Cochlear Implants for Children with Residual Hearing: Supporting Family Decision-making

Eunjung Na

Thesis submitted to the University of Ottawa
in partial Fulfillment of the requirements for the
Doctorate in Philosophy degree in Rehabilitation Sciences

School of Rehabilitation Sciences
Faculty of Health Sciences
University of Ottawa

We now have this light shining in our hearts, but we ourselves are like fragile clay jars containing this great treasure. This makes it clear that our great power is from God, not from ourselves.

2 Corinthians 4:7 (NLT)

Table of Contents

List of Tables

Chapter 1

Chapter 2

Chapter 3

Chapter 4

Chapter 5

List of Figures

Dissertation abstract

Background

Children with residual hearing have become eligible for consideration as cochlear implant (CI) candidates in some pediatric programs because of the positive clinical research outcomes of CIs. However, decision-making about CIs for children with residual hearing is difficult for parents because they experience uncertainty when their children show auditory benefits and are developing language through hearing aids (HAs). Clinicians may be uncomfortable recommending CI for these children due to variability in audiometric candidacy criteria in individual clinical practice. However, there is very limited information about the CI decision-making process and needs to assist the parents of these children and practitioners.

Objectives

We conducted a comprehensive study to better understand and support the CI decision-making experiences of families and practitioners. The objectives of our study were to: 1) explore the clinical characteristics and outcomes of children with residual hearing who received CIs, 2) summarize the evidence about the benefits and risks of CIs compared to HAs in children with residual hearing, and 3) explore the decision-making process and needs for children with residual hearing from the perspective of parents and practitioners.

Methods

This research project combined quantitative and qualitative research designs. A retrospective chart review was conducted to address the first objective. Data on the clinical characteristics of children with residual hearing were extracted from medical charts from a tertiary care pediatric

CI center in Ottawa, Canada. A systematic review was performed on the benefits and risks of CIs versus HAs for children with residual hearing to address the second objective. The third objective was addressed through two sub-studies. The first sub-study involved qualitative semi-structured interviews. A total of 12 parents participated in individual interviews. In the second sub-study, 17 practitioners at a pediatric CI center in Ottawa, Canada, and specialized teachers of the deaf and hard of hearing at local school boards were recruited, and four focus groups and one individual interview were conducted.

Overall Findings

This study showed that a total of 100 of 389 (25.7%) children who received CIs from 1992 to 2018 at the Children's Hospital of Eastern Ontario (CHEO) had residual hearing, representing more than half the children who were implanted in the last two years covered by the study. As documented in our study, overall, children with residual hearing demonstrated benefits in auditory functioning following cochlear implantation. Approximately 70% of these children achieved open-set word perception scores of 80% or more post-CI.

In the systematic reviews, a total of 3265 citations were identified, of which eight studies met inclusion criteria. The articles consisted of four moderate and two weak quality pre-post cohort studies and two weak quality cross-sectional studies. The systematic review confirmed that children with CIs showed significantly better speech perception scores than those with HAs. Limited evidence of improvement in auditory performance and non-significant improvement in speech intelligibility was found. Two aspects of social-emotional functioning (hyperactivity/inattention and pro-social behaviour) showed significant improvement with CIs. Our finding also contributes new information about the loss of residual hearing and device use.

Four studies provided data on risks following CIs; a total of 16 of 43 (37.2%) children showed loss of residual hearing and 14.0% (8/57) of children had discontinued or limited use of their CI or HA.

The qualitative interviews revealed that both parents and practitioners identified children's everyday functioning as an important factor that influenced their decision-making. It was clear through the qualitative research with parents that they held a strong preference for children's inclusion into hearing society. Spoken communication was a core value for the parents of these children and some parents expressed high expectations that their children's hearing would become 'normal'. We found that practitioners primarily supported parental decision-making by providing information on the practical aspects of the benefits and risks of CIs. Overall parents were satisfied with the decision-making process and decision support from practitioners. However, parents stressed the importance of receiving more personalized information that considered their specific concerns, values and preferences related to their child and family's circumstances. Practitioners also noted that more research among children with residual hearing is needed to guide parental CI decision-making.

Conclusion

To our knowledge, the findings from this dissertation are the first to examine decision-making for children with residual hearing. Our study contributes new information about the characteristics of children receiving CIs, the potential benefits and risks for children with residual hearing, and decision-making needs from the perspectives of families and practitioners. In addition, our research is a useful first step in understanding what families need to make better decisions to assist in the CI decision-making process for this specific population.

Acknowledgements

I would like to first thank my incredible thesis supervisor Elizabeth Fitzpatrick. My Ph.D. journey could not have been started and completed without her support, patience, and encouragement. I would also thank my co-supervisor Karine Toupin-April and committee members, Janet Olds and Lucie Brosseau. This research wouldn't have been possible without their tremendous support and guidance. I appreciate their patience in reviewing each part of my dissertation. Their constructive comments and challenging questions helped me to have a clearer sense of my research and to build a strong foundation in research. I am so honoured to have worked with this amazing thesis committee. I am grateful to the research members at the Child Hearing Lab at CHEO who have been very supportive of my research and provided incredible help. In particular, I would like to thank JoAnne Whittingham for her support, encouragement, and tremendous help. I would also like to thank the clinicians at the Audiology Clinic, CHEO who have been interested in this research. Their interest, participation, and enthusiasm for advancing knowledge made this research possible. I would especially like to thank the participants in this research: clinicians, families, and itinerant teachers of the deaf and hard of hearing who shared their time and experiences. I would also like to say thanks to my loving family. Especially to my wonderful husband, Mooseung, who always accompanied me on this journey and supported me in everything I needed physically and emotionally, I give my biggest thanks. Without his love and support, I could not have done this journey. "I love you so much, Moose". To my dad and mom, who never stopped encouraging me and trusted all my decisions. Their support, belief, encouragement, and prayers made everything possible. Finally, I also say thanks to baby Raon, who is growing up strongly in my belly. I would like to tell her when this little one is born, "Mom was able to complete this thesis with the joy of having you".

Chapter 1: Introduction

Chapter 1: Introduction

Statement of the problem

Children with typical auditory development recognize a wide range of acoustic sounds. Permanent childhood hearing loss results in the absence or degradation of sound stimuli, which influences the development of the functional auditory area of the brain. Consequently, speech and language development, academic achievement, social integration, and psychological well-being are affected when hearing loss occurs in early childhood (Hoffman et al., 2018; Kral & O'Donoghue, 2010; Moeller & Tomblin, 2015; Stevenson et al., 2010; Theunissen et al., 2014). Children with hearing loss should therefore receive appropriate interventions as early as possible to overcome difficulties associated with hearing loss (Ching et al., 2017; Kennedy et al., 2006; Sininger et al., 2010).

Early intervention for children with hearing loss includes the provision of hearing technologies (e.g., hearing aids [HAs], cochlear implants [CIs]), information and support for parents, and rehabilitation strategies to minimize the impact of hearing loss (Moeller & Tomblin, 2015; Yoshinaga-Itano, 2004, 2001; Young & Tattersall, 2007). The majority of children with hearing loss can access auditory information through conventional HAs. However, children with severe to profound bilateral sensorineural hearing loss receive little or no benefit from HAs. CIs are widely recommended for these children whose parents choose spoken communication (Geers, 1997, 2002; Miyamoto et al., 2003; Tomblin et al., 1999).

The use of CIs in children with severe to profound hearing loss has led to positive outcomes in speech and language, academic achievement, and social development. In some cases, children with CIs function comparably to their peers with normal, mild, or moderate

hearing loss (Blamey et al., 2001; Ganek et al., 2012; Geers et al., 2003; Marschark et al., 2007; Punch & Hyde, 2010). It has been well documented that children with CIs perform better on language-related outcomes when compared to their peers with severe to profound hearing loss who use HAs (Bat-Chava et al., 2005; Bittencourt et al., 2012; Fitzpatrick et al., 2012; Hoffman et al., 2016).

CI candidacy criteria is less well-defined for children with residual hearing since no clear audiological cut-point exists for CI surgery (Fitzpatrick et al., 2006, 2009; Hyde et al., 2010b; Leigh et al., 2011, 2016) and predicting children's postoperative prognoses remains a challenge (Fitzpatrick et al., 2009; Hanvey et al., 2016; Heman-Ackah et al., 2012; Maggs et al., 2017; Osberger et al., 2002). CI surgery for any child involves possible risks and negative consequences (Duncan, 2009; Zanetti et al., 2015), including surgical and device-related complications (Chute & Nevins, 2002; Dillon & Pryce, 2020). In addition, children with residual hearing are at risk of losing their remaining hearing, which complicates the decision for their parents (Hyde et al., 2010b; Johnston et al., 2008). Although preserving residual hearing may be possible with new developments in surgical techniques and device technologies, loss of residual hearing has been reported following CI surgery (Brown et al., 2010; Kopelovich et al., 2015; Meredith et al., 2017; Snels et al., 2019). For these reasons, decision-making about cochlear implantation for children with residual hearing who already benefit from HAs is less straightforward than for children with bilateral profound hearing loss. Several studies related to CI decision-making have reported that parents of children who had more preoperative residual hearing took more decision-making time and experienced more stress than parents of children with bilateral profound hearing loss (Burger et al., 2005; Fitzpatrick et al., 2009; Hyde et al., 2010a). Parents of these children may experience uncertainty and decisional conflict, which can

lead to delays in decision-making (Anmyr et al., 2016; Hardonk et al., 2010, 2011; O'Connor, 2010).

Shared decision making (SDM) has been widely adopted in healthcare and is one way to resolve decisional conflict and achieve high-quality decision-making (Chorney et al., 2015; Rose et al., 2017; Rosewilliam et al., 2011). SDM is an evidence-based approach in which families and practitioners work together to make health-care decisions related to clinical uncertainties and information imbalances that arise in the relationship between clinicians and patients in clinical settings (Edwards & Elwyn, 2009; Gabe et al., 2004; Légaré et al., 2006a, 2011; Légaré & Witteman, 2013). Through this approach, families and practitioners exchange clinical experiences and results, as well as information about the families' preferences and values to reach a decision together about the most appropriate interventions (Légaré et al., 2011; Makoul & Clayman, 2006).

To apply SDM in clinical practice, both parents and practitioners should contribute to the process. In particular, practitioners are required to have current and accurate information regarding diagnoses, characteristics of patients, factors affecting decisions, intervention options, and the benefits and risks of different options. Parents need to be aware of their own values, preferences, and expectations (Charles et al., 1997; Moumjid et al., 2007). However, to date, very limited information to guide decision-making in pediatric CI exists (Fitzpatrick et al., 2009; Porter et al., 2018). In order to guide parents in making an appropriate decision, it is important to collect more comprehensive evidence about this population of children.

Present state of knowledge

Childhood hearing loss

Approximately 466 million people worldwide have hearing loss, and 34 million of them are children (The World Health Organization [WHO], 2020). Prevalence studies show that approximately 1 to 3 out of every 1,000 children are born with permanent hearing loss (Hawley et al., 2017; Mehra et al., 2009; CDC, 2010; NIDCD, 2016). In Canada, a prevalence study estimated that more than 2,000 children are born with hearing loss every year, representing approximately six in every 1,000 babies (Bagatto et al., 2010; Eskander & Papsin, 2014).

Hearing loss is commonly categorized according to the degree of hearing loss (e.g., mild, moderate, moderate-severe, severe, and profound). It is also categorized based on etiology and type (conductive, sensorineural, or mixed [both conductive and sensorineural]). People with conductive hearing loss have problems in the outer ear and/or middle ear, and sensorineural hearing loss occurs when problems arise in the inner ear or the auditory nerve. Some people with sensorineural hearing loss are not able to hear sounds of certain frequencies, but they can hear some sounds through undamaged frequencies. This hearing ability is called residual hearing. For children, having residual hearing is important because it can impact their hearing and language performance.

Impact of childhood hearing loss

Hearing loss disrupts delivery of acoustic input to the central auditory system. Over time, this leads to reduced development of the primary auditory area in the brain (Flexer, 2011; Kral & Sharma, 2012). Due to limited auditory input, children with hearing loss often experience difficulties in speech perception and language acquisition. Difficulties in these areas can impact

academic achievement (Antia et al., 2009; Sarant et al., 2015), social interactions, (Antia et al., 2009, 2012; Netten et al., 2015; Yoshinaga-Itano, 2003), and overall quality of life (Arnoldner et al., 2014; Borton et al., 2010; Morettin et al., 2013; Roland et al., 2016).

Permanent hearing loss in children can cause a deficit in auditory perceptual processes and lead to delays in language development (Benasich et al., 2002; Ching et al., 2010; Nittrouer & Burton, 2005). According to Pittman et al. (2005), children with hearing loss showed significantly lower scores on a receptive vocabulary test (Peabody Picture Vocabulary Test, PPVT- III) than children with typical hearing over a wide range of ages. Ching and colleagues (2010) found that children with hearing loss exhibited lower scores in receptive and expressive language skills (Preschool Language Scale, PLS-4) compared to an age-matched normative group of children. Moreover, they needed higher signal-to-noise ratios (SNR) than their peers with normal hearing for speech perception (Ching et al., 2017). According to Moeller & Tomblin (2015), children with bilateral mild to severe hearing loss produced significantly fewer words and illustrated deficits in phonology and grammar compared to their peers with typical hearing. Some studies have demonstrated that limited language exposure is associated with reduced language processing efficiency (Fernald et al., 2012; Weisleder & Fernald, 2013).

Childhood hearing loss can also result in lower academic achievement. Although outcomes for these children have improved, on average their academic achievement remains behind their peers with typical hearing (Qi & Mitchell, 2012). Reed et al. (2008) reported that 7 of 25 (28%) children with hearing loss obtained lower academic achievement levels because they experienced a delay in receiving intervention and in language skills. Studies have shown that at the secondary school level, students with hearing loss are significantly behind their peers in reading, math, and science (Marschark et al., 2015; Qi & Mitchell, 2012; Spencer & Marschark,

2010). In particular, a study by Marschark et al (2015) indicated that having better spoken language abilities was associated with higher test scores in passage comprehension, mathematics calculation, science, and social studies.

Behavioural and psychosocial functioning issues may be more common in children with hearing loss compared to their peers with typical hearing. In particular, according to Keilmann et al. (2007), children with hearing loss expressed more anxiety and sadness and reported lower well-being when they were in higher grades. Stevenson et al. (2010) found that emotional and behavioural problems in children with hearing loss were more common than in their peers due to their limited language skills. Another study on older youth with hearing loss indicated that adolescents experienced emotional and behavioural difficulties due to their limitations in language and reading comprehension (Stevenson et al., 2017). In terms of psychosocial functioning, better oral communication skills are related to lower levels of psychopathology (Barker & Briggs, 2009; Percy-Smith et al., 2008; Van Eldik et al., 2004; Theunissen et al., 2015). For example, according to Theunissen et al. (2015), children with profound hearing loss and who have CIs showed lower levels of psychopathological symptoms than children with moderate or severe hearing loss who have HAs.

Additionally, several studies found that children with hearing loss have lower scores on quality of life than do their peers with typical hearing (Borton et al., 2010; Roland et al., 2016; Ronner et al., 2020; Schick et al., 2013). A systematic review and meta-analysis by Roland et al. (2016) confirmed statistically and clinically significant differences in Pediatric Quality of Life Inventory (PedsQL) scores between children with hearing loss and those with typical hearing, specifically in school activities and social interactions. In addition, Rich et al. (2013) found that children with hearing loss showed less mature social skills with fewer quality friendships than

their peers with typical hearing. A recent qualitative study by Dammeyer et al. (2018) asked children with CIs aged 11–15 years (n=65) about their CI use and other factors related to communication, experiences of hearing loss, social participation and friendships, and psychological well-being. A total of 55.4% of these children who participated in the interviews reported that they felt different from their peers with typical hearing and 18.5% indicated that they try to hide their CIs often or all of the time, and these children still failed to achieve the same level of performance and satisfaction as their peers.

Despite improvements in technology, the literature suggests that children with hearing loss are at risk for increased difficulties in literacy, academic, social functioning, and quality of life.

Interventions for childhood hearing loss

Universal newborn hearing screening (UNHS) has been widely implemented in developed countries to identify childhood hearing loss within the first few months of life (Akinpelu et al., 2014; De Leenheer et al., 2011; American Academy of Pediatrics & Joint Committee on Infant Hearing [JCIH], 2007; JCIH, 2019). These programs have led to earlier intervention, providing children with hearing loss with greater opportunities to develop age-appropriate language skills, behaviour skills, and social-emotional competence (Fulcher et al., 2015; Stika et al., 2015).

At diagnosis, parents of children with hearing loss are presented with several intervention options, which include choices about hearing technology and communication development. The severity of hearing loss, that is, the amount of residual hearing, is an important consideration for parents and practitioners when discussing appropriate interventions. Maximizing residual hearing

and ensuring optimal access to sound is the foundation of spoken language intervention programs.

