits to school by the teachers of the hearing impaired, helped identify the best seating position for their child. Parents said that this support was easier to obtain and implement in primary school, as compared to secondary school (11 years and above). They also said that there were occasions when their children found this support annoying, particularly when they could not sit with their friends, or they did not want to "make a big deal of their hearing loss"

> "Yes, yeah. We had a couple of issues with the junior school where the teacher wasn't aware of this even though they had been informed and therefore was at the back of the class, and things like this, and I think he had: We wonder whether actually, thinking back, whether some of his grades weren't quite…he didn't progress quite as well as he was expected to, his development was below what they expected and we wondered whether it was partly that. So it may have been an element of the hearing, but also we put a bit more effort in after school, and things, which we hadn't probably been doing and then things started to come back up where we expected, so I think it was maybes a

combination of hearing, but also actually just hard work, putting the work in."(Luke's dad)

"They have a lady who goes and speaks to the school to check that they've got the right things in place, i.e. where's he sat in class? Is the teacher aware that he has hearing only on one side? And there's a few different things that they asked them to put in place, so he has to be near the front of the class, which annoys Luke, I think." (Luke's Dad)

""Since he's gone to high school, classes have got bigger again, but they all know to keep him at the front and to keep him to the left hand side of the class," (Gerard's dad)

"They were more accommodating with it, and you had somebody from the social that came in and discussed with his teachers, assessed how well he was hearing while he was in school, so they did adapt quite a lot for you. I think at least twice a year wasn't it? They tried to come in once a term just to see how it had changed, and if he were in a different classroom, they would speak to the teacher and change things accordingly. But, at secondary school it just seems to get lost a little bit." (Nathan's mum)

4.3. Implications of active issues and parental concerns

Parents described concerns regarding ongoing issues, in particular recurrent or prolonged infections in the acquired hearing loss group, cosmetic appearance of the ear in the congenital hearing loss group and the trial of hearing aids. Although parents were keen for their child to try hearing aids, they were aware that the hearing aids would attract more attention to their child's hearing loss. The presence of foul smelling otorrhea discharging from the ear canal also drew attention to their child's condition.

In both the congenital and acquired UHL groups, parents talk about active issues, their impact and the need for monitoring. This was a particular concern in the acquired hearing loss group for many of the parents, although not all. They said that the presence of foul-smelling discharge caused by the infection has a negative impact on their child's self-esteem. Parents restricted certain activities (e.g. swimming) to avoid contact with water, as they were advised by health

care providers that this measure reduced the risk of infections. Ongoing infections also raised anxiety and affected their child's mood. Parents in the congenital hearing loss group had accepted the need for monitoring of the normal hearing ear, but did not describe any concerns about infections.

> "It's just difficult sometimes, because he's had so many infections as well, he doesn't always tell us. 'Cause I don't think you identify it all the time. So, that becomes difficult. So, we're trying to keep on top of it, aren't we?" (Nathan's mum)

> "She had a lot of discharge from her ears, and you always worried if this would settle down. We have had to keep her ear dry, but with her long hair, that's easier said than done." (Eva's mum)

Parents in the congenital hearing loss group described the impact of the cosmetic appearance of the ear. They saw their child as a confident young person needing some support through their journey in managing the ear malformation. Parents talked about their own feelings about the appearance, but also about letting their child make decisions relating to any surgery to change the cosmetic appearance. They were keen to support their child's role in the decision making process.

> "Dan wears his hair quite long, whether that changes as he gets older, he might just say, actually, yeah, and probably as the operation on his ear settles and the appearance actually looks even better then he might just be ready then to say, okay I'll try going a bit shorter each time, maybe. (Dan's mum).

> "She was already at secondary school 'cause as a parent I just thought that it would be a good time to have it done going to secondary school but she was still quite happy and obviously we never pushed the decision, it was hers all along and obviously she had a few different options to take. (Ellie's mum)

Parents in the acquired hearing loss group verbalised the benefits and desire to *try* hearing aids to overcome the conductive UHL more than the congenital group. Parents in the congenital hearing loss group talked about how they were told at a very early age that their child had no

functional hearing in one ear, and described how they had accepted it, particularly as they saw their child adapting well to the hearing loss. However, parents in the acquired hearing loss group were keen to explore options such as hearing aids and even further surgery, as the acquired hearing loss was a new condition that parents were dealing with. Parents in both groups whose children had tried hearing aids described benefits such as, speech being quieter, being more attentive in school and generally better hearing. However, they also said that their children did not like the hearing aid for various reasons, including "new sensation", "other people's opinions".

> "I think, it was useful in the sense of it just gave him that, what he'd always been missing, in my opinion, the ability to hear as well as myself.
> He just didn't like the sensation it would give him. I don't know if it was to do with the band or just the tightness of the device next to his skin." (Dan's mum)

> "Eva is so loud, everyone in the house also speaks loudly so that Eva can hear. But when she tried the hearing aid at home for a week, it was amazing how quite the house was." (Eva's mum)

Parental concern was an important category that became apparent as interviews progressed. Some parents worried about the impact on education and future, whilst others were anxious about deterioration in their child's hearing. Worry about safety on roads was something that all parents described. They were worried that there were times when their child was not hearing as well as they could, as the classroom may have been noisy, their child was too far away from the teacher, or may have missed hearing part of instructions from the teacher. They said this resulted in their child falling behind their peers. Parents talked about having to miss lessons to attend hospital appointments or whilst recovering from surgery, which had a negative impact on their child's education. Parents helped their children by arranging additional help at home to overcome this negative impact. Some schools were also able to support these children with additional lessons to improve their academic performance. Parents said that many of these measures had been helpful and welcome. In addition to their academic performance, parents talked about how their child's UHL was affecting their child's concentration and behaviour.

Parents said that the *effort of listening* was sometimes significant and their child was exhausted at the end of the day.

> "He is not hearing what he is being told therefore you feel that he is getting into trouble with what they consider to be inappropriate behaviour." (Edward's mum)

> "I think with the hearing not being on one side, he gets tired easier and then perhaps sometimes his behaviour will occasionally tail off, but that could also be teenager sat in a class as well." (Luke's dad)

> "Because unless he can see you, he doesn't hear you, more often than not do you? Which causes a bit of a problem at school, 'cause a lot of the teachers think he's not paying attention, even though we have had people go into school and explain to them that he needs to sit near the front, and be able to see people's faces when they're talking. But it has caused a few problems, you've had a few detentions over it, teachers not thinking you're paying attention, haven't you?" (Nathan's mum)

Parents in the acquired hearing loss group worried more about the future than those in the congenital hearing loss group. The acquired hearing loss, accompanying infections, and the need for treatment was a new experience that parents had to take into consideration when they talked about the future. Parents of children with congenital hearing loss knew from infancy that their child did not have 'normal' hearing in one ear but tended to describe their children growing up to do most things similar to their siblings. They were able to accept this hearing loss and although it had an impact on their child, they did not describe this as something that would cause long-term problems.