Parents of children with hearing loss can choose from two major hearing technologies, HAs and CIs, based primarily on the severity of their hearing loss. The majority of children with hearing loss can access auditory information through HAs, which constitute the first step in the intervention process for most children (Hoffman & Beauchaine, 2007; JCIH, 2019). While the majority of children with hearing loss benefit from HAs, CIs are recommended for children with severe to profound hearing loss who receive little or no benefit from HAs (Geers, 1997, 2002; Miyamoto et al., 2003; Tomblin et al., 1999).

CI candidacy criteria

In 1990, the Food and Drug Administration (FDA) in the U.S. approved CI surgery in children with bilateral profound hearing loss as young as 2 years of age. Since then, the candidacy criteria have changed and expanded. CI candidacy is primarily determined based on hearing levels. Until early 2019, the FDA candidacy criteria included bilateral sensorineural profound hearing loss [pure-tone average (PTA) > 90 dB HL at 500, 1000, 2000 Hz] for children aged ≤ 2 years. For children aged two years and older, children with bilateral sensorineural severe to profound hearing loss (PTA > 70 dB HL at 500, 1000, 2000 Hz) are also considered CI candidates.

In July 2019, FDA expanded the CI candidacy criteria for one manufacturer to include patients 5 years and older with single-sided deafness (SSD) and asymmetric hearing loss (AHL) who have profound hearing loss in the ear to be implanted and better than moderate hearing loss in the other ear. Most recently, the FDA expanded the criteria to include children 9 months of age or older who have profound hearing loss in both ears (FDA, 2019). In determining CI

candidacy, it is generally agreed that families and practitioners need to consider individual environmental factors and characteristics rather than just adhering to audiometric criteria (Chundu & Flynn, 2014; Fitzpatrick et al., 2009; Leigh et al., 2011, 2016). Assessments of the child's functioning also usually involve the use of auditory behaviour questionnaires, as well as closed-set and open-set word and sentence tests that are selected clinically depending on the child's age and linguistic function. Table 1 outlines the FDA candidacy criteria approved for the devices provided by the three largest CI manufacturers.

Table 1. FDA-labeled indications of pediatric CI candidacy for three manufacturers

	Advanced Bionics Corp.		**Cochlear ™**		**Med EL®**	
Age	12 months – 17 years		9 months – 17 years		12 months – 17 years	
			9 – 23 m	24 m – 17 y	< 5 years	≥ 5 years
Audiometric criteria	• Profound (≥ 90 dB) • Bilateral SNHL		• Profound (≥ 90 dB) • Bilateral SNHL	• Severe to profound • Bilateral SNHL	• Profound (≥ 90 dB) • Bilateral SNHL	SSD or AHL
Age	< 4 years	≥ 4 years	12- 23 m	24 m – 17 y		
Speech perception criteria	• MAIS, ESP or • < 20% open-set words (MLNT or LNT)	• < 12% correct on open-set words (PBK) or • < 30% correct on open-set sentences (HINT-C)	• MAIS, ESP	• ≤ 30% correct on open-set words (MLNT or LNT)	• < 20% correct on open-set words (MLNT or LNT)	

SNHL, Sensorineural hearing loss; SSD, single-sided deafness; AHL, asymmetric hearing loss; IT-MAIS, Infant-Toddler Meaningful Auditory Integration Scale; MAIS, Meaningful Auditory Integration Scale; LNT, Lexical Neighborhood Test; MLNT, Multisyllabic Lexical Neighborhood Test; PBK, Phonetically Balanced-Kindergarten Test; HINT, Hearing in Noise Test for Children; ESP, Early Speech Perception test

Benefits of CI in children with severe to profound hearing loss

Cochlear implantation has been well established as the preferred standard of care for children with severe to profound sensorineural hearing loss whose parents choose spoken language, because it provides more access to auditory information than conventional HAs (Geers, 1997; Leigh et al., 2011; Zwolan et al., 2004). According to a previous study by Peixoto et al. (2017) that examined 10 years of follow-up after CIs, improvements in word recognition scores of 84.6% and sentence test scores of 65.1% were reported in 132 children who participated in this study. Some children with CIs follow a similar trajectory of speech, language, and vocabulary development as their peers with typical hearing (Moog & Geers, 2010; Yoshinaga-Itano et al., 2010; Walker et al., 2019).

Prior to the availability of CIs, many children with severe to profound hearing loss required special education at school (Dye et al., 2009; Knoors & Marschark, 2012). The majority of children who receive early CI intervention are now educated in mainstream classrooms, and some of them show similar academic achievement as their peers with typical hearing (Bell et al., 2019; Mayer & Trezek, 2018; Punch & Hyde, 2010; Sugaya et al., 2015; Westby, 2016). In particular, Punch & Hyde (2010) reported that 79 of 151 (52%) of children with CI were educated in regular or age-appropriate classrooms most or all the time.

Several researchers have reported that children with CIs experience a quality of life similar to that of their peers with typical hearing (Loy et al., 2010; Roland et al., 2016; Warner-Czyz et al., 2009). Quality of life is a multi-dimensional concept which typically involves the physical, social, and emotional well-being of individuals. Various aspects of quality of life have been studied to understand children and youth with hearing loss because of the importance of communication and social participation in everyday life. In particular, children who experienced

a longer period of CI use have higher self-esteem, which is associated with better performance in social interactions (Martin et al., 2011). Percy-Smith et al. (2008) described that children with CI obtained results on measures of confidence, socialization, independence, and happiness similar to their peers with typical hearing. According to a systematic review by Morettin et al. (2013), children who received CIs early in life had better communication skills with their parents and at school. As a result, they had better social interactions and thus showed improvement in overall quality of life.

Risks of CI in children with hearing loss

As noted previously, unlike HAs, CIs are implanted surgically, a procedure that is known to be invasive (Chute & Nevins, 2002; Johnston et al., 2010). While children with CIs have shown good auditory and speech outcomes, multiple risks have been documented (Black et al., 2007; Farinetti et al., 2014; Lassig et al., 2005; Loundon et al., 2010; Migirov et al., 2009; Tarkan et al., 2013). Overall, surgical complications following CI surgery are similar to those of other types of ear surgeries, such as wound infections, temporary taste disturbance, tinnitus and balance disturbance, and facial nerve injury (Gheorghe & Zamfir-Chiru-Anton, 2015; Mylanus et al., 2004; Russell et al., 2013). Some children may experience complications related to anesthesia (Armstrong et al., 2013; Hawksworth & Ravury, 2015; Kim et al., 2017; Yeh et al., 2011). For example, according to Yeh et al. (2011), of 123 implanted children, 8 (6.5%) children showed anesthetic complications. CI surgery is one of the main causes of hospital readmission due to otologic surgeries (Kalejaiye et al., 2017; Kim et al., 2017). According to Loundon et al. (2010), 43 of 434 (9.9%) children with CIs experienced major or minor surgical complications. A total of 24 (5.5%) children experienced major complications, such as severe cutaneous

infection, magnet displacement, meningitis, cholesteatoma, cerebrospinal fluid leakage, and electrode misplacement; 19 (4.4%) children had minor complications, such as vertigo, soft tissue infection, persistent otitis media, and temporal facial palsy. Major complications can lead to serious medical conditions and in some cases, to major surgical revision (Hansen et al., 2010).

According to previous literature, CI revision surgery (re-implantation of a CI device) can occur in 2.9% to 12.9% of children (Brown et al., 2009; Eskander et al., 2011; Lescanne et al., 2011; Marlowe et al., 2009; Sorrentino et al., 2009). Causes of revision surgery include device failure (which is the most common cause), device upgrading and medical issues, such as infection (Blanchard et al., 2015; Brown et al., 2009; Lassig et al., 2005; Wang et al., 2014). Several cohort studies have found that revision rates are higher for children under the age of 15 because of device failure (Côté et al., 2007; Migirov et al., 2006; Sorrentino et al., 2009). Identifying device failure can be challenging in younger children because they may not be able to express perceived changes in hearing. Delay in identifying difficulties may bring negative consequences related to language development (Blanchard et al., 2015).

Loss of residual hearing following CI surgery is another important clinical consideration when making decisions for children with hearing loss. This is especially applicable to children with sufficient hearing to access sound through HAs. In the past decade, a soft surgery CI technique has been widely used to preserve hearing in low frequencies (Buechner et al., 2015; Lenarz et al., 2009; Santa Maria et al., 2014; Snels et al., 2019). Although current CI technology and surgical techniques (e.g., soft surgery) are often successful at preserving residual hearing, the possibility of losing hearing still exists irrespective of the surgical technique, electrode length, and depth of insertion (Brown et al., 2010; Kopelovich et al., 2015; Meredith et al., 2017; Snels et al., 2019). According to Zanetti et al. (2015), the preservation of residual hearing was

successful in only 61 of 155 (39%) patients (82 children and 73 adults) who were implanted using a soft surgery technique. Due to these risk factors, implanting children with hearing loss is still a difficult decision and causes concerns for parents.

CI candidacy criteria for children with residual hearing

As more children receive CIs, and their benefits are documented, candidacy criteria have expanded in clinical practice. Multiple studies have emphasized expanding candidacy criteria to include children who have residual hearing loss. The FDA recognizes that CI programs may determine that some children who do not meet the traditional indications for audiological candidacy can benefit from CI. The FDA document entitled "Off-Label and Investigational Use of Marketed Drugs, Biologics, and Medical Devices" authorizes 'off-label' uses of medical devices in clinical practice (Gifford, 2011, 2012, 2016; FDA, 2011). With the help of this document, CI specialists recommend CIs when their assessment and clinical judgment indicate that children who have usable residual hearing may benefit from the surgery.

There is increasing attention on CI surgery for children with residual hearing in the literature (Fitzpatrick et al., 2009; Gratacap et al., 2015; Hughes et al., 2014; Leigh et al., 2011, 2016). Studies have shown that audiometric candidacy criteria are variable in clinical practice (de Kleijn et al., 2018; Dowell et al., 2004; Fitzpatrick et al., 2009). In the U.S., according to Hughes et al. (2014), children whose speech perception scores are better than the FDA-approved criteria, or who have audiometric thresholds that are better than a severe-profound hearing loss, are off-label CI candidates. In Australia, an evidence-based guideline recommends that implantation be considered for children with unaided PTA hearing levels in the range of 65–80 dB HL, because these children can receive more speech-related benefits from CIs than from HAs

(Leigh et al., 2016). In addition, a recent systematic review (de Kleijn et al., 2018) that included studies from multiple countries reported that CI was recommended for children who have lower thresholds (≥ 80dB HL) at 4-frequency (500, 1000, 2000, and 4000 Hz) based on speech perception and auditory performance subtests. According to a recent survey in the U.S. (Carlson et al., 2018), 63 of 81 (78%) surgeons performed CI surgery for off-label or non-traditional indications in children and adult populations. Of these surgeons, 21 (27%) reported that it would be useful to have FDA approval of CI candidacy for people who have greater degrees of residual hearing.

Benefits of CI in children with residual hearing

An increasing number of studies of children who are outside typical CI candidacy criteria have reported positive outcomes in a number of areas, including improvements in speech, language, and auditory functioning (Chiossi & Hyppolito, 2017; Dettman et al., 2004; Fitzpatrick et al., 2006; Gratacap et al., 2015; Hughes et al., 2014). Several studies have also reported that positive speech-related outcomes after CIs in children with hearing loss are closely associated with younger age at CI and the having a higher amount of preoperative residual hearing (Leigh et al., 2011, 2016).

According to a recent systematic review (Chiossi & Hyppolito, 2017), children with residual hearing obtain positive language-related outcomes after CIs. In a recent study (Michael et al., 2019), children with CIs who had residual hearing also showed significantly lower hyperactivity/inattention and higher pro-social behaviour scores compared to children with severe to profound hearing loss who use HAs. In addition, the development of CI technology for preserving residual hearing in low frequencies (Carlson et al., 2015, 2018; Eshraghi et al., 2017;

Skarzynski, 2012; Sweeney et al., 2015, 2016; Zanetti et al., 2015) has led to improved hearing abilities and speech perception in noise, overall natural sound and music quality, and sound localization by combining acoustic and electronic stimulation (Adunka et al., 2008, 2013; Dunn et al., 2010; Moteki et al., 2017).

An increasing number of children with residual hearing are being considered for CIs (Carlson et al., 2018; Teagle et al., 2019). This is due to the significant improvements in hearing that have been reported for children with residual hearing who have received CIs and the advancements in CI technology. However, the evidence of benefits beyond speech perception such as language, cognition, social-emotional functioning, and academic achievement is still limited. In addition, CI guidelines and the long-term prognosis of outcomes for children with residual hearing are unclear (Heman-Ackah et al., 2012; O'Brien et al., 2010, 2012). For these reasons, parents may experience decisional conflict when making decisions about CI surgery for children with residual hearing.

Parental decision-making in considering cochlear implantation

Decision-making about CI is a considerable challenge for parents of children with hearing loss (Duncan, 2009; Incesulu et al., 2003; Li et al., 2004; Most & Zaidman-Zait, 2001; Sorkin & Zwolan, 2008). Parents of all children with hearing loss are faced with a number of difficult decisions shortly after birth (Decker et al., 2012; Kluwin & Stewart, 2000; Li et al., 2003). Since the implementation of UNHS programs, parents often learn about their child's hearing loss in the first few months of life and need to make decisions about interventions when their child is still very young (Hardonk et al., 2010; McCracken et al., 2008; Young & Andrews, 2001; Young & Tattersall, 2007).

Given the importance of the early years for language acquisition, parents may experience stress and feel pressed for time during the CI decision-making process. According to Incesulu et al. (2003), 22 of 27 (81%) parents responded that CI decision-making was one of the most stressful steps for them because individual outcomes cannot be accurately predicted. The decision has consequences that affect the children's entire lives (Bruin & Ohna, 2015; Crowe et al., 2014; Hyde et al., 2010b; Li et al., 2003, 2004; Punch & Hyde, 2011). In Canada, there are also newcomers (e.g., immigrant and refugee families) whose children may experience late identification of hearing loss because they may not have had access to newborn hearing screening in their country of origin. In this case, parents and children may feel pressure to make a decision with limited time to absorb all the information.

Understanding information about the benefits and risks of CIs is also an important factor in the decision-making process for parents (Duncan, 2009; Li et al., 2003, 2004). A study in the United Kingdom (Athalye et al., 2015) used qualitative methods to explore the perceptions of any CI users (age range 10 to 88), parents, and professionals. Parents and users with CIs wanted more liaison, exchange of information and detailed reports about individual cases, and collaboration with audiology clinics during the decision-making process. Parents and users indicated they wanted to be more involved in the CI decision-making process. However, they felt their involvement was limited because of the lack of shared information between them and the practitioners. Therefore, they suggested using plain English for the information tools and sharing information about updated current CI technology between parents and practitioners. In the U.S., Stewart (2014) presented the results of a survey about parents' experiences and perspectives regarding the CI decision-making process for their school-aged implanted children. Of 50 parents in the study, 44 (88%) indicated that they received information about CIs from

audiologists, particularly during the decision-making process. Parents also reported having searched for information from as many places and people as possible to make well-informed decisions regarding CIs for their children.

According to previous qualitative interviews, parents feel more pressure when they make a CI decision if their children have usable hearing and benefit from HAs (Fitzpatrick et al., 2006; Hyde et al., 2010b), and decision-making can be more complex. Practitioners also hesitate to decide on implantation when children with residual hearing have already received benefits and had usable spoken language function with HAs (Fitzpatrick et al., 2009). For instance, a retrospective study in the United Kingdom reported that 28 children (35 ears) with low-frequency residual hearing (age range 13 months to 12 years) demonstrated more positive clinical outcomes after surgery than they had expected before surgery (Wilson et al., 2016). However, at pre-surgery, parents were more focused on what 'hearing' their child could lose.

Additionally, practitioners are not always confident in recommending CIs for children with residual hearing and take several factors into consideration, such as degree of hearing loss, language development, ease of listening, and functioning at school (Fitzpatrick et al., 2009; Gratacap et al., 2015; Wilson et al., 2016). Wilson and colleagues (2016) reported that when children show usable residual hearing, it is a challenge for practitioners to determine whether they will benefit more from a CI. The lack of scientific evidence about CI candidacy and specific guidelines regarding residual hearing increases the difficulty of the decision-making process (Gratacap et al., 2015). Given the complexity of the process compared to that of typical CI candidates, more research is required to explore the decision-making needs of children, parents, and practitioners.

Shared decision-making in pediatric healthcare

People commonly experience difficulties in making an appropriate decision when there is more than one reasonable option; this can lead to delays in decision-making (O'Connor, 1995; O'Connor & Edwards, 2009). SDM is an evidence-based approach in which families and practitioners work together to make healthcare decisions (Gabe et al., 2004; Légaré et al., 2011). Through this, families and practitioners can decide together the appropriate intervention approach by sharing medical and clinical information and information about the families' preferences and values (Légaré et al., 2011; Makoul & Clayman, 2006). SDM has been widely applied to increase families' participation in the decision-making process and to guide families in making informed and value-sensitive decisions (Elwyn et al., 2013; Légaré et al., 2014).