> "But even like being in a lecture theatre, how is that going to affect him? Is he going to be able to hear? Will he be sat at the front? So that bothers us a little bit, because obviously he was really good at school, and then it had a bit of a knock-on-effect these last few years, and we don't want it to continue to have that knock-on-effect." (Nathan's mum)

"I feel even worse and guilty when it comes to Edward because he now wants to join the army. And I am not sure whether he can, and that has always been his wish to become a Royal Engineer. So, I do not know how we are going to go around that. I am still hoping that there will be a way around that, by means of plastic surgery somewhere; somehow, even if we have to go to America, we will find a way, even if we have to sell our house. That's his dream so; to me that is what my role is as a parent. (Edward's mum)

"No, not that specific worry about future, just the normal worries that you have with your kids, isn't it, really, so, yeah." (Luke's dad)

" I don't think there will be any issues with what he wants to do when he is older, but maybe I think that's partly because Liam will do whatever he wants to. If he sets his mind to do something, he will do it. If whatever he chooses to do, if the hearing situation is a possible difficulty, then I know he will find a way to overcome that. (Liam's mum)

Parents worried about deterioration in hearing over time. In the congenital hearing loss group, parents were anxious about deterioration of hearing in the normal hearing ear as a result of age-related hearing loss or due to exposure to loud noises. Whereas parents of children with acquired hearing loss, describe their concern about on-going deterioration of their child's hearing in the affected ear, as well as worrying about the normal hearing ear.

"Yeah. Well obviously we knew from the moment he was born, they showed us that he didn't have an ear on this side, so they knew that obviously there would be an impact. I guess my concern is as he gets older is, as the hearing goes on this side, that's the concern isn't it? Because I guess we have both sides for it to gradually deteriorate, but Leo unfortunately only has it on one side to deteriorate." (Leo's dad)

"When me and my husband have discussed it, it's little things, like, if it continues to get worse, when he has children of his own, is he going to be able to hear them if they need him?" (Nathan's mum)

> “Well, obviously it would be worse if we got damage in the other ear, ‘cause she’s doing it mainly with one ear now. So, I’d like to confirm that the other ear’s good.” (Sophie’s dad)

> “ We’ve tried to steer him away from being a drummer, and he’s very good, I have to say, he’s a very, very good drummer. And we’ve tried to steer him away from that because of the loud noise issues, but not very successfully. And we worry about him having no hearing at all.” (Liam’s mum)

Parents in both the congenital and acquired hearing loss groups worried about the safety of their child, particularly when crossing roads. They said they were cautious about risks to safety for all of their children but were particularly wary about their child with the UHL. Some parents managed this by educating their child, whereas others chose to avoid exposing their child to situations where they may be exposed to crossing roads independently, for as long as possible.

> “It's just that when he was out among traffic we're always wary, well has he heard a car, has he heard that? If he goes out on his bike, well have you heard anything coming? You know, when he's out at Scouts, one thing and another, we keep telling him, be careful, and all that sort of thing because, you know, half your hearing’s not there and it's…you're only hearing basically from one side, aren't you?” (Gerrard’s dad)

> “So, it is being mindful of everything, and safety is one of them, it’s silly things like it’s only the last few months that I’ve let him go round to the shop on his own, because he’s got to cross the road, and I’m worried that if he doesn’t hear something, and it’s quite a busy road. “ (Nathan’s mum)

> “So I just reiterated to him, remember, you can't hear in one ear, you must take notice of the crossing, and I'm just conscious that if he can't hear traffic coming if he's out and about that you need to be really sensible about it.” (Leo’s mum)

5. Discussion

Parents described hearing difficulties in challenging situations, difficulty localizing sound and adaptations children and families make to overcome these difficulties. A significant finding relating to hearing problems was the parental acceptance of their child's conductive UHL. This is in contrast to our companion study, which investigated the children's perspective of the impact of their conductive UHL. Children normalized their UHL and difficulties were hidden. These hidden difficulties only became apparent as we explored the child's daily routine.

Whilst all parents were able to identify the hearing difficulties their child faced, only a few parents explored interventions to treat their child's conductive UHL. The following reasons could explain why parents did not always pursue treatment options: i. parents focused on managing other issues such as the infections (in the acquired hearing loss group) and the cosmetic appearance (in the congenital hearing loss group), ii. adaptations made by the children, their families and at school were helpful in overcoming the difficulties children with UHL encountered, and iii. parental acceptance of their child's UHL, as we see parents starting to normalize their child's UHL. Advice at the time of the diagnosis in the congenital hearing loss group is likely to have been, "monitoring and review of babies with unilateral hearing loss as current evidence does not support early intervention" [11]. This advice in conjunction with the adaptations children and families make to overcome the impact of conductive UHL contributes to acceptance of the UHL and minimizes parental desire to seek intervention. The finding of acceptance is important when discussing management of UHL with parents of children with a conductive UHL. As parents observe their child adapting to their hearing loss and accept the imposed limitations, they see this as "the New Normal"[72]. Although this can be a positive attribute, in the absence of an effective intervention it could prevent an effective intervention becoming available to these families.

Available literature now highlights the potential negative impact of UHL upon several aspects of childhood development [34, 37-40, 70, 71]. However, evidence supporting intervention is still limited [73-77], which may be attributed to the lack of clinically meaningful outcome measurement instruments (OMI) specifically designed for conductive UHL, that either objectively capture the most important clinical consequences, or are validated as subjective tools to be used in parents by proxy. Clinically relevant OMIs are becoming available, such as

tests designed to measure difficulty hearing in challenging situations (e.g. hearing in noise test - children (HINT-C), speech in quiet and noise and sound localization tests), but as yet they are not used as standard practice. *Generic questionnaires* (e.g. Glasgow benefit inventory[76], PedsQL[47]) and *specific questionnaires* (e.g. SSQ-Child/Parent/Teacher[29]) have limitations as they do not fully identify the impact of UHL in children, particularly as children may have adapted to overcome many difficulties associated with UHL. This hinders the detection of the *hidden impact of UH*L. In our study, parents describe difficulties hearing in certain challenging situations and adaptations made to overcome the negative impact of UHL; valuable information when developing bespoke patient-specific outcome measurement instruments for this group.

Another important finding from our study was the concern and anxiety described by parents of children in the *acquired hearing loss* group regarding the long-term effect of their child's UHL, as compared to the apparent acceptance of the reassurance and advice given to parents of children in the *congenital hearing loss* group (monitoring and support). Parents in the acquired hearing loss group were accepting of the current impact of their child's conductive UHL but were keen to explore all available therapeutic options to improve their child's future hearing performance, perhaps reflecting a feeling of parental responsibility, or guilt, due to the hearing loss not being present at birth and as such being perceived as potentially preventable. The *Regret Theory* proposed by Loomes and Sugden [78] suggests that when making decisions, parents will anticipate risks of doing something and compare these to the risk of not doing anything. In our study, as parents began exploring the impact of UHL, they were keen for their child to try an intervention even when strong evidence of benefit was lacking or limited, with this finding supported in the literature [79, 80] and considered to reflect the need to do everything possible to ensure long-term improvements and prevent future problems.

Our study also highlighted the importance parents placed on support from healthcare providers to optimize educational outcomes, such as additional support in classroom and advising on ideal positioning in the classroom. Noh and Park [54] from their research into speech discrimination in noise, report that students with UHL should be no further than 6.27m away from a teacher in a noisy classroom if they are to achieve comparable speech discrimination scores to normal hearing peers who are not preferentially seated. Measures adapted in school

and greater understanding of the potential consequences of conductive UHL upon learning, will negate some of the difficulties.