However, few studies exist on pediatric decision-making, and limited evidence-based practices regarding the impacts of interventions are available to support SDM with pediatric populations (Feenstra et al., 2014, 2015; Strauss et al., 2015; Wyatt et al., 2015). Studies suggest that decision support interventions need to include children's views in decisions related to their own health (Coyne et al., 2014; Feenstra et al., 2014; Hinds et al., 2005; Lipstein et al., 2015). Recently, Adams and Levy (2017) pointed out that practitioners should involve older children in decision-making and help children to understand options. However, children's participation in health-related decision-making is dependent on their cognitive capabilities and personalities (Franck & Callery, 2004; Gabe et al., 2004; Garnett et al., 2016; Kodish, 2003). Therefore, in some cases, the core decision-makers are the parents, depending on the competency level of their children (Madrigal et al., 2012; Miller & Nelson, 2012; Stewart et al., 2005).

There are numerous factors to consider for children and their families confronted with the SDM process. For example, the children's developmental stages and their abilities, the time

frame for decision-making, the severity of the health-related condition, the acuity or chronicity of primary conditions, and the presence of comorbidities can impede interactions between children, their parents, and practitioners (Zajicek-Farbe et al., 2015). A systematic review, which targeted children less than 18 years of age and their caregivers, found that parents experience challenges in balancing their personal knowledge, emotions, and beliefs. In addition, parents expressed wanting to share those difficulties in the decision-making process with the practitioners and with their children (Wyatt et al., 2015). Evolving over time, there has been a growth of service delivery models that have moved from a practitioner-centred framework to a more family-centred approach, engaging clients to participate in health-related decisions using SDM.

In audiology, to date, no studies have explored the use of SDM during the decision-making process between parents and practitioners (Porter et al., 2018); however, a few studies in pediatric audiology have mentioned SDM (Johnston, 2008, 2009; O'Connor et al., 2008; Li et al., 2003, 2004). For example, Johnston et al. (2009) developed a decision aid for parents of children with unilateral CIs who were interested in obtaining a second CI. It was developed based on the results of a needs assessment which indicated that parents required additional information on the risks and benefits of bilateral CIs (Johnston et al., 2008). There has been growing interest in using SDM and decision-making in audiology, and the approach has been advocated to assist families in making evidence-informed choices (Boisvert et al., 2017). In this context, through the SDM process, clinicians can guide processes to elicit the preferences of families of children with residual hearing in CI decision-making, therefore aligning their care with family-centred and evidence-based practices (Hoffmann et al., 2014). To apply SDM in

parental CI decision-making for children with residual hearing, it is necessary to collect evidence about these children with residual hearing.

In order to assist parents and clinicians in making decisions about CIs, it is important to conduct a comprehensive research study about the characteristics and outcomes of the children with residual hearing who receive CIs and the benefits and risks of the intervention options, as well as the decision-making experiences of families and practitioners.

Conceptual framework

The Ottawa Decision Support Framework (ODSF) (O'Connor et al., 1998) was applied as the framework to guide the studies. The ODSF (see Figure 1) is a commonly used framework developed by the Ottawa Hospital Research Institute (OHRI) that guides practitioners and to assess patient's decisional needs, provide decision support interventions, and evaluate their effects on decisional outcomes (O'Connor et al., 1998; Feenstra et al., 2014; Légaré et al., 2006a, 2006b). It is appropriate for health-related decisions that: "1) are stimulated by a new circumstance, diagnosis, or developmental transition, 2) require careful deliberation because of the uncertain and/or value-sensitive nature of the benefits and risks, and 3) need relatively more effort during the deliberation phase than the implementation phase" (O'Connor et al., 1998). This framework consists of three elements: 1) assessment of needs, 2) decision support interventions, and 3) evaluation of the decision-making process and outcomes of decisions (O'Connor et al., 1998).

In a decision support intervention for children with residual hearing and their parents who are confronting CI decision-making, it is important to explore families' decisional needs including the children's characteristics and situations of the families. Decisional needs include

factors that influence decision-making such as elements of the decision (type, timing, stage, and leaning), clinical characteristics, decisional conflict, available options, realistic expectations, and values. A collaborative approach between CI practitioners and decision-makers is important during the decision-making process.

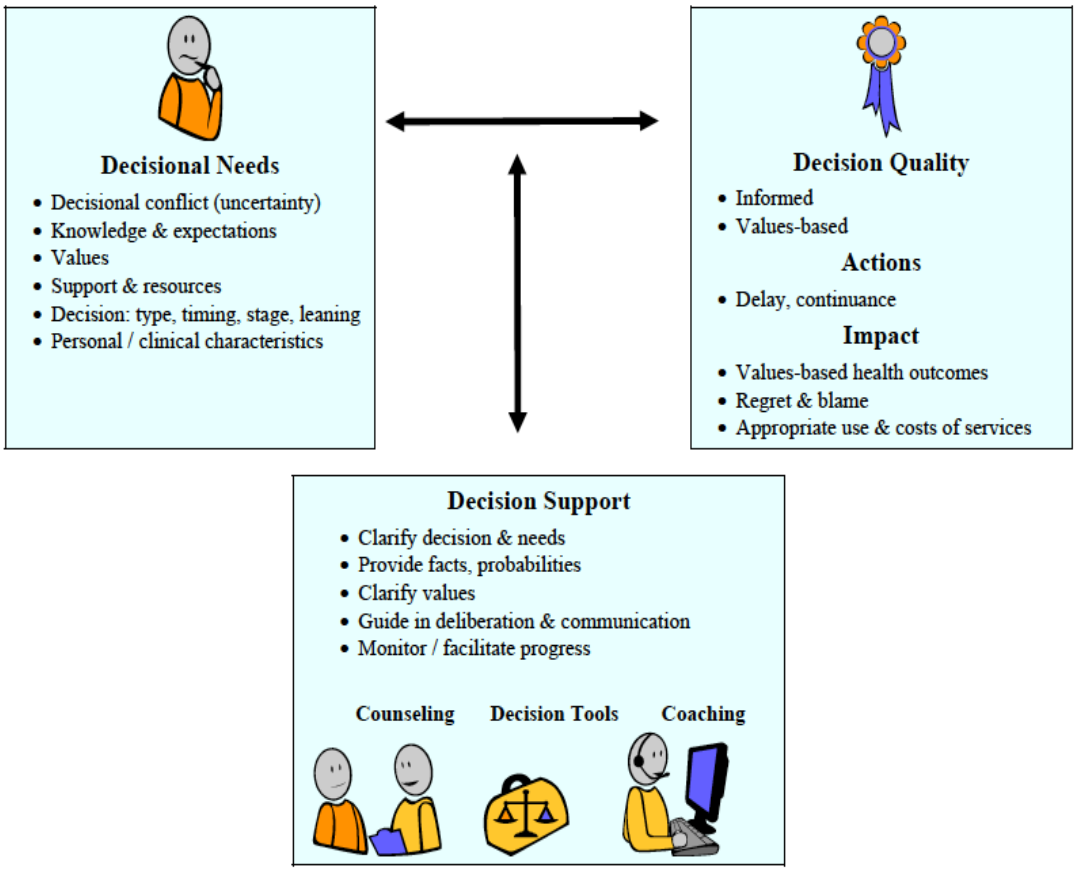


Figure 1. Ottawa Decision Support Framework

This framework is useful for describing factors that influence CI practitioners' decisions, decreasing parents' decisional conflicts, and to integrating their values, preferences, and needs (O'Connor et al., 1998). Each of these will be explored for children with residual hearing and their parents in the process of making a CI decision. This thesis places emphasis on describing

the decisional needs of children with residual hearing, their parents, and practitioners, and on collecting evidence to facilitate SDM. A better understanding of the factors influencing parents' decisions and their needs can help determine optimal decision support interventions for families.

Objectives

Given the complexity of the decision-making process for children with residual hearing who are considered for a CI, a better understanding of decision support needs is required. However, there is very limited information about CI decision-making to assist the parents and practitioners of this population of children (Porter et al., 2018). It is important therefore to accumulate information about the clinical characteristics of children with residual hearing who have received CIs, the benefits and the risks of the technology options (CIs and HAs), and the perspectives of parents and practitioners.

The overall purpose of this thesis is to examine the characteristics and outcomes of children with residual hearing who received CIs and to better understand the decision-making experiences of families and practitioners. The objectives of this research were to: 1) explore the clinical characteristics and outcomes of children with residual hearing who received CIs; 2) summarize the evidence about the benefits and risks of CIs compared to HAs in children with residual hearing; and 3) explore the decision-making process and needs from the perspectives of parents and practitioners.

Methodology

Table 2 shows an overview of each chapter's methodologies and objectives. To address the first objective, a retrospective medical chart review was performed (Chapter 2). A systematic review was conducted to review and summarize the evidence about the benefits and risks of CIs compared to HAs (Chapter 3). The third objective was addressed by conducting interviews with parents (Chapter 4) and practitioners (Chapter 5).

Table 2. Study methodologies and objectives

Chapter	Study design	Methodology	Objectives
2	Retrospective chart review	Quantitative	To explore the clinical characteristics and outcomes of children with residual hearing who received CIs
3	Systematic review	Qualitative	To summarize the evidence about the benefits and risks of CIs compared to HAs in children with residual hearing
4	Parent interviews	Qualitative	To explore the decision-making process and needs from the perspectives of parents and practitioners
5	Practitioner interviews	Qualitative	

CI, cochlear implant; HA, hearing aid

Chapter 2. To explore the clinical characteristics and outcomes of children with residual hearing who received CIs (Objective 1)

A retrospective chart review was conducted in a tertiary care pediatric CI center in the capital region of Canada which provides CI surgical and audiological follow-up services. The clinical data set for this study included all children who received CIs over a 26-year period from 1992 to

2018. Audiological assessments, including speech perception test data, were also extracted from the medical charts.

Chapter 3. To summarize the evidence about the benefits and risks of CIs compared to HAs in children with residual hearing (Objective 2)

A systematic review that investigated the benefits and risks of pediatric CIs was employed. The aim of our systematic review was to review and synthesize the benefits and risks of CIs compared to those of HAs in children who have residual hearing. The research questions addressed in this review were: 1) What are the benefits for children with preoperative residual hearing who received CIs, compared to their pre-implant outcomes or compared to children with HAs? and 2) What are the risks and negative outcomes among children with residual hearing who received CIs?

Chapter 4. To explore the decision-making process and needs from the perspectives of parents (Objective 3)

A qualitative research design was employed to better understand the CI decision-making process of parents. An interview guide (Appendix F) was developed based on the Decisional Needs Assessment in Populations (Jacobsen et al., 2013). The interview guide was reviewed by two senior researchers to adapt the questions to the context and topic of the study. Semi-structured individual interviews were applied to obtain in-depth information from parents of children with residual hearing. Participants were asked open-ended questions to encourage them to share their perspectives on experiences with the CI decision-making process.

Chapter 5. To explore the decision-making process and needs from the perspectives of practitioners (Objective 3)

Four qualitative focus groups and an individual interview were conducted with two groups of practitioners: one group was recruited from hospital-based CI programs and the other from local school boards. An interview guide (Appendix F) was developed to explore practitioners ' experiences and perspectives in supporting CI decision-making by parents of children with residual hearing. A semi-structured format was selected to guide the interviews. The interviewer asked questions to encourage practitioners to share their experiences about the process of supporting decision-making for parents and children, as well as their needs in supporting parents.

Chapter 6. Discussion

The final chapter summarizes and integrates the findings of the overall thesis project. The results are interpreted in relation to the conceptual framework that guided this project. In addition, Chapter 6 concludes with suggestions for future research.

References

Adams, R. C., & Levy, S. E. (2017). Shared decision-making and children with disabilities: Pathways to consensus. *Pediatrics*, *139*(6), e20170956. https://doi.org/10.1542/peds.2017-0956

Adunka, O. F., Buss, E., Clark, M. S., Pillsbury, H. C., & Buchman, C. A. (2008). Effect of preoperative residual hearing on speech perception after cochlear implantation. *Laryngoscope*, *118*(11), 2044–2049. https://doi.org/10.1097/MLG.0b013e3181820900

Adunka, O. F., Dillon, M. T., Adunka, M. C., King, E. R., Pillsbury, H. C., & Buchman, C. A. (2013). Hearing preservation and speech perception outcomes with electric-acoustic stimulation after 12 months of listening experience. *Laryngoscope*, *123*(10), 2509–2515. https://doi.org/10.1002/lary.23741

Akinpelu, O. V, Peleva, E., Funnell, W. R. J., & Daniel, S. J. (2014). Otoacoustic emissions in newborn hearing screening: A systematic review of the effects of different protocols on test outcomes. *Int J Pediatr Otorhinolaryngol*, *78*(5), 711–717. https://doi.org/10.1016/j.ijporl.2014.01.021

American Academy of Pediatrics, Joint Committee on Infant Hearing (2007). Year 2007 position statement: Principles and guidelines for early hearing detection and intervention programs. *Pediatrics*, *120*(4), 898–921. https://doi.org/10.1542/peds.2007-2333

Anmyr, L., Larsson, K., & Olsson, M. (2016). Parents 'stress and coping related to children's use of a cochlear implant: A qualitative study. *J Soc Work Disabil Rehabil*, *15*(2), 150–167. https://doi.org/10.1080/1536710X.2016.1162123

Antia, S. D., Jones, P. B., Reed, S., & Kreimeyer, K. H. (2009). Academic status and progress of deaf and hard-of-hearing students in general education classrooms. *J Deaf Stud Deaf Educ*,

14(3), 293–311. https://doi.org/10.1093/deafed/enp009

Antia, S. D., Kreimeyer, K. H., Metz, K. K., & Spolsky, S. (2012). *Peer interactions of deaf and hard-of-hearing children.* (M. Marschark & P. E. Spencer, Eds.), *The Oxford Handbook of Deaf Studies, Language, and Education* (2nd ed.). Oxford University Press, USA. https://doi.org/10.1093/oxfordhb/9780199750986.013.0013

Armstrong, M., Maresh, A., Buxton, C., Craun, P., Wowroski, L., Reilly, B., & Preciado, D. (2013). Barriers to early pediatric cochlear implantation. *Int J Pediatr Otorhinolaryngol*, *77*(11), 1869–1872. https://doi.org/10.1016/j.ijporl.2013.08.031

Arnoldner, C., Lin, V. Y., Honeder, C., Shipp, D., Nedzelski, J., & Chen, J. (2014). Ten-year health-related quality of life in cochlear implant recipients: Prospective SF-36 data with SF-6D conversion. *Laryngoscope*, *124*(1), 278–282. https://doi.org/10.1002/lary.24387

Athalye, S., Archbold, S., Mulla, I., Lutman, M., & Nikolopoulous, T. (2015). Exploring views on current and future cochlear implant service delivery: The perspectives of users, parents and professionals at cochlear implant centres and in the community. *Cochlear Implants Int*, *16*(5), 241–253. https://doi.org/10.1179/1754762815Y.0000000003

Bagatto, M., Scollie, S. D., Hyde, M., & Seewald, R. (2010). Protocol for the provision of amplification within the Ontario Infant hearing program. *Int J Audiol*, *49*(SUPPL. 1), S70–S79. https://doi.org/10.3109/14992020903080751

Barker, E. J., & Briggs, R. J. (2009). Cochlear implantation in children with keratitis-ichthyosis-deafness (KID) syndrome: outcomes in three cases. *Cochlear Implants Int*, *10*(3), 166–173. https://doi.org/https://dx.doi.org/10.1179/cim.2009.10.3.166

Bat-Chava, Y., Martin, D., & Kosciw, J. G. (2005). Longitudinal improvements in communication and socialization of deaf children with cochlear implants and hearing aids:

evidence from parental reports. *J Child Psychol Psychiatry*, *46*(12), 1287–1296. https://doi.org/10.1111/j.1469-7610.2005.01426.x

Bell, N., Angwin, A. J., Wilson, W. J. & Arnott, W. L. (2019). Reading development in children with cochlear implants who use spoken language: A psycholinguistic investigation. *J Speech Lang Hear Res. 62*(2), 456-469. https://doi.org/10.1044/2018_JSLHR-H-17-0469

Benasich, A. A., Thomas, J. J., Choudhury, N., & Leppänen, P. H. (2002). The importance of rapid auditory processing abilities to early language development: Evidence from converging methodologies. *Dev Psychobiol. 40*(3), 278–292.

Bittencourt, A. G., Torre, A. A. G. Della, Bento, R. F., Tsuji, R. K., & Brito, R. de. (2012). Prelingual deafness: Benefits from cochlear implants versus conventional hearing aids. *Int Arch Otorhinolaryngol*, *16*(3), 387–390. https://dx.doi.org/10.7162/S180999777201200300014

Black, I. M., Bailey, C. M., Albert, D. M., Leighton, S. E. J., Hartley, B. E. J., Chatrath, P., & Patel, N. (2007). The Great Ormond Street Hospital paediatric cochlear implant programme 1992-2004: A review of surgical complications. *Cochlear Implants Int*, *8*(2), 53–67. https://doi.org/10.1002/cii.330

Blamey, P. J., Sarant, J. Z., Paatsch, L. E., Barry, J. G., Bow, C. P., Wales, R. J., Wright, M., Psarros, C., Rattigan, K., & Tooher, R. (2001). Relationships among speech perception, production, language, hearing loss, and age in children with impaired hearing. *J Speech Lang Hear Res*, *44*(April), 264–285. https://doi.org/10.1044/1092-4388(2001/022)

Blanchard, M., Thierry, B., Glynn, F., De Lamaze, A., Garabedian, E. N., & Loundon, N. (2015). Cochlear implant failure and revision surgery in pediatric population. *Ann Otol Rhinol*

Laryngol, *124*(3), 227–231. https://doi.org/10.1177/0003489414551931

Boisvert, I., Clemesha, J., Lundmark, E., Crome, E., Barr, C., & McMahon, C. M. (2017). Decision-making in audiology: Balancing evidence-based practice and patient-centered care. *Trends Hear*, *21*, 233121651770639. https://doi.org/10.1177/2331216517706397

Borton, S. A., Mauze, E., & Lieu, J. E. C. (2010). Quality of life in children with unilateral hearing loss: A pilot study. *Am J Audiol*, *19*(1), 61–72. https://doi.org/10.1044/1059-0889(2010/07-0043)

Brown, K. D., Connell, S. S., Balkany, T. J., Eshraghi, A. E., Telischi, F. F., & Angeli, S. A. (2009). Incidence and indications for revision cochlear implant surgery in adults and children. *Laryngoscope*, *119*(1), 152–157. https://doi.org/10.1002/lary.20012

Brown, R. F., Hullar, T. E., Cadieux, J. H., & Chole, R. A. (2010). Residual hearing preservation after pediatric cochlear implantation. *Otol Neurotol*, *31*(8), 1221–1226. https://doi.org/10.1097/MAO.0b013e3181f0c649

Bruin, M., & Ohna, S. E. (2015). Negotiating reassurance: parents 'narratives on follow-up after cochlear implantation. *Eur J Spec Needs Educ*, *30*(4), 518–534. https://doi.org/10.1080/08856257.2015.1046741

Buechner, A., Vaerenberg, B., Gazibegovic, D., Brendel, M., Ceulaer, G. De, Govaerts, P., & Lenarz, T. (2015). Evaluation of the 'Fitting to Outcomes eXpert' (FOX ®) with established cochlear implant users. *Cochlear Implants Int*, *16*(1), 39–46. https://doi.org/10.1179/1754762814Y.0000000085

Burger, T., Spahn, C., Richter, B., Eissele, S., Lohle, E., & Bengel, J. (2005). Parental distress: the initial phase of hearing aid and cochlear implant fitting. *Am Ann Deaf*, *150*(1), 5–10. https://doi.org/10.1353/aad.2005.0017

Carlson, M. L., O'Connell, B. P., Lohse, C. M., Driscoll, C. L., & Sweeney, A. D. (2018). Survey of the American Neurotology Society on cochlear implantation. *Otol Neurotol*, *39*(1), e12–e19. https://doi.org/10.1097/MAO.0000000000001631

Carlson, M. L., Sladen, D. P., Haynes, D. S., Driscoll, C. L., DeJong, M. D., Erickson, H. C., Sunderhaus, L. W., Hedley-Williams, A., Rosenzweig, E. A., Davis, T. J., & Gifford, R. H. (2015). Evidence for the expansion of pediatric cochlear implant candidacy. *Otol Neurotol*, *36*(1), 43–50. https://doi.org/10.1097/MAO.0000000000000607

Charles, C., Gafni, A., & Whelan, T. (1997). Shared decision-making in the medical encounter: what does it mean? (or it takes at least two to tango). *Soc Sci Med*, *44*(5), 681–692.

Ching, T. Y. C., Crowe, K., Martin, V., Day, J., Mahler, N., Youn, S., Street, L., Cook, C., & Orsini, J. (2010). Language development and everyday functioning of children with hearing loss assessed at 3 years of age. *Int J Speech Lang Pathol*, *12*(2), 124–131.

Ching, T. Y. C., Zhang, V. W., Flynn, C., Burns, L., Hou, S., Mcghie, K., Buynder, P. Van. (2017). Factors influencing speech perception in noise for 5-year-old children using hearing aids or cochlear implants. *Int J Audiol*, *0*(0), 1–11. https://doi.org/10.1080/14992027.2017.1346307

Chiossi, J. S. C., & Hyppolito, M. A. (2017). Effects of residual hearing on cochlear implant outcomes in children: A systematic-review. *Int J Pediatr OtorhiInt J Pediatr Otorhinolaryngol*, *100*, 119–127. https://doi.org/10.1016/j.ijporl.2017.06.036

Chorney, J., Haworth, R., Graham, M. E., Ritchie, K., Curran, J. A., & Hong, P. (2015). Understanding shared decision making in pediatric otolaryngology. *Otolaryngol Head Neck Surg*, *152*(5), 941–947. https://doi.org/10.1177/0194599815574998

Chundu, S., & Flynn, S. L. (2014). Audiogram and cochlear implant candidacy – UK

perspective. *Cochlear Implants Int*, *15*(4), 241–244. https://doi.org/10.1179/1754762813Y.0000000052

Chute, P. M., & Nevins, M. E. (2002). *The parents 'guide to cochlear implants*. Gallaudet University Press.

Côté, M., Ferron, P., Bergeron, F., & Bussières, R. (2007). Cochlear reimplantation: Causes of failure, outcomes, and audiologic performance. *Laryngoscope*, *117*(7), 1225–1235. https://doi.org/10.1097/MLG.0b013e31805c9a06

Coyne, I., Amory, A., Kiernan, G., & Gibson, F. (2014). Children's participation in shared decision-making: Children, adolescents, parents and healthcare professionals 'perspectives and experiences. *Eur J Oncol Nurs*, *18*(3), 273–280. https://doi.org/10.1016/j.ejon.2014.01.006

Crowe, K., Fordham, L., Mcleod, S., & Ching, T. Y. C. (2014). 'Part of Our World': Influences on caregiver decisions about communication choices for children with hearing loss. *Deafness Educ Int*, *16*(2), 61–85. https://doi.org/10.1179/1557069X13Y.0000000026

Dammeyer, J., Chapman, M., & Marschark, M. (2018). Experience of hearing loss, communication, social participation, and psychological well-being among adolescents with cochlear implants. *Am Ann Deaf*, *163*(4), 424–439. https://doi.org/10.1353/aad.2018.0027

de Kleijn, J. L., van Kalmthout, L. W. M., van der Vossen, M. J. B., Vonck, B. M. D., Topsakal, V., & Bruijnzeel, H. (2018). Identification of pure-tone audiologic thresholds for pediatric cochlear implant candidacy: A systematic review. *JAMA Otolaryngol Head Neck Surg*, *144*(7), 630–638. https://doi.org/https://dx.doi.org/10.1001/jamaoto.2018.0652

De Leenheer, E. M. R., Janssens, S., Padalko, E., Loose, D., Leroy, B. P., & Dhooge, I. J. (2011). International journal of pediatric otorhinolaryngology etiological diagnosis in the hearing-

impaired newborn: Proposal of a flow chart. *Int J Pediatr Otorhinolaryngol*, *75*(1), 27–32. https://doi.org/10.1016/j.ijporl.2010.05.040

Decker, K. B., Vallotton, C. D., & Johnson, H. a. (2012). Parents 'communication decision for children with hearing loss: sources of information and influence. *Am Ann Deaf*, *157*(4), 326–339. https://doi.org/10.1353/aad.2012.1631

Dettman, S. J., D'Costa, W. A., Dowell, R. C., Winton, E. J., Hill, K. L., & Williams, S. S. (2004). Cochlear implants for children with significant residual hearing. *Arch Otolaryngol Head Neck Surg*, *130*(5), 612. https://doi.org/10.1001/archotol.130.5.612

Dillon, B., & Pryce, H. (2020). What makes someone choose cochlear implantation? An exploration of factors that inform patient decision-making. *Int J Audiol*, *59*(1), 24–32. https://doi.org/10.1080/14992027.2019.1660917 LK -

Dowell, C., Hollow, R., & Winton, E. (2004). Outcomes for cochlear implant users with significant residual hearing. *Arch Otolaryngol Head Neck Surg*, *130*(May), 575–581. https://doi.org/10.1001/archotol.130.5.575

Duncan, J. (2009). Parental readiness for cochlear implant decision-making. *Cochlear Implants Int*, *10*(1), 38–42. https://doi.org/10.1002/cii.384

Dunn, C. C., Perreau, A., Gantz, B., & Tyler, R. S. (2010). Benefits of localization and speech perception with multiple noise sources in listeners with a short-electrode cochlear implant. *JAAA*, *21*(1), 44–51. https://doi.org/10.3766/jaaa.21.1.6

Dye, M. W. G., Hauser, P. C., & Bavelier, D. (2009). Is visual selective attention in deaf individuals enhanced or deficient? The case of the useful field of view. *PLoS ONE*, *4*(5), e5640. https://doi.org/10.1371/journal.pone.0005640

Edwards, A., & Elwyn, G. (2009). Shared decision-making in health care: Achieving evidence-

based patient choice. In A. Edwards & G. Elwyn (Eds.), *Oxford University Press* (illustrate). OUP Oxford, 2009. https://doi.org/10.1007/s13398-014-0173-7.2

Elwyn, G., Lloyd, A., Joseph-Williams, N., Cording, E., Thomson, R., Durand, M. A., & Edwards, A. (2013). Option grids: Shared decision making made easier. *Patient Educ Couns*, *90*(2), 207–212. https://doi.org/10.1016/j.pec.2012.06.036

Eshraghi, A. A., Ahmed, J., Krysiak, E., Ila, K., Ashman, P., Telischi, F. F., Angeli, S. A., Prentiss, S., Martinez, D., & Valendia, S. (2017). Clinical, surgical, and electrical factors impacting residual hearing in cochlear implant surgery. *Acta Oto-Laryngologica*, *137*(4), 384–388. https://doi.org/10.1080/00016489.2016.1256499

Eskander, A, Gordon, K., Kadhim, L., Papaioannou, V., Cushing, S. L., James, A. L., & Papsin, B. C. (2011). Low pediatric cochlear implant failure rate. *Arch Otolaryngol Head Neck Surg*, *137*(12), 1190–1196. https://doi.org/10.1001/archoto.2011.200

Eskander, Antoine, & Papsin, B. (2014). Screening infants for hearing impairment in Canada. *CMAJ*, *186*(14), 1048–1049. https://doi.org/10.1503/cmaj.131685

Farinetti, A., Ben Gharbia, D., Mancini, J., Roman, S., Nicollas, R., & Triglia, J. M. (2014). Cochlear implant complications in 403 patients: Comparative study of adults and children and review of the literature. *Eur Ann Otorhinolaryngol Head Neck Dis*, *131*(3), 177–182. https://doi.org/10.1016/j.anorl.2013.05.005

Feenstra, B., Boland, L., Lawson, M. L., Harrison, D., Kryworuchko, J., Leblanc, M., & Stacey, D. (2014). Interventions to support children's engagement in health-related decisions: A systematic review. *BMC Pediatr*, *14*(1), 109. https://doi.org/10.1186/1471-2431-14-109

Feenstra, B., Lawson, M. L., Harrison, D., Boland, L., & Stacey, D. (2015). Decision coaching using the Ottawa family decision guide with parents and their children: A field testing

study. *BMC Med Inform Decis Mak*, *15*(1), 5. https://doi.org/10.1186/s12911-014-0126-2

Fernald, A., Marchman, V., & Weisleder, A. (2012). SES differences in language processing skill and vocabulary are evident at 18 months. *Dev. Sci*, *16*(2), 234–248. https://doi.org/10.1111/desc.12019

Fitzpatrick, E. M., McCrae, R., & Schramm, D. (2006). A retrospective study of cochlear implant outcomes in children with residual hearing. *BMC Ear Nose Throat Disord*, *6*(7), 1–6. https://doi.org/10.1186/1472-6815-6-7

Fitzpatrick, E. M., Olds, J., Durieux-Smith, A., McCrae, R., Schramm, D., & Gaboury, I. (2009). Pediatric cochlear implantation: How much hearing is too much? *Int J Audiol*, *48*(2), 91–97. https://doi.org/10.1080/14992020802516541

Fitzpatrick, E. M., Olds, J., Gaboury, I., McCrae, R., Schramm, D., & Durieux-Smith, A. (2012). Comparison of outcomes in children with hearing aids and cochlear implants. *Cochlear Implants Int*, *13*(1), 5–15. https://doi.org/https://dx.doi.org/10.1179/146701011X12950038111611

Flexer, C. (2011). Cochlear implants and neuroplasticity: Linking auditory exposure and practice. *Cochlear Implants Int*, *12 Suppl 1*(July), S19–S21. https://doi.org/10.1179/146701011X13001035752255

Franck, L. S., & Callery, P. (2004). Re-thinking family-centred care across the continuum of children's healthcare. *Child Care Health Dev*, *30*(3), 265–277. https://doi.org/10.1111/j.1365-2214.2004.00412.x

Fulcher, A. N., Purcell, A., Baker, E., & Munro, N. (2015). Factors influencing speech and language outcomes of children with early identified severe / profound hearing loss: Clinician-identified facilitators and barriers. *Int J Speech Lang Pathol*, *17*(3), 325–333.

https://doi.org/10.3109/17549507.2015.1032351

Gabe, J., Olumide, G., & Bury, M. (2004). “It takes three to tango”: A framework for understanding patient partnership in paediatric clinics. *Soc Sci Med*, *59*(5), 1071–1079. https://doi.org/10.1016/j.socscimed.2003.09.035

Ganek, H., McConkey Robbins, A., & Niparko, J. K. (2012). Language outcomes after cochlear implantation. *Otolaryngol Clin North Am*, *45*(1), 173–185. https://doi.org/https://dx.doi.org/10.1016/j.otc.2011.08.024

Garnett, V., Smith, J., & Ormandy, P. (2016). Child-parent shared decision making about asthma management. *Nurs Child Young People*, *28*(4), 16–22. https://doi.org/10.7748/ncyp.28.4.16.s20

Geers, A. E. (1997). Comparing implants with hearing aids in profoundly deaf children. Otolaryngol Head Neck Surg, 117, 150–154. https://doi.org/10.1016/S0194-5998(97)70167-0

Geers, A. E. (2002). Factors affecting the development of speech, language, and literacy in children with early cochlear implantation. *Lang Speech Hear Serv Sch*, *33*, 172–184. https://doi.org/0161–1461/02/3303–0172

Geers, A. E, Nicholas, J. G., & Sedey, A. L. (2003). Language skills of children with early cochlear implantation. *Ear Hear*, *24*(1S), 46–58. https://doi.org/10.1097/01.AUD.0000051689.57380.1B

Gheorghe, D., & Zamfir-Chiru-Anton, A. (2015). Complications in cochlear implant surgery. *J Med Life*, *8*(3), 329–332.