6. Strengths and Limitations

In depth interviews with parents to comprehensively understand parental perception regarding the impact of hearing loss on childhood development is the main strength of our study. Likewise, we chose to focus on an under-studied group of children with UHL, in whom historically the consequences of hearing loss were not given priority. Our use of multi-disciplinary analysis was instrumental in understanding the issues that parents described.

Our study had the following limitations, i. response rate for recruitment was only 32%, perhaps reflecting reluctance to attend for a formal interview, avoidance of discussing a distressing problem, or a belief that their child's hearing loss was not a significant problem, ii. parents may have been reluctant to disclose, or downplayed, certain difficulties or anxieties in order to not cause their child distress, and iii. the numbers of children in the respective groups were relatively small, partly negated by the depth of enquiry.

7. Conclusion

Our study highlighted three main categories interlinked with each other: i. problems perceived by parents and their acceptance, ii. advice, monitoring and support to overcome these problems, and iii. parental concerns and implications of the hearing loss. Parents did not report significant difficulties in speech & language development. The main problems identified were hearing in challenging situations and necessary lifestyle adaptations made by the child, with parental acceptance of these problems being a significant finding. This report highlights the need for bespoke outcome measurement instruments (OMI) to quantify the true extent of hearing problems in children with conductive unilateral hearing loss, to enable meaningful assessment of need and effectiveness of therapeutic interventions.

Conflict of interest: None

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Legend for Table

Table 1.4 Demographic details of participants.

Table 2.4 Summary of main categories, categories and subcategories.

Table 1.4 Demographic details of participants.

Pseudonym	**Age (years)**	**Sex**	**Type of hearing loss**	**Duration of hearing loss (age at onset of hearing loss)**
Dan	11	M	Congenital	11 years (Birth)
Edward	13	M	Acquired	11 years (2 years)
Ellie	14	F	Congenital	14 years (Birth)
Eva	13	F	Acquired	6 years (7 years)
Gerrard	13	M	Acquired	9 years (4 years)
John	17	F	Acquired	7 years (10 years)
Leo	12	M	Acquired	5 years (7 years)
Liam	11	M	Congenital	11 years (Birth)
Luke	12	M	Congenital	12 years (Birth)
Nathan	13	M	Acquired	5 years (8 years)
Sophie	17	F	Acquired	12 years (5 years)

Table 2.4 Summary of main categories, categories and subcategories.

Categories	**Subcategories**
Problems perceived by parents and their acceptance	Perceived hearing problems and the acceptance
	Impact on speech & language development
Advice, monitoring and support	
Implications of active issues and parental concerns	

Appendix 1.4 The Impact of Unilateral Hear loss: Topic Guide for Parent interviews

The interviews will aim to investigate the impact of the child's hearing loss by looked at various aspects such as the effect on education, behaviour and development.

N.B: The interview will take the form of a conversation. Interviewees will have the opportunity to raise issues of importance to them. This topic guide may therefore change as data collection progresses.

At the start of the interview

- Thank the parent(s) for agreeing to be interviewed
- Brief outline of the purpose of the research
- Reassurance about anonymity, confidentiality, and non-impact on service delivery
- Check it is still OK to record the conversation (even though written consent will have been received)
- Ask about any concerns before starting
- Have they got any questions?
- Make clear it is OK to stop at any point or refuse to answer questions during interview

Questions

- Let's talk about their daily routine- Weekday and weekend
- Interaction within the family
- What wakes them up in the morning?
- Getting out and about, are there any aspects they do differently?
- Moving around in school and with friends- Any preferences?
- Any preference about classroom seating?
- Other activities such as hanging out with friends, music, sports, computer games
- Have they tried hearing aids, what do they think about them?
- Are there things they like to do but find them difficult?
- What about the future?
- Any questions or concerns?

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Chapter 5 Discussion

Current limitations in the available evidence

The aim of our study was to evaluate the impact of unilateral hearing loss (UHL) in children, more specifically conductive UHL. The systematic review investigated and evaluated the evidence available in the literature regarding the consequences of UHL in children. We found several limitations in the identified literature:

- Failure to distinguish between different types of hearing loss (congenital/ acquired/ sensorineural/conductive).
- Failure to report the severity of the UHL (mild/moderate/severe).
- Failure to adequately differentiate between unilateral and mild bilateral hearing loss.
- Combining, and not differentiating between, children and adults.
- Heterogeneity in outcome domains evaluated.
- Heterogeneity in outcome measurement instruments used to evaluate a particular outcome domain.
- Absence of quality of life questionnaires specific to hearing impairment in children or UHL.

Not withstanding these limitations, it is clear from the available literature that UHL has potential impact on multiple domains of childhood development. The impact of hearing loss on domains such as *effort of listening* and resulting *fatigue*[1] in difficult listening condition are now receiving research attention. Although the literature identifies key areas such as *difficulty hearing in challenging situations* with some confidence, the ramifications of these difficulties upon childhood development are not fully understood. Quantifying the impact of UHL on childhood development, the long-term significance of these consequences, and the identification of the patient groups most susceptible to negative sequelae, require further investigation.

Lessons from the qualitative study

Our qualitative study highlighted that children are keen to normalise their hearing loss, making detection of the full impact difficult. Only with detailed exploration of a child's daily routine did it become apparent that children have adapted strategies to limit the effects of challenging listening conditions. They consciously positioned themselves and turned their head in relation to

the source of sound, focused or concentrated more, or sought help from friends and families, to overcome their difficulties. This effort resulted in new and *hidden consequences*, such as tiredness, distraction, loss of concentration, behavioural issues and feelings of frustration; all of which are potentially important when seeking to meaningfully assess the impact of the UHL or benefit from a therapeutic intervention.

Understanding what is important to children with UHL is essential, so that parents and teachers can provide the necessary support and healthcare providers can use patient-specific outcome domains that are most relevant to the specified patient group. In addition, to identifying patient-specific outcome domains, there is also a need to use appropriate instruments, validated to detect the hidden impact. Our study also highlighted the reluctance to use certain interventions, such as conventional hearing aids. As a result of the desire to normalise their hearing loss, children were reluctant to use an intervention that would highlight the problem they were seeking to overcome or ignore.

By contrast, parents did not attempt to normalise the impact of the conductive UHL, rather choosing to focus on describing the hearing difficulties their child faced and the adaptations needed to overcome these difficulties. Acceptance of their child's UHL was a novel finding, presumably resulting from observation of their child appearing to successfully overcome the difficulties with the help of subtle changes to daily living. Some of the imposed limitations described included the need for support at school and home, ongoing monitoring of impact on education and concerns about the future. Of concern, acceptance may serve as a barrier to seeking, or complying with, an intervention.

Other barrier towards managing conductive UHL- the choice of these outcomes being influenced by clinician opinion regarding the likely consequences and the nature of available outcome measurement instruments (OMI). There is a need to identify bespoke patient-specific outcome domains to help evaluate the impact of UHL across the stages of childhood, both within and between affected individuals, to enable greater understanding of the impact upon development and facilitate meaningful evaluation of the effectiveness of novel and existing therapeutic interventions.