Gifford, R. H. (2011). Who is a cochlear implant candidate? *Hear J*, *64*(6), 16–18. https://doi.org/10.1097/01.HJ.0000399149.53245.b1

Gifford, R. H. (2012). Journal Club: FDA Indications for pediatric cochlear implantation fail to reflect current research. Hear J. https://doi.org/10.1097/01.HJ.0000421133.83343.d1

Gifford, R. H. (2016). Expansion of pediatric cochlear implant indications. *Hear J, December*, 8–10. https://doi.org/10.1097/01.HJ.0000511125.71672.3e

Gratacap, M., Thierry, B., Rouillon, I., Marlin, S., Garabedian, N., & Loundon, N. (2015). Pediatric cochlear implantation in residual hearing candidates. *Ann Otol Rhinol Laryngol*, *124*(6), 443–451. https://doi.org/10.1177/0003489414566121

Hansen, S., Anthonsen, K., Stangerup, S., Jensen, J. H., Thomsen, J., & Cayé-thomasen, P. E. R. (2010). Unexpected findings and surgical complications in 505 consecutive cochlear implantations: a proposal for reporting consensus. *Acta Oto-Laryngologica*, *130*, 540–549. https://doi.org/10.3109/00016480903358261

Hanvey, K., Ambler, M., Maggs, J., & Wilson, K. (2016). Criteria versus guidelines: Are we doing the best for our paediatric patients? *Cochlear Implants Int*, *17*, 78–82. https://doi.org/10.1080/14670100.2016.1157310

Hardonk, S., Bosteels, S., Desnerck, G., Loots, G., Van Hove, G., Van Kerschaver, E., Vanroelen, C., & Louckx, F. (2010). Pediatric cochlear implantation: A qualitative study of parental decision-making processes in Flanders, Belgium. *Am Ann Deaf*, *155*(3), 339–352. https://doi.org/10.1353/aad.2010.0012

Hardonk, S., Daniels, S., Desnerck, G., Loots, G., Van Hove, G., Van Kerschaver, E., Sigurjónsdóttir, H. B., Vanroelen, C., & Louckx, F. (2011). Deaf parents and pediatric cochlear implantation: An exploration of the decision-making process. *Am Ann Deaf*, *156*(3), 290–304. https://doi.org/10.1353/aad.2011.0027

Hawksworth, C., & Ravury, S. (2015). An audit of anesthesia safety in a pediatric cochlear

implantation program. *Paediatric Anaesthesia*, *25*(6), 630–635. https://doi.org/10.1111/pan.12613

Hawley, K. A., Goldberg, D. M., & Anne, S. (2017). Utility of a multidisciplinary approach to pediatric hearing loss. *Am J Otolaryngol*, *38*(5), 547–550. https://doi.org/10.1016/j.amjoto.2017.05.008

Heman-Ackah, S. E., Roland, J. T., Haynes, D. S., & Waltzman, S. B. (2012). Pediatric cochlear implantation: Candidacy evaluation, medical and surgical considerations, and expanding criteria. *Otolaryngologic Clinics of North America*, *45*(1), 41–67. https://doi.org/10.1016/j.otc.2011.08.016

Hinds, P. S., Drew, D., Oakes, L. L., Fouladi, M., Spunt, S. L., Church, C., & Furman, W. L. (2005). End-of-life care preferences of pediatric patients with cancer. *J Clin Oncol*, *23*(36), 9146–9154. https://doi.org/10.1200/JCO.2005.10.538

Hoffman, J., & Beauchaine, K. (2007). Babies with hearing loss: Steps for effective intervention. *ASHA Lead*, *12*(2), 8–23. https://doi.org/10.1044/leader.FTR3.12022007.8

Hoffman, M. F., Cejas, I., & Quittner, A. L. (2016). Comparisons of longitudinal trajectories of social competence: Parent ratings of children with cochlear implants versus hearing peers. *Otol Neurotol*, *37*(2), 152–159. https://doi.org/10.1097/MAO.0000000000000938

Hoffman, M., Tiddens, E., Quittner, A. L., & Team, Cd. I. (2018). Comparisons of visual attention in school-age children with cochlear implants versus hearing peers and normative data. *Hear Res*. https://doi.org/10.1016/j.heares.2018.01.002.

Hoffmann, T. C., Montori, V. M., & Del Mar, C. (2014). The connection between evidence-based medicine and shared decision making. *JAMA*, *312*(13), 1295–1296. https://doi.org/10.1001/jama.2014.10186

Hughes, M. L., Neff, D. L., Simmons, J. L., & Moeller, M. P. (2014). Performance outcomes for borderline cochlear implant recipients with substantial preoperative residual hearing. *Otol Neurotol*, *35*(8), 1373–1384. https://doi.org/10.1097/MAO.0000000000000367

Hyde, M., Punch, R., & Komesaroff, L. (2010a). A comparison of the anticipated benefits and received outcomes of pediatric cochlear implantation: parental perspectives. *Am Ann Deaf*, *155*(3), 322–338. https://doi.org/10.1353/aad.2010.0020

Hyde, M., Punch, R., & Komesaroff, L. (2010b). Coming to a decision about cochlear implantation: Parents making choices for their deaf children. *J Deaf Stud Deaf Educ*, *15*(2), 162–178. https://doi.org/10.1093/deafed/enq004

Incesulu, A., Vural, M., & Erkam, U. (2003). Children with cochlear implants: Parental perspective. *Otol Neurotol*, *24*, 605–611. https://doi.org/10.1097/00129492-200307000-00013

Jacobsen, M. J., O'Connor, A., & Stacey, D. (2013). Decisional Needs Assessment in Populations: A workbook for assessing patients 'and practitioners 'decision-making needs. *Decisional Needs Assessment in Populations*.

Johnston, J. C., Durieux-Smith, A., Fitzpatrick, E., O'Connor, A., Benzies, K., & Angus, D. (2008). An assessment of parents 'decision-making regarding paediatric cochlear implants. *Can J Speech-Lang Pathol Audiol*, *32*(4), 169–182.

Johnston, J. C., Smith, A. D., O'Connor, A., Benzies, K., Fitzpatrick, E. M., & Angus, D. (2009). The development and piloting of a decision aid for parents considering sequential bilateral cochlear implantation for their child with hearing loss. *Volta Rev*, *109*(2–3), 121–141.

Johnston, J. C., Smith, A. D., Fitzpatrick, E., O'Connor, A., Angus, D., Benzies, K., & Schramm, D. (2010). Estimation of risks associated with paediatric cochlear implantation. *Cochlear*

Implants Int, *11*(3), 146–169. https://doi.org/10.1002/cii.421

Joint Committee on Infant Hearing. (2019). *Year 2019 position statement: Principles and guidelines for early hearing detection and intervention programs-exclusive summary*. http://www.jcih.org/JCIH_2019_Executive_Summary.pdf

Kalejaiye, A., Ansari, G., Ortega, G., Davidson, M., & Kim, H. J. (2017). Low surgical complication rates in cochlear implantation for young children less than 1 year of age. *Laryngoscope*, *127*(March), 720–724. https://doi.org/10.1002/lary.26135

Keilmann, A., Limberger, A., & Mann, W. J. (2007). Psychological and physical well-being in hearing-impaired children. *Int J Pediatr Otorhinolaryngol*, *71*(11), 1747–1752. https://doi.org/10.1016/j.ijporl.2007.07.013

Kennedy, C. R., McCann, D. C., Campbell, M. J., Law, C. M., Mullee, M., Petrou, S., Watkin, P., Worsfold, S., Yuen, H. M., & Stevenson, J. (2006). Language ability after early detection of permanent childhood hearing impairment. *N Engl J Med*, *354*(20), 2131–2141. https://doi.org/10.1056/NEJMoa054915

Kim, Y., Patel, V. A., Isildak, H., Carr, M. M., & Virginia, W. (2017). An analysis of safety and adverse events following cochlear implantation in children under 12 months of age. *Otol Neurotol*, *38*(10), 1426–1432. https://doi.org/10.1097/MAO.0000000000001585

Kluwin, T. N., & Stewart, D. A. (2000). Cochlear implants for younger children: A preliminary description of the parental decision process and outcomes. *Am Ann Deaf*, *145*(1), 26–32. https://doi.org/10.1353/aad.2012.0247

Knoors, H., & Marschark, M. (2012). Language planning for the 21st century: Revisiting bilingual language policy for deaf children. *J Deaf Stud Deaf Educ*, *17*(3), 291–305. https://doi.org/https://dx.doi.org/10.1093/deafed/ens018

Kodish, E. (2003). Informed consent in pediatric research. *J Pediatr*, *13*(4), 346–358. https://doi.org/10.1067/mpd.2003.64

Kopelovich, J. C., Reiss, L. A. J., Etler, C. P., Xu, L., Bertroche, J. T., Gantz, B. J., & Hansen, M. R. (2015). Hearing loss after activation of hearing preservation cochlear implants might be related to afferent cochlear innervation injury. *Otol Neurotol*, *36*(6), 1035–1044. https://doi.org/https://dx.doi.org/10.1097/MAO.0000000000000754

Kral, A., & O'Donoghue, G. (2010). Profound deafness in childhood. *N Engl J Med*, *363*(15), 1438–1450.

Kral, A., & Sharma, A. (2012). Developmental neuroplasticity after cochlear implantation. *Trends Neurosci*, *35*(2), 111–122. https://doi.org/10.1016/j.tins.2011.09.004

Lassig, A.-A. D., Zwolan, T. A., & Telian, S. A. (2005). Cochlear implant failures and revision. *Otol Neurotol*, *26*(4), 624–634. https://doi.org/10.1097/01.mao.0000178123.35988.96

Légaré, F., Stacey, D., Turcotte, S., Mj, C., Kryworuchko, J., Id, G., Lyddiatt, A., Mc, P., Thomson, R., & Elwyn, G. (2014). Interventions for improving the adoption of shared decision making by healthcare professionals. *Cochrane Database Syst Rev*, *15*(9), https://doi.org/10.1002/14651858

Légaré, F., O'Connor, A. C., Graham, I., Saucier, D., Côté, L., Cauchon, M., & Paré, L. (2006a). Supporting patients facing difficult health care decisions: Use of the Ottawa Decision Support Framework. *Canadian Family Physician*, *52*(4), 476–477.

Légaré, F., O'Connor, A. M., Graham, I. D., Wells, G. A., & Tremblay, S. (2006b). Impact of the Ottawa Decision Support Framework on the agreement and the difference between patients' and physicians' decisional conflict. *Med Decis Making*, *26*(4), 373–390. https://doi.org/10.1177/0272989X06290492

Légaré, F., Stacey, D., Brière, N., Desroches, S., Dumont, S., Fraser, K., Murray, M. A., Sales, A., & Aubé, D. (2011). A conceptual framework for interprofessional shared decision-making in-home care: Protocol for a feasibility study. *BMC Health Serv Res*, *11*(1), 1–7. https://doi.org/10.1186/1472-6963-11-23

Légaré, F., & Witteman, H. (2013). Shared decision making: Examining key elements and barriers to adoption into routine clinical practice. *Health Affairs*. https://doi.org/10.1377/hlthaff.2012.1078

Leigh, J. R., Dettman, S. J., Dowell, R.C., & Sarant, J. Z. (2011). Evidence-based approach for making cochlear implant recommendations for infants with residual hearing. *Ear Hear*, *32*(3), 313–322. https://doi.org/10.1097/AUD.0b013e3182008b1c

Leigh, J. R., Dettman, S. J., & Dowell, R. C. (2016). Evidence-based guidelines for recommending cochlear implantation for young children: Audiological criteria and optimizing age at implantation. *Int J Audiol*, *55 Suppl 2*, S9–S18. https://doi.org/https://dx.doi.org/10.3109/14992027.2016.1157268

Lenarz, T., Buechner, A., Lesinski-schiedat, A., Patrick, J., & Pesch, J. (2009). Hearing conservation surgery using the Hybrid-L Electrode. *Audiol Neurootol*, *14*(suppl 1), 22–31. https://doi.org/10.1159/000206492

Lescanne, E., Zahrani, M. A., Bakhos, D., Robier, A., & Morinière, S. (2011). International journal of pediatric otorhinolaryngology revision surgeries and medical interventions in young cochlear implant recipients. *Int J Pediatr Otorhinolaryngol*, *75*(10), 1221–1224. https://doi.org/10.1016/j.ijporl.2011.07.003

Li, Y., Bain, L., & Steinberg, A. G. (2003). Parental decision making and the choice of communication modality for the child who is deaf. *Arch Pediatr Adolesc Med*, *157*(2), 162–

168. https://doi.org/10.1001/archpedi.157.2.162

Li, Y., Bain, L., & Steinberg, A. G. (2004). Parental decision-making in considering cochlear implant technology for a deaf child. *Int J Pediatr Otorhinolaryngol*, *68*(8), 1027–1038. https://doi.org/10.1016/j.ijporl.2004.03.010

Lipstein, E. A., Brinkman, W. B., Fiks, A. G., Hendrix, K. S., Kryworuchko, J., Miller, V. A., Prosser, L. A., Ungar, W. J., & Fox, D. (2015). An emerging field of research: Challenges in pediatric decision making. *Med Decis Making*, *35*(5), 403–408. https://doi.org/10.1177/0272989X14546901

Loundon, N., Blanchard, M., Roger, G., Denoyelle, F., & Garabedian, E. N. (2010). Medical and surgical complications in pediatric cochlear implantation. *Arch Otolaryngol Head Neck Surg*, *136*(1), 12–15. https://doi.org/ 10.1001/archoto.2009.187

Loy, B., Warner-Czyz, A. D., Tong, L., Tobey, E. A., & Roland, P. S. (2010). The children speak: An examination of the quality of life of pediatric cochlear implant users. *Am J Otolaryngol*, *142*(2), 247–253. https://doi.org/10.1016/j.otohns.2009.10.045

Madrigal, V. N., Carroll, K. W., Hexem, K. R., Faerber, J. A., Morrison, W. E., & Feudtner, C. (2012). Parental decision-making preferences in the pediatric intensive care unit. *Crit Care Med*, *40*(10), 2876–2882. https://doi.org/10.1097/CCM.0b013e31825b9151

Maggs, J., Ambler, M., & Hanvey, K. (2017). Trends in cochlear implant candidacy in children. *J Paediatr Child Health*, *27*(10), 454–458. https://doi.org/10.1016/j.paed.2017.06.002

Makoul, G., & Clayman, M. L. (2006). An integrative model of shared decision making in medical encounters. *Patient Educ Couns*, *60*(3), 301–312. https://doi.org/10.1016/j.pec.2005.06.010

Marlowe, A. L., Chinnici, J. E., Rivas, A., Niparko, J. K., & Francis, H. W. (2009). Revision

cochlear implant surgery in children: The Johns Hopkins experience. *Otol Neurotol*, *31*(1), 74–82. https://doi.org/10.1097/MAO.0b013e3181c29fad

Marschark, M., Rhoten, C., & Fabich, M. (2007). Effects of cochlear implants on children's reading and academic achievement. *J Deaf Stud Deaf Educ*, *12*(3), 269–282. https://doi.org/10.1093/deafed/enm013

Marschark, M., Shaver, D. M., Nagle, K. M., & Newman, L. A. (2015). Predicting the academic achievement of deaf and hard-of-hearing students from individual, household, communication, and educational factors. *Except Child*, *81*(3), 350–369. https://doi.org/10.1177/0014402914563700.Predicting

Martin, D., Bat-Chava, Y., Lalwani, A., & Waltzman, S. B. (2011). Peer relationships of deaf children with cochlear implants: Predictors of peer entry and peer interaction success. *J Deaf Stud Deaf Educ*, *16*(1), 108–120. https://doi.org/10.1093/deafed/enq037

Mayer, C., & Trezek, B. J. (2018). Literacy outcomes in deaf students with cochlear implants: Current state of the knowledge. *J Deaf Stud Deaf Educ*, *23*(1), 1–16. https://doi.org/10.1093/deafed/enx043

McCracken, W., Young, A., & Tattersall, H. (2008). Universal newborn hearing screening: Parental reflections on very early audiological management. *Ear Hear*, *29*(1), 54–64. https://doi.org/10.1097/AUD.0b013e31815ed8d0

Mehra, S., Eavey, R. D., & Keamy, D. G. (2009). The epidemiology of hearing impairment in the United States: Newborns, children, and adolescents. *Otolaryngol Head Neck Surg*, *140*, 461–472. https://doi.org/10.1016/j.otohns.2008.12.022

Meredith, M. A., Rubinstein, J. T., Sie, K. C. Y., & Norton, S. J. (2017). Cochlear implantation in children with postlingual progressive steeply sloping high-frequency hearing loss. *J Am*

Acad Audiol, *28*(10), 913–919. https://doi.org/10.3766/jaaa.16115

Michael, R., Attias, J., & Raveh, E. (2019). Cochlear implantation and social-emotional functioning of children with hearing loss. *J Deaf Stud Deaf Educ*, *24*(1), 25–31. https://doi.org/https://dx.doi.org/10.1093/deafed/eny034

Migirov, L, Muchnik, C., Kaplan-Neeman, R., & Kronenberg, J. (2006). Surgical and medical complications in paediatric cochlear implantation: A review of 300 cases. *Cochlear Implants Int*, *7*(4), 194–201. https://doi.org/10.1002/cii.319

Migirov, Lela, Dagan, E., & Kronenberg, J. (2009). Surgical and medical complications in different cochlear implant devices. *Acta Oto-Laryngologica*, *129*(7), 741–744. https://doi.org/10.1080/00016480802398954

Miller, V. A., & Nelson, R. M. (2012). Factors related to voluntary parental decision-making in pediatric oncology. *Pediatrics*, *129*(5), 903–909. https://doi.org/10.1542/peds.2011-3056

Miyamoto, R., Houston, D., Kirk, K., Perdew, A., & Svirsky, M. (2003). Language development in deaf infants following cochlear implantation. *Acta Oto-Laryngologica*, *123*(2), 241–244. https://doi.org/10.1080/00016480310001079

Moeller, M. P., & Tomblin, J. B. (2015). An introduction to the outcomes of children with hearing loss study. *Ear Hear*, *36*(1), 4S-13S. https://doi.org/10.1097/AUD.0000000000000210

Moog, J. S., & Geers, A. E. (2010). Early educational placement and later language outcomes for children with cochlear implants. *Otol Neurotol*, *31*(8), 1315–1319. https://doi.org/10.1097/MAO.0b013e3181eb3226

Morettin, M., dos Santos, M. J. D., Stefanini, M. R., de Lourdes Antonio, F., Bevilacqua, M. C., & Cardoso, M. R. A. (2013). Measures of quality of life in children with cochlear implant:

Systematic review. *Braz J Otorhinolaryngol*, *79*(3), 375–381. https://doi.org/10.5935/1808-8694.20130066

Most, T., & Zaidman-Zait, A. (2001). The needs of parents of children with cochlear implants. *Volta Rev*, *103*(2), 99–113.