The problem of meaningfully measuring the impact of UHL and/or therapeutic interventions-

Measurement of outcomes in children is predominantly influenced by clinician opinion, which is based largely on the experience of managing children with bilateral moderate to profound sensorineural hearing loss. Available outcome measurement instruments (OMI) are also designed to assess difficulties these children face and to measure the effectiveness of interventions used in managing this loss. Although this transferrable information is useful, there is emerging evidence that problems faces by children are unique. The findings of our study revealed hidden impact on behaviour, fatigue, tiredness and lack of concentration. These difficulties are often secondary to the adaptations children make to overcome difficulties faced in challenging listening environment. Instruments to measure the burden of these difficulties are lacking. Questionnaires such as the Speech Spatial and Qualities-Child (SSQ-C) may be helpful in measuring the impact on this domain. Novel instruments such as the Fatigue assessment scale, effort assessment scale are also being utilised in some studies to evaluate their suitability in testing the impact of monaural hearing.

In an attempt to identify relevant instruments, our participants completed three questionnaires at the end of the qualitative interviews. Nine of the 11 children completed the questionnaires. We chose Speech Spatial and Qualities- Child (SSQ-C) questionnaire, Fatigue Assessment Score (FAS) and Listening Effort Score (EAS). As our participants were selected for a qualitative study, we had a small sample size, so we have not tested the data statistically, but present descriptive findings. (Table 1)

Fatigue assessment score measures fatigue and the score ranges from 0-40. The lower the score, the less fatigue the individual perceives. The median fatigue assessment score was 7/40. Only 2 patients scored above the median score. (12/40 & 27/40). Most children had low fatigue assessment scores, although in the interviews, children talked about their tiredness. Similarly listening effort assessment score (EAS) is scored from 0-60 and is used to detect listening effort, with higher scores indicating significant effort. . The median score in our cohort was 35 and only 3 children scored above the median. As shown in the scatter plot in Figure 1, the percentage scores of EAS and FAS do not correlate.

SSQ –C identifies difficulties faced by children in difficult hearing conditions and tests binaural hearing. The SSQ-C questionnaire evaluates aspects of listening that are not evaluated with

standard speech perception testing[2]. The three domains evaluate the speech, spatial hearing and qualities of hearing, with higher scores suggesting better functioning. The median score in the speech domain in our participants was 63/100, spatial hearing 78/130 and qualities of hearing 75/100. Although this is a useful tool to measure listening difficulties, normative data are not available and it relies on comparison of scores, of before and after intervention.

Available instruments for measuring impact of UHL measure individual domains. The lack of an instrument that measures the relevant domain important to children is another important factor highlighted by the SR.

Figure 1.5 Scatter plots of EAS and FAS scores

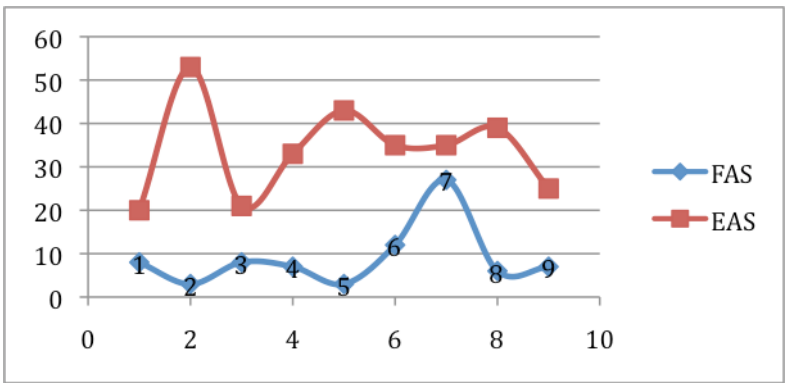

Table-1.5 Summary data of questionnaires

Patient	FAS (40)	EAS (60)	Speech (100)	Spatial (130)	Qualities (100)
Eva	8	20	63	70	64
Liam	3	53	83	64	93
Ellie	8	21	45	51	37
Luke	7	33	72	88	82
Leo	3	43	78	94	80
Dan	12	35	69	81	65

Nathan	27	35	45	36	35
Gerrard	6	39	59	78	76
Edward	7	25	50	81	76

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Chapter 6: Conclusion and Future directions

Our study highlights that the potential impact of UHL in childhood is not inconsequential, varying between children and within childhood. If we are to evaluate UHL in children in a clinically meaningful manner, it is necessary that we address the consequences perceived by parents and children, and not simply select from existing tools and instruments that may not be directly relevant to the problems experienced, or patient group. Using parent and child interviews, we identified *hidden problems* consequent upon children adapting to hearing difficulties in challenging situations, such as concerns about *behaviour, tiredness*, and d*istraction*. Likewise, although there are positive consequences from parental acceptance of their child's hearing problem, it could prove to be a barrier to seeking intervention and/or supporting their child, and as such parental education and support should be emphasised.

Future work

This study highlights the following areas of unmet research need, which either hinder evaluation of the problem or limit options for clinical management:

- Greater understanding of the impact of conductive UHL in children.
- Need for bespoke outcome measurement instruments (OMI) to quantify the true extent of problems in children with conductive UHL.
- Identification of interventions effective and acceptable in this patient group.

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Appendices

Appendix 1.3. Topic Guide for Child / Young person interviews

The Impact of Unilateral Hear loss: Topic Guide for Child / Young person interviews

The interviews will aim to investigate the impact of the child's hearing loss by looked at various aspects such as the effect on education, behaviour and development.

N.B: The interview will take the form of a conversation. Interviewees will have the opportunity to raise issues of importance to them. This topic guide may therefore change as data collection progresses.

At the start of the interview

- Thank the child / young person for agreeing to be interviewed
- Briefly tell them about the research, why you are doing it and what you are aiming to find out
- Check it is still OK to record the conversation (even though written consent will have been received). Let them know that a written copy of the interview will be made and that it will not have their name on it. It will be anonymous
- Reassurance that anything they tell you will be kept anonymous (their name will not appear on any of the written copies of the interview or on any of the questionnaires they fill in)
- Ask about any worries about the interview before starting
- Have they got any questions?
- Make clear it is OK to stop at any point or refuse to answer questions during interview

Questions

- Lets talk about your daily routine- Weekday and weekend
- What wakes you up in the morning?
- Getting out and about, are there any aspects you do differently?
- Moving around in school and with friends- Any preferences?
- Any preference about classroom seating?
- Other activities such as hanging out with friends, music, sports, computer games
- Have you tried hearing aids, what do you think about them?
- Are there things you like to do but find them difficult?
- What about the future?
- Any questions or concerns?

Appendix 1.4. Topic Guide for Parent interviews

MANCHESTER 1824
The University of Manchester

Central Manchester University Hospitals NHS
NHS Foundation Trust

The Impact of Unilateral Hear loss: Topic Guide for Parent interviews

The interviews will aim to investigate the impact of the child's hearing loss by looked at various aspects such as the effect on education, behaviour and development.

N.B: The interview will take the form of a conversation. Interviewees will have the opportunity to raise issues of importance to them. This topic guide may therefore change as data collection progresses.