Moteki, H., Nishio, S., Miyagawa, M., Tsukada, K., Iwasaki, S., & Usami, S. (2017). Long-term results of hearing preservation cochlear implant surgery in patients with residual low frequency hearing. *Acta Oto-Laryngologica*, *137*(5), 516–521. https://doi.org/10.1080/00016489.2016.1252061

Moumjid, N., Gafni, A., Brémond, A., & Carrère, M. O. (2007). Shared decision-making in the medical encounter: Are we all talking about the same thing? *Med Decis Making*, 27(5), 539–546. https://doi.org/10.1177/0272989X07306779

Mylanus, E. A. M., Rotteveel, L. J. C., & Leeuw, L., R. (2004). Congenital malformation of the inner ear and pediatric cochlear implantation. *Otol Neurotol*, *25*(3), 308–317.

Netten, A. P., Rieffe, C., Theunissen, S. C., Soede, W., Dirks, E., Korver, A. M, Konings, S., Oudesluys-Murphy, A. M., Dekker, F. W., Frijns, J. H., & DECIBEL Collaborative study group. (2015). Early identification: Language skills and social functioning in deaf and hard of hearing preschool children. *Int J Pediatr Otorhinolaryngol*, *79*(12), 2221–2226. https://doi.org/10.1016/j.ijporl.2015.10.008

Nittrouer, S., & Burton, L. T. (2005). The role of early language experience in the development of speech perception and phonological processing abilities: Evidence from 5-year-olds with histories of otitis media with effusion and low socioeconomic status. *J Commun Disord*, *38*, 29–63. https://doi.org/10.1016/j.jcomdis.2004.03.006

O'Brien, G. L. C., Kenna, M., Neault, M., Clark, T. A., Kammerer, B., Johnston, J., Waldman,

E., Pierce, S., Forbes, P., & Licameli, G. R. (2010). Not a “sound” decision: Is cochlear implantation always the best choice? *Int J Pediatr Otorhinolaryngol*, *74*(10), 1144–1148. https://doi.org/10.1016/j.ijporl.2010.07.002

O’Brien, L. C. G., Valim, C., Neault, M., Kammerer, B., Clark, T., Johnston, J., Culver, S., Zhou, J., Kenna, M. A., & Licameli, G. R. (2012). Prognosis tool based on a modified children’s implant profile for use in pediatric cochlear implant candidacy evaluation. *Ann Otol Rhinol Laryngol*, *121*(2), 73–84. https://doi.org/10.1177/000348941212100201

O’Connor, A. M. (2010). User manual—Decisional conflict scale. In *Ottawa: Ottawa Hospital Research Institute*. http://decisionaid.ohri.ca/docs/develop/User_manuals/UM_Decisional_Conflict.pdf

O’Connor, A. M., & Edwards, A. (2009). The role of decision aids in promoting evidence-based patient choice. In *Shared decision-making in health care: Achieving evidence-based patient choice* (pp. 191–200).

O’Connor, A. M. (1995). Validation of a decisional conflict scale. *Med Decis Making*, *15*(1), 25–30.

O’Connor, A. M., Tugwell, P., Wells, G. A., Elmslie, T., Jolly, E., Hollingworth, G., McPherson, R., Bunn, H., Graham, I., & Drake, E. (1998). A decision aid for women considering hormone therapy after menopause: decision support framework and evaluation. *Patient Educ Couns*, *33*(3), 267–279. https://doi.org/10.1016/S0738-3991(98)00026-3

Osberger, M. J., Zimmerman-Phillips, S., & Koch, D. B. (2002). Cochlear implant candidacy and performance trends in children. *Ann Otol Rhinol Laryngol*, *111*(5 II), 62–65.

Peixoto, M., Bento, M., & Oliveira, S. (2017). Cochlear implant in speech understanding in noise. *Otolaryngol Head Neck Surg*, *157*(Supplement 1), P238–P239.

https://doi.org/http://dx.doi.org/10.1177/0194599817717250

Percy-Smith, L., Cayé-Thomasen, P., Gudman, M., Jensen, J. H., & Thomsen, J. (2008). Self-esteem and social well-being of children with cochlear implant compared to normal-hearing children. *Int J Pediatr Otorhinolaryngol*, *72*(7), 1113–1120. https://doi.org/10.1016/j.ijporl.2008.03.028

Pittman, A. L., Lewis, D. E., Hoover, B. M., & Stelmachowicz, P. G. (2005). Rapid word-learning in normal-hearing and hearing-impaired children: effects of age, receptive vocabulary, and high-frequency amplification. *Ear Hear*, *26*, 619–629. https://doi.org/10.1097/01.aud.0000189921.34322.68

Porter, A., Creed, P., Hood, M., & Ching, T. Y. C. (2018). Parental decision-making and deaf children: A systematic literature review. *J Deaf Stud Deaf Educ*, *23*(4), 295–306. https://doi.org/10.1093/deafed/eny019

Punch, R., & Hyde, M. (2010). Children with cochlear implants in Australia: Educational settings, supports, and outcomes. *J Deaf Stud Deaf Educ*, *15*(4), 405–421. https://doi.org/10.1093/deafed/enq019

Punch, R., & Hyde, M. B. (2011). Communication, psychosocial, and educational outcomes of children with cochlear implants and challenges remaining for professionals and parents. *Int J Otolaryngol*. https://doi.org/10.1155/2011/573280

Qi, S., & Mitchell, R. E. (2012). Large-scale academic achievement testing of deaf and hard-of-hearing students: Past, present, and future. *J Deaf Stud Deaf Educ*, *17*(1), 1–18. https://doi.org/10.1093/deafed/enr028

Reed, S., Antia, S. D., & Kreimeyer, K. H. (2008). Academic status of deaf and hard-of-hearing students in public schools: Student, home, and service facilitators and detractors. *J Deaf*

Stud Deaf Educ, *13*(4), 485–502. https://doi.org/10.1093/deafed/enn006

Rich, S., Levinger, M., Werner, S., & Adelman, C. (2013). Being an adolescent with a cochlear implant in the world of hearing people: Coping in school, in society and with self-identity. *Int J Pediatr Otorhinolaryngol*, *77*, 1337–1344. https://doi.org/10.1016/j.ijporl.2013.05.029

Roland, L., Fischer, C., Tran, K., Rachakonda, T., Kallogjeri, D., & Lieu, J. E. C. (2016). Quality of life in children with hearing impairment: Systematic review and meta-analysis. *Am J Otolaryngol Head Neck Surg*, *155*(2), 208–219. https://doi.org/10.1177/0194599816640485

Ronner, E. A., Benchetrit, L., Levesque, P., Basonbul, R. A., & Cohen, M. S. (2020). Quality of life in children with sensorineural hearing loss. *Otolaryngol Head Neck Surg*, *162*(1), 129–136.

Rose, A., Rosewilliam, S., & Soundy, A. (2017). Shared decision-making within goal setting in rehabilitation settings: A systematic review. In *Patient Educ Couns*, *100* (1), 65–75. https://doi.org/10.1016/j.pec.2016.07.030

Rosewilliam, S., Roskell, C. A., & Pandyan, A. D. (2011). A systematic review and synthesis of the quantitative and qualitative evidence behind patient-centred goal setting in stroke rehabilitation. *Clin Rehabil*, *25*(6), 501–514. https://doi.org/10.1177/0269215510394467

Russell, J. L., Pine, H. S., & Young, D. L. (2013). Pediatric cochlear implantation: Expanding applications and outcomes. *Pediat Clin N*, *60*(4), 841–863. https://doi.org/10.1016/j.pcl.2013.04.008

Santa Maria, P. L., Gluth, M. B., Yuan, Y., Atlas, M. D., & Blevins, N. H. (2014). Hearing preservation surgery for cochlear implantation: A meta-analysis. *Otol Neurotol*, *35*(10), 256–269. https://doi.org/10.1097/MAO.0000000000000561

Sarant, J. Z., Harris, D. C., & Bennet, L. A. (2015). Academic outcomes for school-aged children

with severe–profound hearing loss and early unilateral and bilateral cochlear implants. *J Speech Lang Hear Res, 58,* 1017–1032. https://doi.org/10.1044/2015_JSLHR-H-14-0075

Schick, B., Skalicky, A., Edwards, T., Kushalnagar, P., Topolski, T., & Patrick, D. (2013). School placement and perceived quality of life in youth who are deaf or hard of hearing. *J Deaf Stud Deaf Educ, 18*(1), 47–61. https://doi.org/10.1093/deafed/ens039

Sininger, Y. S., Grimes, A., & Christensen, E. (2010). Auditory development in early amplified children: Factors influencing auditory-based communication outcomes in children with hearing loss. *Ear Hear*, *31*(2), 166–185.

Skarzynski, H. (2012). Ten years experience with a new strategy of partial deafness treatment. *J Hear Sci, 2*(2), 11–18.

Snels, C., Inthout, J., Mylanus, E., Huinck, W., & Dhooge, I. (2019). Hearing preservation in cochlear implant surgery: A meta-analysis. *Otol Neurotol*, *40*(2), 145–153. https://doi.org/10.1097/MAO.0000000000002083

Sorkin, D. L., & Zwolan, T. A. (2008). Parental perspectives regarding early intervention and its role in cochlear implantation in children. *Otol Neurotol*, *29*(2), 137–142. https://doi.org/10.1097/mao.0b013e3181616c88

Sorrentino, T., Côté, M., Eter, E., Laborde, M., Cochard, N., Deguine, O., & Fraysse, B. (2009). Cochlear reimplantation: Technical and surgical failures. *Acta Oto-Laryngologica, 129*(4), 380–384. https://doi.org/10.1080/00016480802552576

Spencer, P. E., & Marschark, M. (2010). *Evidence-based practice in educating deaf and hard-of-hearing students* (Oxford University Press (ed.)).

Stevenson, J., McCann, D., Watkin, P., Worsfold, S., & Kennedy, C. (2010). The relationship between language development and behaviour problems in children with hearing loss. *J*

Child Psychol Psychiatry, *51*(1), 77–83. https://doi.org/10.1111/j.1469-7610.2009.02124.x

Stevenson, J., Pimperton, H., Kreppner, J., Worsfold, S., Terlektsi, E., & Kennedy, C. (2017). Emotional and behaviour difficulties in teenagers with permanent childhood hearing loss. *Int J Pediatr Otorhinolaryngol*, *101*, 186–195. https://doi.org/10.1016/j.ijporl.2017.07.031

Stewart, J. L., Pyke-Grimm, K. A., & Kelly, K. P. (2005). Parental treatment decision making in pediatric oncology. *Semin Oncol Nurs*, *21*(2), 89–97. https://doi.org/10.1016/j.soncn.2004.12.003

Stewart, M. (2014). Parental decision-making during the cochlear implant selection process. http://digitalcommons.wustl.edu/pacs_capstones/683

Stika, C. J., Eisenberg, L. S., Johnson, K. C., Henning, S. C., Colson, B. G., Ganguly, D. H., & DesJardin, J. L. (2015). Developmental outcomes of early-identified children who are hard of hearing at 12 to 18months of age. *Early Hum Dev 91*(1), 47–55. https://doi.org/10.1016/j.earlhumdev.2014.11.005

Strauss, A. T., Martinez, D. A., Garcia-arce, A., Taylor, S., Mateja, C., Fabri, P. J., & Zayas-castro, J. L. (2015). A user needs assessment to inform health information exchange design and implementation. *BMC Med Inform Decis Mak*, *15*(81), 1–11. https://doi.org/10.1186/s12911-015-0207-x

Sugaya, A., Fukushima, K., Kasai, N., Kataoka, Y., Maeda, Y., Nagayasu, R., Toida, N., Ohmori, S., Fujiyoshi, A., Taguchi, T., Omichi, R., & Nishizaki, K. (2015). Impact of early intervention on comprehensive language and academic achievement in Japanese hearing-impaired children with cochlear implants. *Int J Pediatr Otorhinolaryngol*, *79*(12), 2142–2146. https://doi.org/10.1016/j.ijporl.2015.09.036

Sweeney, A. D., Carlson, M. L., Zuniga, M. G., Bennett, M. L., Wanna, G. B., Haynes, D. S., &

Rivas, A. (2015). Impact of perioperative oral steroid use on low-frequency hearing preservation after cochlear implantation. *Otol Neurotol*, *36*(9), 1480–1485. https://doi.org/10.1097/MAO.0000000000000847

Sweeney, A. D., Hunter, J. B., Carlson, M. L., Rivas, A., Bennett, M. L., Gifford, R. H., Noble, J. H., Haynes, D. S., Labadie, R. F., & Wanna, G. B. (2016). Durability of hearing preservation after cochlear implantation with conventional-length electrodes and scala tympani insertion. *Otolaryngol Head Neck Surg*, *154*(5), 907–913. https://doi.org/10.1177/0194599816630545

Tarkan, Ö., Tuncer, Ü., Özdemir, S., Sürmelioğlu, Ö., Çetik, F., Kiroğlu, M., Kayikçioğlu, E., & Kara, K. (2013). Surgical and medical management for complications in 475 consecutive pediatric cochlear implantations. *Int J Pediatr Otorhinolaryngol*, *77*(4), 473–479. https://doi.org/10.1016/j.ijporl.2012.12.009

Teagle, H. F. B., Park, L. R., Brown, K. D., Zdanski, C., & Pillsbury, H. C. (2019). Pediatric cochlear implantation: A quarter century in review. *Cochlear Implants Int*, *20*(6), 288–298. https://doi.org/10.1080/14670100.2019.1655868

The World Health Organization. (2020). *Deafness and hearing loss*. http://www.who.int/news-room/fact-sheets/detail/deafness-and-hearing-loss

Theunissen, S. C. P. M., Rieffe, C., Kouwenberg, M., De Raeve, L. J. I., Soede, W., Briaire, J. J., & Frijns, J. H. M. (2014). Behavioral problems in school-aged hearing-impaired children: the influence of sociodemographic, linguistic, and medical factors. *Eur Child Adolesc Psychiatry*, *23*(4), 187–196. https://doi.org/https://dx.doi.org/10.1007/s00787-013-0444-4

Theunissen, S. C. P. M., Rieffe, C., Soede, W., Briaire, J. J., Ketelaar, L., Kouwenberg, M., & Frijns, J. H. M. (2015). Symptoms of psychopathology in hearing-impaired children. *Ear*

Hear, *36*(4), e190–e198. https://doi.org/10.1097/AUD.0000000000000147

Tomblin, J. B., Spencer, L., Flock, S., Tyler, R., & Gantz, B. (1999). A comparison of language achievement in children with cochlear implants and children using hearing aids. *J Speech Lang Hear Res*, *42*(2), 497–509. https://doi.org/1092-4388/99/4202-0497

U.S. Food and Drug Administration. (2011). *"Off-Label" and Investigational Use Of Marketed Drugs, Biologics, and Medical Devices*. https://www.fda.gov/RegulatoryInformation/Guidances/ucm126486.htm

Van Eldik, T., Treffers, P. D. A., Veerman, J. W., & Verhulst, F. C. (2004). Mental health problems of deaf Dutch children as indicated by parents' responses to the child behavior checklist. *Am Ann Deaf*, *148*, 390–395. https://doi.org/10.1353/aad.2004.0002

Walker, E. A., Redfern, A., & Oleson, J. J. (2019). Linear mixed-model analysis to examine longitudinal trajectories in vocabulary depth and breadth in children who are hard of hearing. *J Speech Lang Hear Res*, *62*(3), 525–542. https://doi.org/10.1044/2018_JSLHR-L-ASTM-18-0250

Wang, J. T., Wang, A. Y., Psarros, C., & Da Cruz, M. (2014). Rates of revision and device failure in cochlear implant surgery: A 30-year experience. *Laryngoscope*, *124*(10), 2393–2399. https://doi.org/10.1002/lary.24649

Warner-Czyz, A. D., Loy, B., Roland, P. S., Tong, L., & Tobey, E. A. (2009). Parent versus child assessment of quality of life in children using cochlear implants. *Int J Pediatr Otorhinolaryngol*, *73*(10), 1423–1429. https://doi.org/10.1016/j.ijporl.2009.07.009

Weisleder, A., & Fernald, A. (2013). Talking to children matters: Early language experience strengthens processing and builds vocabulary. *Psychol Sci*, *24*(11), 2143–2152. https://doi.org/10.1177/0956797613488145

Westby, C. (2016). Academic performance of Children with cochlear implants. *Word of Mouth*, *27*(3), 1–5.