At the start of the interview

- Thank the parent(s) for agreeing to be interviewed
- Brief outline of the purpose of the research
- Reassurance about anonymity, confidentiality, and non-impact on service delivery
- Check it is still OK to record the conversation (even though written consent will have been received)
- Ask about any concerns before starting
- Have they got any questions?
- Make clear it is OK to stop at any point or refuse to answer questions during interview

Questions

- Lets talk about their daily routine- Weekday and weekend
- Interaction within the family
- What wakes them up in the morning?
- Getting out and about, are there any aspects they do differently?
- Moving around in school and with friends- Any preferences?
- Any preference about classroom seating?
- Other activities such as hanging out with friends, music, sports, computer games
- Have they tried hearing aids, what do they think about them?
- Are there things they like to do but find them difficult?
- What about the future?
- Any questions or concerns?

Appendix 3. Letter of Invitation

Central Manchester University Hospitals NHS
NHS Foundation Trust

Royal Manchester Children's Hospital
Oxford Road
Manchester
M13 9WL

LETTER OF INVITATION

Title of study: Impact of Unilateral hearing loss in children

Dear Parent,

I am writing to you about a study that is being carried out by researchers from the University of Manchester. The aim of this study is to gain a better understanding of the impact of hearing loss in one ear in childhood. The researchers hope that this will allow them to manage this condition better in the future.

I am contacting you because you have a child who has hearing loss in one ear and attends Royal Manchester Children's Hospital. You do not have to take part in the study but you may be contacted to check that you have received the information. Your details will not be passed to the researchers without your permission, but you may be contacted by a member of the clinical team to check that you have received this information.

In this envelope, you will find detailed information about the study for both you and your child. This tells you exactly what is involved and how you can take part.

What do I have to do?

- Read the parent information sheet and decide if you want to take part.
- Pass on the children's information sheet to your son or daughter who has hearing loss in one ear and see if they would be happy to talk to a researcher
- If you and your child are interested in taking part and would like to be contacted by our researchers, please complete and return the slip (on the second page). Agreeing to be contacted by a researcher does not mean that you have to take part.
- If you have any questions, please contact the researchers named below.

Thank you for taking the time to read this letter. If you require any further details of the project you can speak directly to a member of the research team, Jaya Nichani who works at Royal Manchester Children's Hospital. The contact number is 0161 7010994. You can also send them an email at: jaya.nichani@cmft.nhs.uk

Yours sincerely

Mrs Jaya Nichani
Consultant Paediatric Otolaryngologist

PERMISSION SLIP

MANCHESTER 1824
The University of Manchester

Central Manchester University Hospitals NHS
NHS Foundation Trust

As the parent(s)/guardian(s) of: _____

I give permission to be contacted by the researchers to discuss the study further.

Parent(s) name: _____

Telephone/Mobile no.: _____

Email address: _____

Signed: _____ Date: _____

Please enclose the completed reply slip into the stamped addressed envelope provided and return to: ***Mrs Jaya Nichani, Royal Manchester Children's Hospital, Oxford Road, Manchester, M13 9WL.***

Appendix 4. Assent form for young people, 11 – 15 years old

MANCHESTER 1824
The University of Manchester

Central Manchester University Hospitals NHS
NHS Foundation Trust

The Impact of Single Sided Deafness

(The Impact of Unilateral Hearing Loss)

Study ID: 172404

Assent form for young people, 11 – 15 years old (V1.0, Apr 2016)

Young person (or, if unable, parent on their behalf) to circle all they agree with:

Has somebody explained this study to you?	Yes / No
Do you understand what this study is about?	Yes / No
Have you asked all the questions you have?	Yes / No
Have you had your questions answered in a way you understand?	Yes / No
Do you understand that it's OK to stop taking part at any time?	Yes / No
Do you agree to be interviewed	Yes / No
Do you agree to your interview being recorded	Yes / No
Are you happy to take part?	Yes / No

If ANY answers are "No" or you don't want to take part, DON'T sign your name!

If you DO want to take part, you can write your name, todays date and sign below.

Your name: _____ Date _____

Sign: _____

The doctor / nurse who explained this project to you needs to sign too:

Print Name: _____

Sign: _____

Date: _____

1 copy for participant, 1 for researcher file, 1 (original) to be kept in patients notes

Appendix 5. Adolescent (16 – 17 years) Consent form

MANCHESTER 1824
The University of Manchester

Central Manchester University Hospitals NHS
NHS Foundation Trust

The Impact of Unilateral Hearing Loss
Study id:172404

Adolescent (16 – 17 years) Consent form
Version 2.0, June 2016

Please initial each of the boxes

1.	I confirm that I have read the patient information sheet dated..... (Version.....) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.	Yes / No ☐
2.	I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without their medical care or legal rights being affected.	Yes / No ☐
3.	I agree to be interviewed as part of this study	Yes / No ☐
4.	I agree to the interview being audio recorded and transcribed	Yes / No ☐
5.	I understand and agree that anonymous quotations from my interview can be used in reports, publications or conferences to illustrate the findings of the research study	Yes / No ☐
6.	I agree to take part in the above study.	Yes / No ☐
7.	I understand that data collected during the study may be looked at by individuals from the University of Manchester, from regulatory authorities or from the NHS Trust, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my data.	Yes / No

Patients name (Print)	Signature	Date

Name of person taking consent (Print)	Signature	Date

1 copy for patient, 1 copy for researcher site file and 1 (original) to be kept in the patient's medical notes

Appendix 6. Adult Consent form

MANCHESTER 1824 The University of Manchester

Central Manchester University Hospitals NHS NHS Foundation Trust

The Impact of Unilateral Hearing Loss

Adult Consent form
Version 2.0, June 2016, Study Id: 172404

Please initial each of the boxes

1.	I confirm that I have read the parent information sheet dated..... (Version.....) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.	Yes / No ☐
2.	I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without the medical care or legal rights being affected.	Yes / No ☐
3.	I agree to be interviewed as part of this study	Yes / No ☐
4.	I agree to the interview being audio recorded and transcribed	Yes / No ☐
5.	I understand and agree that anonymous quotations from my interview can be used in reports, publications or conferences to illustrate the findings of the research study	Yes / No ☐
6.	I agree to take part in the above study.	Yes / No ☐
7.	I understand that data collected during the study may be looked at by individuals from the University of Manchester, from regulatory authorities or from the NHS Trust, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my data.	Yes / No ☐

Parent / guardian's name (Print)	Signature	Date
Name of person taking consent (Print)	Signature	Date

1 copy for parent, 1 copy for researcher site file and 1 (original) to be kept in the child's medical notes

Appendix 7. Adult information Sheet

The impact of one sided hearing loss in children

Adult information Sheet (Version 2, June 2016)

We would like to invite you and your child to take part in our research study. Joining the study is entirely up to you. Before you decide, we would like you to understand why the research is being done and what it would involve for your child and you. One of our team will go through this information sheet with you again at your convenience and answer any questions you have. Please take time to read the information and feel free to talk to others about the study if you wish.

Important things you need to know

- We would like to find out how the hearing loss effects your child
- We would like to interview you. The interview will be a conversation and each interview will take 30 – 45 minutes

Why are we doing this research?

Doctors are now able to identify children with single sided hearing loss (loss of hearing in one ear) a lot earlier than previously. As such there is a need to understand how this hearing loss affects a growing child. However, the full effect of this hearing loss is not fully understood. The aim of this study is to investigate the effect of single sided hearing loss in children aged 11 – 17 years of age.

Why have you been asked to take part?