Wilson, K., Ambler, M., Hanvey, K., Jenkins, M., Jiang, D., Maggs, J., & Tzifa, K. (2016). Cochlear implant assessment and candidacy for children with partial hearing. *Cochlear Implants Int*, *17 Suppl 1*, 66–69. https://doi.org/https://dx.doi.org/10.1080/14670100.2016.1152014

Wyatt, K. D., List, B., Brinkman, W. B., Lopez, G. P., Asi, N., Erwin, P., Wang, Z., Pablo, J., Garces, D., Montori, V. M., & Leblanc, A. (2015). Shared decision making in pediatrics: A systematic review and meta-analysis. *Acad Pediatr*, *15*(6), 573–583. https://doi.org/10.1016/j.acap.2015.03.011

Yeh, J. S., Mooney, K. L., Gingrich, K., Kim, J. T., & Lalwani, A. K. (2011). Anesthetic complications in pediatric patients undergoing cochlear implantation. *Laryngoscope*, *121*(10), 2240–2244. https://doi.org/10.1002/lary.21924

Yoshinaga-Itano, C. (2003). From screening to early identification and intervention: Discovering predictors to successful outcomes for children with significant hearing loss. *J Deaf Stud Deaf Educ*, *8*(1), 11–30. https://doi.org/10.1093/deafed/8.1.11

Yoshinaga-Itano, C. (2004). Levels of evidence: Universal newborn hearing screening (UNHS) and early hearing detection and intervention systems (EHDI). J Commun Disord, *37*(5), 451–465. https://doi.org/10.1016/j.jcomdis.2004.04.008

Yoshinaga-Itano, C. (2001). The social-emotional ramifications of universal newborn hearing screening: Early identification and intervention of children who are deaf or hard of hearing. *A Sound Foundation Through Early Amplification Survey*, *November*, 8–10.

Yoshinaga-Itano, C., Baca, R. L., & Sedey, A. L. (2010). Describing the trajectory of language

development in the presence of severe-to-profound hearing loss: A closer look at children with cochlear implants versus hearing aids. *Otol Neurotol*, *31*(8), 1268–1274. https://doi.org/10.1097/MAO.0b013e3181f1ce07

Young, A., & Andrews, E. (2001). Parents 'experience of Universal Neonatal Hearing Screening: A critical review of the literature and its implications for the implementation of new UNHS programs. *J Deaf Stud Deaf Educ*, *6*(3), 149–160. https://doi.org/10.1093/deafed/6.3.149

Young, A., & Tattersall, H. (2007). Universal newborn hearing screening and early identification of deafness: Parents 'responses to knowing early and their expectations of child communication development. *J Deaf Stud Deaf Educ*, *12*(2), 209–220. https://doi.org/10.1093/deafed/enl033

Zajicek-Farbe, M. L., Lotrecchiano, G. R., Long, T. M., & Farber, J. M. (2015). Parental perceptions of family centered care in medical homes of children with neurodevelopmental disabilities. *Matern Child Health J*, *19*(8), 1744–1755. https://doi.org/10.1007/s10995-015-1688-z

Zanetti, D., Nassif, N., & Redaelli De Zinis, L. O. (2015). Factors affecting residual hearing preservation in cochlear implantation. *Acta Otorhinolaryngol Ital*, *35*(6), 433–441. https://doi.org/10.14639/0392-100X-619

Zwolan, T. A., Ashbaugh, C. M., Alarfaj, A., Kileny, P. R., Arts, H. A., El-Kashlan, H. K., & Telian, S. A. (2004). Pediatric cochlear implant patient performance as a function of age at implantation. *Otol Neurotol*, *25*(2), 112–120.

Chapter 2: Chart review

Clinical characteristics and outcomes of children with cochlear implants who had preoperative residual hearing

Formatted for the International Journal of Audiology

Accepted on 14 February 2021, Published online on 24 March 2021 in the International Journal of Audiology

Clinical characteristics and outcomes of children with cochlear implants who had preoperative residual hearing

Eunjung Na* [1, 2], Karine Toupin-April [1, 2, 3], Janet Olds [2, 4, 5], JoAnne Whittingham [2]

Elizabeth M. Fitzpatrick [1, 2]

[1] School of Rehabilitation Sciences, Faculty of Health Sciences, University of Ottawa, Ottawa, ON, Canada

[2] Children's Hospital of Eastern Ontario Research Institute, Ottawa, ON, Canada

[3] Department of Pediatrics, Faculty of Medicine, University of Ottawa, Ottawa, ON, Canada

[4] Children's Hospital of Eastern Ontario, Ottawa, ON, Canada

[5] Department of Otolaryngology - Head and Neck Surgery, Faculty of Medicine, University of Ottawa, Ottawa, ON, Canada

Corresponding author: Elizabeth M. Fitzpatrick, Ph.D. Faculty of Health Sciences, University of Ottawa, 451 Smyth Road, Ottawa, ON K1H 8M5, elizabeth.fitzpatrick@uottawa.ca; Telephone: 613-562-5800

Abstract

Objective: Cochlear implant (CI) candidacy criteria have expanded to include children with residual hearing. This study explored the clinical profiles and outcomes of children with CIs who had preoperative residual hearing in at least one ear.

Design: A retrospective chart review was conducted to collect clinical characteristics and speech perception data. Pre- and post-CI auditory and speech perception data were analyzed using a modified version of the Pediatric Ranked Order Speech Perception (PROSPER) score.

Study sample: This study included all children with residual hearing who received CIs in one Canadian pediatric center from 1992 to 2018.

Results: A total of 100 of 389 (25.7%) children with CIs had residual hearing (median 77.6 dB HL, better ear). The proportion of children with residual hearing increased from 1992 to 2018. Children who had auditory behaviour and speech perception tests (n=83) showed higher modified PROSPER scores post-CI compared to pre-CI. Phonologically Balanced Kindergarten (PBK) test scores were available for 71 children post-CI; 81.7% (58/71) of children achieved > 80% on the PBK.

Conclusions: One in four children who received CIs had residual hearing, and most of them had severe hearing loss pre-CI. These children showed a high level of speech perception with CIs.

Keywords: *cochlear implantation, speech perception, pediatric, residual hearing, cochlear implant candidate, children*

Introduction

Cochlear implants (CIs) are the standard of care for children who receive little benefit from hearing aids (HAs) (Moog, 2002; Osberger et al., 2002; Raine, 2013; Skarzynski et al., 2009). It has been well-documented that children with severe and profound hearing loss who receive CIs can achieve successful outcomes, in the areas of speech, language, academic achievement, and social development (Dettman et al., 2016; Sarant et al., 2015, 2018; Thoutenhoofd, 2006; Yoshinaga-Itano et al., 2018).

In 1990, the Food and Drug Administration (FDA) in the United States approved CI surgery in children with bilateral profound hearing loss as young as 2 years of age. Since then, the candidacy criteria have expanded to include children with more residual hearing and higher levels of auditory function. This topic has received increasing attention in the literature (Fitzpatrick et al., 2009; Gratacap et al., 2015; Hughes et al., 2014; Leigh et al., 2011, 2016). Studies have shown that audiometric candidacy criteria are variable across clinics (de Kleijn et al., 2018; Dowell et al., 2004; Fitzpatrick et al., 2009). An evidence-based guideline published in Australia by Leigh et al. (2016) recommended that CIs could be considered for children with pure tone average (PTA) between 65 and 85 dB HL because these children may receive more speech-related benefits from CIs than from HAs. In addition, a recent systematic review (de Kleijn et al., 2018) that included studies from multiple countries reported that children who have PTA (at 500, 1000, 2000, and 4000 Hz) ≥ 80 dB HL could also benefit from cochlear implantation. According to a recent survey in the United States, 63 of 81 (78%) surgeons performed CI surgery for off-label or non-traditional indications in children and adult populations (Carlson et al., 2018).

An increasing number of studies of children who are outside FDA CI candidacy criteria

have reported positive outcomes in a number of areas, including improvements in speech, language and auditory functioning (Hyde et al., 2010a; Nicholas & Geers, 2007; Thoutenhoofd, 2006). In addition, in a recent study (Michael et al., 2019), children with CIs who had better than severe hearing loss showed significantly lower hyperactivity/inattention and higher pro-social behaviour scores compared to children with severe to profound hearing loss who use HAs. Several studies have also reported that positive speech-related outcomes after CIs in children with hearing loss are closely associated with the amount of preoperative residual hearing (Leigh et al., 2016; Chiossi & Hyppolito, 2017). In addition, the development of CI technology for preserving residual hearing in low frequencies (Carlson et al., 2015, 2018; Eshraghi et al., 2017; Skarzynski, 2012; Sweeney et al., 2015, 2016; Zanetti et al., 2015) has led to improved hearing abilities and speech perception in noise, overall natural sound and music quality, and sound localization by combining acoustic and electronic stimulation (Adunka et al., 2008, 2013; Dunn et al., 2010; Moteki et al., 2017).

Decision-making about CI for children with residual hearing can be particularly difficult for parents and clinicians because no clear audiological cut-point exists for CI surgery resulting in variability in audiometric candidacy criteria (Fitzpatrick et al., 2009, 2006; Hyde et al., 2010b; Gifford, 2011; Holcomb & Smeal, 2020). Parents and clinicians may feel uncomfortable recommending CI until they can obtain reliable measures of speech perception scores; therefore, they may require more time to make a decision about CIs for this specific population than for children with bilateral profound hearing loss (Fitzpatrick et al., 2009; 2015, Hyde et al., 2010b; Leigh et al., 2011).

In order to assist parents and clinicians in making decisions about CIs, it is important to learn more about the characteristics and outcomes of children with residual hearing who received

CIs. According to the Ottawa Decision Support Tutorial (O'Connor et al., 2015), patient clinical characteristics (e.g., diagnosis, stage of the disease, and duration of the condition) and demographic characteristics (e.g., sex, age, developmental stage, education, occupation, socioeconomic status, ethnicity) are important factors in clinical decision-making. For example, practitioners can support patients' decisions by providing information about the outcomes observed among other patients with similar clinical characteristics (O'Connor et al., 2015). However, very little research has focused specifically on the clinical characteristics of children with residual hearing who received CIs.

This population-based study aimed to document the number of children with residual hearing who received CIs in one Canadian pediatric center. We also explored the clinical profiles as well as the auditory behaviour and speech perception abilities of the children.

Methods

Study design

This study involved a retrospective chart review conducted in a tertiary care pediatric CI center in the capital region of Canada which provides CIs surgical and audiological follow-up services. The clinical data set for this study included all children who received CIs over a 26-year period from 1992 to 2018. Audiological assessments, including speech perception test data, were also extracted from the medical charts. This study was approved by the institutional review boards of the Children's Hospital of Eastern Ontario (CHEO) and the University of Ottawa.

Participants

Data for children who received CIs in the CHEO CI program from 1992 to 2018 were reviewed for the study. CHEO is the only pediatric academic health care facility in a catchment area (the Eastern Ontario area of Canada) of approximately one million. As a Canadian program, all children who were implanted with CIs received care in a publicly funded health care setting, including audiological follow-up services and rehabilitation to develop auditory and spoken language skills after CIs. For this study, data for the full clinical population of children with CIs were reviewed to select children with residual hearing who met the following criteria.

a) Chronological age ≤ 18 years at the time of CI surgery
b) Preoperative thresholds with a PTA ≤ 90 dB HL (average of thresholds at 500, 1000, 2000 Hz) in at least one ear (either the CI ear or the non-CI ear, including children with single-sided deafness).

Procedures

Using a data-specific form (Appendix A), clinical information for each participant was extracted from medical charts. The information included sex, diagnosis of hearing loss including hearing screening status, age of diagnosis, and degree of hearing loss, as well as etiology, progression of hearing loss, and other disabilities. Clinical characteristics of the children that were also documented including age at CI candidacy, audiological characteristics pre-CI, age at implantation and age at follow-up.

Pre- and post-CI surgery speech perception scores were also extracted. Assessment tools used in the clinical protocol in the CHEO CI program consisted of auditory behaviour questionnaires, closed-set, and open-set words and sentence tests. The assessments varied throughout the 26 years (1992 to 2018) covered by this study as new tools became available and

were adopted as part of the assessment protocol (see Table 1). Children were assessed in their best aided condition (binaural HAs pre-implant for most children and unilateral CI, unilateral CI and HA or bilateral CIs post-implant). The pre-and post-CI assessment measures administered were based on clinical judgement for each child, for a diverse clinical population with a wide age range, different communication modes, and variable linguistic function. The typical clinical protocol administered in the clinic included the following tools. LittlEARS® Auditory Questionnaire (LittlEARS) (Coninx et al., 2009) and Infant-Toddler Meaningful Auditory Integration Scale (IT-MAIS) (Zimmerman-Phillips et al., 2001) parent questionnaires were commonly used to assess auditory behaviors in infants and young children. As receptive language skills emerged, assessments were completed using closed-set speech perception test, primarily the Early Speech Perception (ESP) (Moog & Geers, 1991) which consists of three subtests: 1) Pattern Perception, 2) Spondee Identification, 3) Monosyllable Identification. Clinicians administered open-set word tests, typically the Phonologically Balanced Kindergarten (PBK) test (Haskins, 1949) at age 4-5 years. When a child could not complete the PBK test, other open-set tests including the Glendonald Auditory Screening Procedure (GASP) (Erber, 1982) and/or the Multisyllabic Lexical Neighborhood Tests (MLNT) (Kirk et al., 1995) were selected or a closed-set test, Word Intelligibility by Picture Identification (WIPI) (Ross & Lerman, 1970) was administered. For the PBK, a 25-word list was administered at 60 dB SPL using recorded material. This variation in clinical measures precluded the possibility of comparing pre-and post-CI speech perception data using a single outcome measure in this study population.

We modified the Pediatric Ranked Order Speech Perception (PROSPER) score, originally developed by Trimble et al. (2008) to rank speech perception measures in a

hierarchical order from parent questionnaires to PBK open-set word tests. This ranking method was based on early work by Geers & Moog (1987) and allows for the integration of all available auditory behaviour and speech perception data into categories of performance (Trimble et al., 2008). Our modification of the original PROSPER scores to rank the assessments that were used in the CI program at CHEO is presented in Table 2. For example, measures such as the LittlEars test were not used in the original PROSPER but were part of the CHEO protocol. As shown in Table 2, within each category, a score was assigned to the specific assessments. Assessments in the original PROSPER that were not used to assess children in this study were not included. The scoring system was organized into five broad category scores, with 0 corresponding to "could not test" (could not be done pre-CI due to child's low language function but had shown improvement at post-CI), 1 to parent questionnaires, 2 to closed-set to easy open-set tests, 3 to LNT open-set word tests, and 4 to PBK open-set word tests (see Table 2).

Data Analysis

Data management and analysis were carried out using the Statistical Package for the Social Sciences (SPSS) 26.0 (IBM Corp.). Descriptive statistics were used to summarize the clinical characteristics of the children (e.g., frequencies and means or medians of continuous variables, as appropriate). Using the Wilcoxon signed-rank test, we compared ages at key time points (e.g., age at diagnosis, pre-CI assessment, CI surgery, and post-assessment) between the residual hearing group and the children with bilateral profound hearing loss who received CIs in the clinic. Pre-operative thresholds for individual frequencies were compared between the CI ear and the contralateral non-CI ear for children with residual hearing who received a unilateral CI

or sequential bilateral CIs. The differences in the PTA between the CI ear and the non-CI ear were compared using paired t-tests.

Modified PROSPER scores and category scores at pre- and post-CI were calculated and recorded in Microsoft Excel for initial classification and then transferred to SPSS. PBK results pre- and post-CI were compared with paired t-tests. Two-tailed tests were applied for all analyses with statistical significance set at $p<0.05$.

Results

Proportion of children with residual hearing

During the study period (1992-2018), 389 children underwent unilateral or bilateral CIs at CHEO. The proportion of children with residual hearing tended to increase annually after 2004, ranging from 30 to 60% of children implanted. Notably, in the last two years (2017 and 2018), more than 60% of all children implanted had residual hearing (Figure 1).

A total of 100 of the 389 (25.7%) children with CIs were identified as having residual hearing based on the hearing in the better ear. Of these, 67 (67.0%) children underwent CI surgery in one ear and the remaining 33 (33.0%) in both ears (see Figure 2).

Clinical characteristics of children

Table 3 summarizes the clinical characteristics of all 389 children who underwent CIs at CHEO and of the 100 children with residual hearing. The group of children with residual hearing was diagnosed with hearing loss at a median age of 13.0 months (interquartile range [IQR]: 3.6, 25.0), similar to that of the bilateral profound hearing loss group (12.2 months [IQR: 6.2, 21.7]) *($p=0.795$)*. The residual hearing group received their first CI at 46.2 months (IQR: 29.3, 94.0),

several months older than the profound hearing loss group at 39.4 months (IQR: 22.1, 81.0), but not significantly different *(p=0.122)*. However, the median time from diagnosis to CI surgery was significantly longer in the residual hearing group (29.6 months [IQR: 11.8, 61.4]) than in the bilateral profound group (16.7 months [IQR: 7.8, 46.8]) *(p<0.0001).*

As shown in Table 3, over half of the 100 children (52.0%) were screened. Children who were not screened (27.0%) were born before the implementation of universal hearing screening in 2002. Children whose route to identification was unknown (21.0%) either transferred or were referred from other provinces or countries and information on screening was not available. Almost half of the 100 (46.0%) children had congenital hearing loss or early-onset hearing loss (diagnosed ≤ 6 months of age), and 14 children (14.0%) presented with late-onset hearing loss (diagnosed > 6 months of age).