You are a parent of a child with single sided hearing loss, who is between the ages of 16 and 17 years old.

What would taking part involve?

We would like to interview you to find out what impact your child's hearing loss has had on them and you.

Interview

The interview will be a conversation, which will take place at a routine clinic visit and will take about 30 minutes. The conversation/s will be audio recorded so that we can listen carefully to everything that has been said and make a written copy of the interview. We will not use anybody's real name in the study reports, publications and presentations.

Do we have to take part?

It is up to you whether you take part in this study. Not taking part will have no effect on the care your child receives now or in the future. If you decide you do want to take part you will be asked to sign a consent form this is to show that you understand what will happen. Even after signing the consent, if you decide at any time that you no longer want to take part that is OK and it will not affect your child's care.

What are the possible benefits of taking part?

This study will not help you or your child directly. Instead, it will help us to understand the effect of single sided deafness on children and their everyday lives.

What are the possible risks of taking part?

We do not expect there to be any risk in taking part. During the interview we will ask you about the effect of your child's' hearing loss on their daily living. If you feel uncomfortable answering any of the questions we ask, you can say you would rather

not answer that particular question/s. If you need to discuss this further you can contact the clinicians looking after your child.

What if there is a problem?

Minor complaints
If you have a minor complaint then you need to contact the researcher(s) in the first instance. Mrs Nichani may be contacted on 0161 701 0994, email jaya.nichani@cmft.nhs.uk. Alternatively you may speak to Professor Peter Callery, who is one of the supervisors in this study. Professor Peter Callery may be contacted on 0161 3067612, email peter.callery@manchester.ac.uk

Formal Complaints
If you wish to make a formal complaint or if you are not satisfied with the response you have gained from the researchers in the first instance then please contact the Research Governance and Integrity Manager, Research Office, Christie Building, University of Manchester, Oxford Road, Manchester, M13 9PL, by emailing: research.complaints@manchester.ac.uk or by telephoning 0161 275 2674 or 275 2046.

Will taking part in the study be kept confidential?
Most of the information will be anonymised but any personal data collected will be processed in line with the University's policies and the Data Protection Act 1998. . Any information we collect will be labelled with a code instead of your name so that no one will know it is your record. The information with the code will be entered into the main computer (database). The database is kept securely on encrypted computers. *A member of the research team* entering the information will have a personal password to access the database. We will keep records safe so that nobody except the researchers can see it. The University of Manchester for up to 15 years will store paper documents securely. Everything will be confidential.

The only other time someone might need to look at the study information is during an audit or monitoring visit. This is when people from the University of Manchester, NHS Trust or regulatory authorities review all of the data to make sure the study is being carried out as planned. If you agree, they will include your identifiable data when doing checks (they will see it belongs to you). Anyone that does look at the data will have a duty to keep it confidential.

What will happen to the results of the study?
The results of the study will be published in the medical journals, perhaps used in further research and presented at medical conferences. This study also forms part of a student project, registered with the University of Manchester, and the results will be published as a thesis. You will also receive a summary of the study findings

Who is organising and funding the study?
The University of Manchester has overall responsibility for organising the study which will be carried out at Central Manchester University Hospital NHS Foundation Trust This study has not received any extra external funding.

Who has reviewed this study?
All research in the NHS is looked at by an independent group of people, called a Research Ethics Committee, to protect your / your child's interests. This study has been reviewed and given favourable opinion by [*Insert name*] Research Ethics Committee.

What do I do if I want to take part?

You can either complete the reply slip you received with the invite letter in the stamped address envelope or you can ring or email the researcher Jaya Nichani at 0161 7010994 or email jaya.nichani@cmft.nhs.uk

Further information about research

Dir. Jaya Nichani (Chief Investigator), [0161 7010994]

For general information about public involvement in health research you can contact INVOLVE on 023 8065 1088. INVOLVE is a national advisory group.

Thank you for reading this Parent information sheet and considering yours and your child's participation in this study.

Appendix 8. Young Person's (16 -17 years) information Sheet

The impact of one sided hearing loss in children

Young Person's (16 -17 years) information Sheet

We would like to invite you and your parent/s to take part in our study. Joining the study is entirely up to you. So before you decide, we would like you to understand why the research is being done. One of our team will go through the information sheet with you and answer any questions you have.
Please take time to read the information and feel free to talk to others about the study if you wish.

Important things you need to know

- We would like to interview you and your parent/s. The interview will be a chat and will take 30 – 45 minutes
- We would like to find out how your hearing loss effect you.
- We will also give you some questionnaires to fill in

Why are we doing this research?
Doctors are now able to identify children with hearing loss in one ear earlier than previously. However, the full effect of this hearing loss is not understood. The aim of our study is to investigate the effect of hearing loss in one ear in children aged 11 – 17 years of age.

Why have you been asked to take part?
You have hearing loss affecting one ear and are aged between 16 and 17 years old.

What would taking part involve?
We would like to ask you how your hearing loss affects you on a daily basis. At the end of the chat with you we would ask you to fill in some questionnaires. We would also like to interview your parent/s, as we would like to know how they feel your hearing loss has affected you.

Interview
The interview will be a chat, which could take place at a routine clinic visit. and will take about 30 minutes. The interview will be audio recorded so that we can listen carefully to everything that has been said. We will also make a written copy of the interview. We will not use anybody's real name in the study reports.

Questionnaires
We would like you to complete three questionnaires. These ask questions about your ability to hear in different situations. They will also ask about how tiring hearing may sometimes be for you.

Do we have to take part?
It is up to you whether you and your parent take part in this study. Not taking part will have no effect on the care you receives now or in the future.
If you decide you do want to take part you will be asked to sign a consent form. This is to say that you understand what we have suggested and you are happy to go ahead. Even after signing the consent form, if you decide at any time that you no longer want to take part that is OK.

What are the possible benefits of taking part?

This study will not help you directly. Instead it will help us to understand the effect of single sided hearing loss on children and their everyday lives. This may help you in the future.

What are the possible risks of taking part?
We do not expect there to be any risks but it is possible you might feel uncomfortable answering some of the questions we ask. If this happens, you can say you would rather not answer that particular question. . If you need to discuss this further you can contact the clinicians looking after you.

Will taking part in the study be kept confidential?
Most of the information will be anonymised but any personal data collected will be processed in line with the University's policies and the Data Protection Act 1998. Any information we collect will be labelled with a code instead of your name so that no-one will know it is your record. The information with the code will be entered into the main computer (database). The database is kept securely on encrypted computers. *A member of the research team* entering the information will have a personal password to access the database. We will keep records safe so that nobody except the researchers can see it. Paper documents will be stored securely by the University of Manchester for up to 15 years. Everything will be confidential.

The only other time someone might need to look at the study information is during an audit or monitoring visit. This is when people from the University of Manchester, NHS Trust or regulatory authorities review all of the data to make sure the study is being carried out as planned. If you agree, they will include your identifiable data when doing checks (they will see it belongs to you). Anyone that does look at the data will have a duty to keep it confidential.

What if there is a problem?

Minor complaints
If you have a minor complaint then you need to contact the researcher(s) in the first instance. Mrs Nichani may be contacted on 0161 701 0994, email jaya.nichani@cmft.nhs.uk. Alternatively you may speak to Professor Peter Callery, who is one of the supervisors in this study. Professor Peter Callery may be contacted on 0161 3067612, email peter.callery@manchester.ac.uk

Formal Complaints
If you wish to make a formal complaint or if you are not satisfied with the response you have gained from the researchers in the first instance then please contact the Research Governance and Integrity Manager, Research Office, Christie Building, University of Manchester, Oxford Road, Manchester, M13 9PL, by emailing: research.complaints@manchester.ac.uk or by telephoning 0161 275 2674 or 275 2046.

Will your information be kept confidential?
Yes. All information collected about you will be kept confidential. Your name will be removed from all the information we collect and it will be given a code so that you cannot be identified.
Only anonymous quotes from written copy of the interview will be used in reports.

Your GP will be told of your participation in the study. With your permission relevant medical records may be looked at by the study team and authorities. This is to check that the study is being carried out correctly.

What will happen to the results of the study?
This study forms part of a student project for a thesis at the University of Manchester. We also plan to write articles for people to read – but we will not use anybody's real name. You will also receive a summary of the study findings

Who is organising and funding the study?

The University of Manchester has overall responsibility for organising the study which will be carried out at Central Manchester University Hospital NHS Foundation Trust This study has not received any extra external funding.

Who has reviewed this study?
All research in the NHS is looked at by an independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed and given favourable opinion by [*Insert name*] Research Ethics Committee

What do I do if I want to take part?
You can either complete the reply slip you received with the invite letter in the stamped address envelope or you can ring or email the researcher Jaya Nichani at 0161 7010994 or email jaya.nichani@cmft.nhs.uk

Further information about research

Dr. Jaya Nichani (Chief Investigator), [0161 7010994]

For general information about public involvement in health research you can contact INVOLVE on 023 8065 1088. INVOLVE is a national advisory group.

Thank you for reading this information sheet and considering your participation in this study.

Appendix 9. Parent / guardian information Sheet

The impact of one sided hearing loss in children

Parent / guardian information Sheet (Version 1, Mar 2016)

We would like to invite you and your child to take part in our research study. Joining the study is entirely up to you. Before you decide, we would like you to understand why the research is being done and what it would involve for your child and you. One of our team will go through this information sheet with you again at your convenience and answer any questions you have. Please take time to read the information and feel free to talk to others about the study if you wish.

Important things you need to know

- We would like to find out how the hearing loss effects your child
- We would like to interview your child and you. The interview will be a conversation and will take 30 – 45 minutes
- We will also give you some questionnaires to fill in

Why are we doing this research?

Doctors are now able to identify children with single sided hearing loss (loss of hearing in one ear) a lot earlier than previously. As such there is a need to understand how this hearing loss effects a growing child. However, the full effect of this hearing loss is not fully understood. The aim of this study is to investigate the effect of single sided hearing loss in children aged 11 – 17 years of age.

Why have you been asked to take part?

You are a parent of a child with single sided hearing loss, who is between the ages of 11 and 17 years old.

What would taking part involve?

We would like to interview you to find out what impact your child's hearing loss has had on them and you. We would also like to interview your child if they agree, as we would like to know how they feel their hearing loss has affected them. Your child will also fill in some questionnaires at the end of the interview.

Interview

The interview will be a conversation, which will take place at a routine clinic visit and will take about 30 - 45 minutes. The conversation/s will be recorded so that we can listen carefully to everything that has been said and make a written copy of the interview. We will not use anybody's real name in the study reports, publications and presentations.

Questionnaire

We would also like your child to complete some questionnaires. These questionnaires will ask about your child's ability to hear in different situations and the possible impact of hearing with one ear. We will help your child to fill in the questionnaire. We can show you these questionnaires at any time before the start of the study should you wish to see them.

Do we have to take part?

It is up to you whether you and your child take part in this study. Not taking part will have no effect on the care your child receives now or in the future. If you decide you do want to take part you will be asked to sign a consent form and your child to sign an assent form. This is to show that you both understand what will happen. Even after signing the consent and assent form, if either of you decide at any time that you no longer want to take part that is OK and it will not effect your child's care.

What are the possible benefits of taking part?

This study will not help you or your child directly. Instead, it will help us to understand the effect of single sided deafness on children and their everyday lives.

What are the possible risks of taking part?
We do not expect there to be any risk in taking part. During the interview we will ask you and your child about the effect of your child's hearing on their daily living. If you feel uncomfortable answering any of the questions we ask., you can say you would rather not answer that particular question/s. If you need to discuss this further the researcher can arrange for you to meet the clinicians looking after your child.

What if there is a problem?
If you have a concern about any aspect of this study, you should ask to speak to the researcher (Mrs Nichani) who will do their best to answer your questions (0161 701 0994). Alternatively you may speak to Professor Peter Callery, who is one of the supervisors in this study. Professor Peter Callery may be contacted on 0161 3067612, If you remain unhappy and wish to complain formally, you can do this by contacting hospitals Patient Advice and Liaison Service (PALS). Details can be obtained from 0161 701 8711

Will our information be kept confidential?
Yes. All information collected about you and your child will be kept confidential and stored anonymously and securely under the provisions of the 1998 Data Protection Act.

Your name and your child's name will be removed from all the information we collect and the information will be given a code so that you and they cannot be identified. The information with the code will be entered into the main computer (database). The database is kept securely on encrypted computers. *A member of the research team* entering the information will have a personal password to access the database.

Only anonymous quotes from written copy of the interview will be used in reports, publications or conferences to show the findings of the study.

Your child's GP will be told of their participation in the study. With your permission, your child's relevant medical records may be inspected by the study team and regulatory authorities. This is to check that the study is being carried out correctly.

What will happen to the results of the study?
The results of the study will be published in the medical journals, perhaps used in further research and presented at medical conferences. This study also forms part of a student project, registered with the University of Manchester, and the results will be published as a thesis.

Who is organising and funding the study?
The organisations responsible for the study are University of Manchester and Central Manchester University Hospital NHS Foundation Trust. This study has not received any extra external funding.

Who has reviewed this study?
All research in the NHS is looked at by an independent group of people, called a Research Ethics Committee, to protect your / your baby's interests. This study has been reviewed and given favourable opinion by [*Insert name*] Research Ethics Committee.

Further information about research

Dr. Jaya Nichani (Chief Investigator), [0161 7010994]

For general information about public involvement in health research you can contact INVOLVE on 023 8065 1088. INVOLVE is a national advisory group.

Thank you for reading this Parent information sheet and considering yours and your child's participation in this study.

Appendix 10. Child information Sheet

The impact of one sided hearing loss in children

Why are we doing this research?

Doctors are now able to identify children with single sided hearing loss (loss of hearing in one ear) a lot earlier than previously. However, the full effect of this hearing loss is not understood. We would like to ask you how your hearing loss affects you.

What happens if I take part?

If you agree, we will have a chat with you about how your hearing loss affects you on a daily basis. There are no right or wrong answers. We will talk to you for about half an hour. You can stop at any time. Nobody will mind.
We would like to record this chat. This is so we can listen carefully to what you say. We don't want to miss things. We will show you how the recorder works. You can turn it off if you wish. Only the researchers will listen to the recording.

We would like you to complete some questionnaires. This questionnaire will ask questions about how well you hear in different situations. They will also ask about how tiring or effortful hearing is for you. We can show you these questionnaires at any time before the start of the study if you wish.

Will taking part help me?

No. We will use what you to understand the effect of the hearing loss. We hope

you will enjoy taking part

Do I have to take part?

We will ask your parent first. But it is up to you to decide if you want to take part. We won't mind if you say no. It will not change how you are looked after

What happens afterwards?

We will write reports for doctors, audiologists and other people who look after children with a hearing loss in one ear. We will not use your name in these reports.

Thank you for reading about our study.

Appendix 11. Listening Effort Assessment Scale

MANCHESTER 1824
The University of Manchester

Central Manchester University Hospitals NHS
NHS Foundation Trust

Listening Effort Assessment Scale

Date Completed:	Participants Initials:	Participant Id No.:
__ __ / __ __ / __ __ __ __	___ / ___ / ___	__ / ___

The following statements ask about the level of effort that you use when listening in daily life.

On the line below each statement, please circle the number that best indicates how you usually feel.

1. Do you have to put in a lot of effort to hear what is being said in conversation with others?

Lots of effort — *No effort*

0 1 2 3 4 5 6 7 8 9 10

2. How much do you have to concentrate when listening to someone?

Concentrate hard — *No need to concentrate*

0 1 2 3 4 5 6 7 8 9 10

3. How easily can you ignore other sounds when trying to listen to something?

Not easily ignored — *Easily ignored*

0 1 2 3 4 5 6 7 8 9 10

4. Do you have to put in a lot of effort to follow discussion in a class, a meeting or a lecture?

Lots of effort — *No effort*

0 1 2 3 4 5 6 7 8 9 10

Appendix 12. The Speech, Spatial, and Qualities of Hearing Scale (SSQ) for Children with Impaired Hearing

The Speech, Spatial, and Qualities of Hearing Scale (SSQ) for Children with Impaired Hearing

(Based on the adult SSQ developed by William Noble & Stuart Gatehouse; modifications by Karyn Galvin)

Child's Name: _____ Age: _____yrs_____mths__

Evaluation point: _____

Device Condition: Unilateral: Cochlear Implant Hearing Aid

Bilateral: Cochlear Implants Hearing Aids

Cochlear Implant + Hearing Aid

Right device: Type: _____ Date fitted: _____

Current usage: all day school only other: _____

Left device: Type: _____ Date fitted: _____

Current usage: all day school only other: _____

Appendix 13. The Fatigue Assessment Scale (FAS)

The Fatigue Assessment Scale (FAS)

Date Completed:	Participants Initials:	Participant Id No.:
__ __ / __ __ / __ __ __ __	___ / ___ / ___	__ / ___

The following ten statements refer to how you usually feel on a daily basis.
For each statement, choose one out of the five answers.
Please give an answer to each statement, even if you do not have any complaints at the moment.

Statement	Never	Sometimes	Regularly	Often	Always
1. I am bothered by fatigue	0	1	2	3	4
2. I get tired very quickly	1	1	2	3	4
3. I do not do much during the day	1	1	2	3	4
4. I have enough energy for everyday life	4	3	2	3	0
5. Physically, I feel exhausted	0	1	2	3	4
6. I have problems starting things	0	1	2	3	4
7. I have problems thinking clearly	0	1	2	3	4
8. I have no desire to do anything	0	1	2	3	4
9. Mentally, I feel exhausted	0	1	2	3	4
10. When I am doing something, I can concentrate quite well	4	3	2	3	0

Total score: ____/ 40

Jolanda De Vries et al, British Journal of Health Psychology (2004), 9, 279-291

Appendix 14. Letter of HRA Approval

NHS
Health Research Authority

Mrs Jaya Nichani
Consultant Paediatric Otolaryngologist
Central Manchester University Hospitals NHS Trust
Paediatric Otolaryngologist
Royal Manchester Children's Hospital
Oxford Road, Manchester
M13 9WL

Email: hra.approval@nhs.net

08 September 2016

Dear

Letter of HRA Approval

Study title:	**Impact of Unilateral hearing loss in children**
IRAS project ID:	**172404**
REC reference:	**16/LO/1080**
Sponsor	**University of Manchester**

I am pleased to confirm that HRA Approval has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications noted in this letter.

Participation of NHS Organisations in England

The sponsor should now provide a copy of this letter to all participating NHS organisations in England.

Appendix B provides important information for sponsors and participating NHS organisations in England for arranging and confirming capacity and capability. **Please read *Appendix B* carefully**, in particular the following sections:

- *Participating NHS organisations in England* – this clarifies the types of participating organisations in the study and whether or not all organisations will be undertaking the same activities
- *Confirmation of capacity and capability* - this confirms whether or not each type of participating NHS organisation in England is expected to give formal confirmation of capacity and capability. Where formal confirmation is not expected, the section also provides details on the time limit given to participating organisations to opt out of the study, or request additional time, before their participation is assumed.
- *Allocation of responsibilities and rights are agreed and documented (4.1 of HRA assessment criteria)* - this provides detail on the form of agreement to be used in the study to confirm capacity and capability, where applicable.

Further information on funding, HR processes, and compliance with HRA criteria and standards is also provided.

Appendix 15. Sponsorship Letter UoM

Faculty of Medical & Human Sciences
The University of Manchester
Oxford Road
Manchester M13 9PT

www.manchester.ac.uk

16 May 2016

To whom it may concern

Sponsor Reference: 16163

Role of the Research Sponsor under the Research Governance Framework for Health & Social Care and the Medicines for Human Use (Clinical Trials) Regulations 2004 (SI2004/1031)

I hereby confirm that the University of Manchester would be prepared to accept the role of research sponsor as currently defined in the *Research Governance Framework for Health & Social Care Version 2 (DoH 2005)* and the *Medicines for Human Use (Clinical Trials) Regulations 2004 (SI2004/1031)*, in relation to the study:

Impact of Unilateral Hearing loss in children.

I have been informed that this study will be led by **Dr Jaya Nichani** of The University of Manchester.

Sponsorship is subject to the following conditions:

1) The lead investigator for the study must be an employee of the University of Manchester. For student research the academic supervisor is considered to be the lead investigator.
2) An appropriate contract must be agreed between the University and the funding body.
3) The research must be reviewed and approved by appropriate ethics, NHS and regulatory bodies and registered in accordance with University insurance requirements.

To enable the sponsor to meet their responsibilities as listed in section 3.8 of the Research Governance Framework, Chief Investigators are asked to adhere to the responsibilities as outlined in section 3.6 of the Framework (available at: https://www.gov.uk/government/publications). In line with this requirement **Dr Jaya Nichani** must ensure that all involved in the research project understand and discharge their responsibilities in accordance with the agreed protocol and any relevant management, ethical and regulatory approvals.

Chief Investigators are also reminded that they must register NHS REC approval with The University of Manchester Research Ethics Office.

If you have any queries about sponsorship of this project then please address them to Professor Nalin Thakker, Associate Vice President for Research Integrity, The University of Manchester, Christie Building, Oxford Road, Manchester M13 9PL, or email research-governance@manchester.ac.uk

Yours Faithfully,

Lynne MacRae

Lynne MacRae Dated: **16.05.2016**
Research Practice Coordinator
Faculty of Medical & Human Sciences