The sample of 100 children demonstrated a range of hearing profiles. A total of 87 (87.0%) children had sensorineural or mixed hearing loss, and the remaining 13 (13.0%) had auditory neuropathy spectrum disorder (ANSD). From initial diagnosis to CI surgery, 68 of 100 (68.0%) children showed > 20 dB HL deterioration in hearing thresholds in at least one ear.

Severity of hearing loss based on the better ear initial auditory brainstem response (ABR) testing at diagnosis and PTA (at 500, 1000 and 2000 Hz) at CI surgery is shown in Table 3. At diagnosis, children had a median threshold of 71.5 dB HL (IQR: 53.7, 80.0); 43 (43.0%) children had mild to moderate loss, 38 (38.0%) severe, and 10 (10.0%) profound hearing loss. Severity of hearing loss at diagnosis could not be verified for the remaining 9 (9.0%) children because they were referred from other regions. At CI surgery, the median PTA in the better ear for these 100 children was 77.6 dB HL (IQR: 73.8, 86.7); 82 (82.0%) had severe hearing loss, 17 (17%) had moderate or moderate-severe hearing loss, and 1 (1.0%) had single-sided deafness.

Figure 3 provides preoperative individual hearing thresholds (both CI and non-CI ears) for 89 children with unilateral CI (n=67) or sequential bilateral CIs (n=22). The 11 children with simultaneous bilateral CIs are not included. In these 89 children the mean audiometric threshold in the CI ear was significantly worse (91.7±15.5 dB HL [range: 43.3, 120.0]) than the non-CI ear (78.6±17.0 dB HL [range: 1.7, 120.0]) *(p=0.001)*. The mean low-frequency average (250 and 500 Hz), of 78.7±19.7 dB HL, was significantly better than the mean high-frequency average (1000 to 4000Hz) of 97.1±15.8 dB HL in the CI ear (*p<0.001*). This pattern is the same for the non-CI ear.

Auditory behaviour and speech perception

Assessments of auditory behaviour and speech perception were available for 83 (83.0%) children with residual hearing (see Figure 4). Reasons for the 17 incomplete assessments included pre-CI assessments completed elsewhere and not available (n=5), family relocated after CI surgery (n=3), low language function due to additional disabilities (n=6), using sign language only (n=1), and two were unknown.

Age of children at assessment

The ages of the children at pre- and post-CI assessment and at surgery are shown in Table 4. The children had a median age of 45.8 months (IQR: 22.7, 104.0) at pre-CI assessment and 115.2 months (IQR: 69.3, 161.5) at their most recent post-CI assessment. The median duration of CI use was 34.3 months (IQR: 19.0, 75.3). Age of the children at assessment in each of the modified PROSPER categories is also reported in Table 4. As indicated, children had different assessment measures at different ages. The children in the modified PROSPER category 1

(Parent Questionnaires) were assessed at age 19.6 months (IQR: 13.8, 24.5); younger children are often not able to do speech perception testing, and parent questionnaires provide a proxy assessment of auditory behaviour. Children tested with PBK open-set word assessments (modified PROSPER category 4) were older, with a median age of 104.1 months (IQR: 69.8, 150.1).

Modified PROSPER outcomes

All 83 children showed higher modified PROSPER category scores post-CI compared to their pre-CI scores (Figure 5). For example, as shown, of the 15 children in category 0 (could not test) pre-CI, 10 children achieved category 4 (PBK words) post-CI. A total of 71 of the 83 children had PBK scores post-CI; the remaining 12 children did not have post-CI PBK scores because they were additional disabilities (n= 5), too young (< 48 months) to participate in PBK (n= 4), had behavioural issues (n= 2), or used sign language (n=1). As shown in Figure 5, all 12 children showed improvement in category scores but did not advance beyond category 1 (n=4) or category 2 (n=8).

Of the 71 children with open-set scores post-implant, 37 children completed less advanced tests pre-CI and the other 34 completed the PBK test both pre- and post-CI. As shown in Figure 6, a total of 37 of the 71 children who were assessed pre-CI with less advanced tests (e.g., modified PROSPER category scores "0-3": could not test, parent questionnaire, closed-set/easy open-set, and LNT open-set words) were able to complete the PBK test (modified PROSPER category score 4) post-CI, obtaining a mean score of 85.3% ±13.10 (range 48.0%-100.0%). Figure 7 shows scores for the 34 children who completed the PBK test pre- and post-CI. Duration of CI use for these children was a median of 30.7 months (IQR: 14.0, 54.2). These

children showed significant improvement in speech perception abilities, from a mean score of 34.0±23.1% (range: 4.0-80.0) pre-CI to 89.8±9.81% (range: 92.0-100.0) post-CI *($p<0.001$)*. Overall, 58 of the 71 (81.7%) children achieved PBK scores of 80.0% or more post-CI, including 15 (21.1%) who obtained scores of 100.0%.

Discussion

The purpose of this study was to describe the clinical characteristics as well as auditory behaviour and speech perception abilities of children with residual hearing who received CIs. This retrospective population-based study provides comprehensive data on an entire population of children with CIs in one region of Canada. We found that about one-quarter of the 389 (25.7%) children who received CI from 1992 to 2018 had preoperative residual hearing, and in the last two years of the study, these represented more than half the children who received CIs. This finding is consistent with data from Teagle et al. (2019) who reported that the number of implants in children with lesser degrees of hearing loss increased after 2008. In addition, publications from several countries suggest there is a considerable increase in implantation for children with residual hearing (British Cochlear Implant Group, 2007; Carlson et al., 2018; De Raeve & Wouters, 2013; Leigh et al., 2011, 2016). However, there are still limited population-based studies on the proportion of children with residual hearing who receive CIs.

Our findings showed no significant difference in the age at CI surgery between children with residual hearing and those with bilateral profound hearing loss in the CI program where our study was conducted. However, for children with residual hearing, the interval between diagnosis and CI surgery was longer than for children with little or no residual hearing. This finding may reflect a need for more decision-making time for parents and practitioners due to

lack of specific audiometric criteria and to uncertainty about outcomes (Fitzpatrick et al., 2009, 2015; Hyde et al., 2010b).

Our study also found that almost half (43.0%) of the children with residual hearing had better than a severe loss in their better ear at diagnosis. At the time of CI, almost one in five (18.0%) children had hearing within the mild to moderate range in their better ear. Children had a median preoperative PTA threshold of 77.6 dB HL, consistent with thresholds reported in a recent systematic review (de Kleijn et al., 2018). This review reported that a CI was recommended for children with audiometric thresholds in the range of 70-90 dB HL. In addition, an evidence-based guideline from Australia recommended that children with hearing loss in the range of 65–80 dB HL be considered for CIs (Leigh et al., 2016). A previous study that explored the perspectives of from all pediatric centers across Canada also found that the definition of borderline audiometric thresholds ranged from 70 dB HL to 85 dB HL (Fitzpatrick et al., 2009). According to both our findings and previous findings, audiometric thresholds for pediatric CI candidacy applied in clinical practice are lower than those typically found in current candidacy criteria.

Another hearing profile that characterized children with residual hearing at CI surgery was the presence of substantial low-frequency hearing. This population had significantly better hearing thresholds in both CI and non-CI ears across 250 and 500 Hz than across 1000 and 4000 Hz. This is consistent with reports from other studies that some children with residual hearing are likely to have profound high-frequency hearing loss and much better hearing in the low frequencies (Gratacap et al., 2015; Wilson et al., 2016; Meredith et al., 2017). Since these children are often considered well-functioning with their HAs, many of them are not referred for CI candidacy assessment. However, difficulties in audiological, speech and language

abilities, academic progress, social integration, and social-emotional functioning may be observed in this population (Fitzpatrick et al., 2009; Michaels, et al., 2019). Given this information, in clinical practice, monitoring of children's hearing functioning in multiple areas beyond audiometric criteria is needed when considering CI candidacy.

This study also explored speech recognition outcomes and provides insights into the comprehensive clinical profiles of this specific population. This study applied the modified PROSPER category score to compare pre- and post-CI auditory behaviour and speech perception scores of the children with different ages at pre-post-CI assessments and duration of CI use. A total of 83 children out of 100 had pre- and post-CI auditory behaviour or speech perception outcomes, and all these children had documented results that showed a higher level of the modified PROSPER score post-CI. Of these children, a total of 71 children achieved open-set word scores post-CI, and over 80% children obtained PBK scores of 80% or more post-CI. The 34 children who completed both pre- and post-CI open-set PBK word tests improved from a mean score of 34% to 90% with CIs. Our findings are consistent with several previous studies which reported that post-CI open-set speech perception test scores for children with residual hearing were significantly higher than preoperative results with HAs (Fitzpatrick et al., 2006; Gratacap et al., 2015, Leigh et al., 2016, Chiossi & Hyppolito, 2017). However, while this study shows a high level of open-set functioning with CIs in these children, our retrospective review does not permit us to determine how much of the improvement is due to the CIs. It is reasonable to expect that after almost three years of listening experience, children with residual hearing would show some growth in their auditory skills.

There are several limitations to this study. Consistent with clinical populations, there were variations in age at testing, age at CI surgery, and duration of CI use across the children.

We reported most recent outcomes for children from the clinical charts and duration of implant use was not taken into account in this study. An important limitation of our study is the lack of a control group of children with residual hearing without CIs, therefore, we can not necessarily conclude that a CI alone led to the improvements. In addition, data were collected from only one CI program. Since CI programs have different definitions and practices related to residual hearing, CI candidacy decisions and consequently the characteristics of children with residual hearing who receive CIs may differ between programs. Future research is needed to replicate the findings in different centers. Data were limited to audiological and clinical characteristics and other areas (i.e., spoken language abilities, progress in therapy, social functioning, academic achievement, classroom and social functioning, and listening fatigue) (Fitzpatrick et al., 2009) that are important to families were not examined. For this reason, further research is needed to explore the overall functioning of children beyond auditory functioning.

Conclusion

This study provides comprehensive population-based data on the audiological characteristics of children with residual hearing who received CIs in one region over a 26-year period. Our findings also contribute information on speech perception from a larger clinical population than previous studies. These findings can provide additional insights into the profiles of children who receive CIs. This information may assist in explaining differences in individual outcomes for children with residual hearing and provide new perspectives on decisions for CI candidacy. However, there is still limited information about this population in everyday functioning, such as communication, classroom and social participation. Future research should focus on a better understanding of these children's functioning to assist families in decision-making.

References

Adunka, O. F., Dillon, M. T., Adunka, M. C., King, E. R., Pillsbury, H. C., & Buchman, C. A. (2013). Hearing preservation and speech perception outcomes with electric-acoustic stimulation after 12 months of listening experience. *Laryngoscope, 123*(10), 2509–2515. https://doi.org/10.1002/lary.23741

Adunka, O. F., Buss, E., Clark, M. S., Pillsbury, H. C., & Buchman, C. A. (2008). Effect of preoperative residual hearing on speech perception after cochlear implantation. *Laryngoscope, 118*(11), 2044–2049. https://doi.org/10.1097/MLG.0b013e3181820900

British Cochlear Implant Group ENT UK. (2007). *Cochlear implats for deafness in children and adults*. British Academy of Audiology.

Carlson, M. L., O'Connell, B. P., Lohse, C. M., Driscoll, C. L., & Sweeney, A. D. (2018). Survey of the American Neurotology Society on cochlear implantation. *Otol Neurotol, 39*(1), e12–e19. https://doi.org/10.1097/MAO.0000000000001631

Carlson, M. L., Sladen, D. P., Haynes, D. S., Driscoll, C. L., DeJong, M. D., Erickson, H. C., Sunderhaus, L. W., Hedley-Williams, A., Rosenzweig, E. A., Davis, T. J., & Gifford, R. H. (2015). Evidence for the expansion of pediatric cochlear implant candidacy. *Otol Neurotol, 36*(1), 43–50. https://doi.org/https://dx.doi.org/10.1097/MAO.0000000000000607

Ching, T. Y. C., & Hill, M. (2007). The Parents' Evaluation of Aural/Oral Performance of Children (PEACH) scale: Normative data. *J Am Acad Audiol,* 18(3), 220–235. https://doi.org/10.3766/jaaa.18.3.4

Chiossi, J. S. C., & Hyppolito, M. A. (2017). Effects of residual hearing on cochlear implant outcomes in children: A systematic-review. *Int J Pediatr OtorhiInt J Pediatr Otorhinolaryngol, 100,* 119–127. https://doi.org/10.1016/j.ijporl.2017.06.036

Coninx, F., Weichbold, V., Tsiakpini, L., Autrique, E., Bescond, G., Tamas, L., Compernol, A., Georgescu, M., Koroleva, I., Le Maner-Idrissi, G., Liang, W., Madell, J., Mikić, B., et al. (2009). Validation of the LittlEARS® Auditory Questionnaire in children with normal hearing. *Int J Pediatr Otorhinolaryngol,* 73(12), 1761–1768. https://doi.org/10.1016/j.ijporl.2009.09.036

de Kleijn, J. L., van Kalmthout, L. W. M., van der Vossen, M. J. B., Vonck, B. M. D., Topsakal, V., & Bruijnzeel, H. (2018). A systematic review to define the audiologic thresholds for pediatric cochlear implant candidacy. *J Hear Sci 144*(7), 630–638. https://doi.org/ 10.1001/jamaoto.2018.0652

De Raeve, L., & Wouters, A. (2013). Accessibility to cochlear implants in Belgium: State of the art on selection, reimbursement, habilitation, and outcomes in children and adults. *Cochlear Implants Int, 14*(1), 18–25. https://doi.org/10.1179/1467010013Z.00000000078

Dettman, S. J., Dowell, R. C., Choo, D., Arnott, W., Abrahams, Y., Davis, A., Dornan, D., Leigh, J., Constantinescu, G., Cowan, R., & Briggs, R. J. (2016). Long-Term communication outcomes for children receiving cochlear implants younger than 12 months: A multicenter study. *Otol Neurotol, 37*(2), e82-95. https://doi.org/10.1097/MAO.0000000000000915

Dunn, C. C., Perreau, A., Gantz, B., & Tyler, R. S. (2010). Benefits of localization and speech perception with multiple noise sources in listeners with a short-electrode cochlear implant. *J Am Acad Audiol, 21*(1), 44–51. https://doi.org/10.3766/jaaa.21.1.6

Dowell, C., Hollow, R., & Winton, E. (2004). Outcomes for cochlear implant users with significant residual hearing. *Arch Otolaryngol Head Neck Surg, 130*(May), 575–581. https://doi.org/10.1001/archotol.130.5.575

Eshraghi, A. A., Ahmed, J., Krysiak, E., Ila, K., Ashman, P., Telischi, F. F., Angeli, S. A., Prentiss, S., Martinez, D., & Valendia, S. (2017). Clinical, surgical, and electrical factors impacting residual hearing in cochlear implant surgery. *Acta Oto-Laryngologica*, *137*(4), 384–388. https://doi.org/10.1080/00016489.2016.1256499

Erber, N. (1982). Auditory training. *Alexander Graham Bell Association for the Deaf.*

Fitzpatrick, E. M., Ham, J., & Whittingham, J. (2015). Pediatric Cochlear Implantation: Why Do Children Receive Implants Late?. *Ear Hear*, *36*(6), 688–694. https://doi.org/https://dx.doi.org/10.1097/AUD.0000000000000184

Fitzpatrick, E. M., McCrae, R., & Schramm, D. (2006). A retrospective study of cochlear implant outcomes in children with residual hearing. *BMC Ear Nose Throat Disord*, *6*(7), 1–6. https://doi.org/10.1186/1472-6815-6-7

Fitzpatrick, E., Olds, J., Durieux-Smith, A., McCrae, R., Schramm, D., & Gaboury, I. (2009). Pediatric cochlear implantation: How much hearing is too much? *Int J Audiol*, *48*(2), 91–97. https://doi.org/10.1080/14992020802516541

Geers, A. E., & Moog, J. S. (1987). Predicting spoken language acquisition of profoundly hearing-impaired children. *J Speech Lang Hear Res*, *52*(1), 84–94. https://doi.org/10.1044/jshd.5201.84

Gratacap, M., Thierry, B., Rouillon, I., Marlin, S., Garabedian, N., & Loundon, N. (2015). Pediatric cochlear implantation in residual hearing candidates. *Ann Otol Rhinol Laryngol*, *124*(6), 443–451. https://doi.org/10.1177/0003489414566121

Gifford, R. H. (2011). Who is a cochlear implant candidate? *Hear J*, *64*(6), 16–18. https://doi.org/10.1097/01.HJ.0000399149.53245.b1

Haskins H. (1949). *A phonetically balanced test of speech discrimination for children*.

Northwestern University, Evanston, IL.

Holcomb, M., & Smeal, M. (2020). Pediatric cochlear implantation: Who is a candidate in 2020? *Hear J, 73*(7), 8–9. https://doi.org/10.1097/01.HJ.0000689404.85842.2e

Hughes, M. L., Neff, D. L., Simmons, J. L., & Moeller, M. P. (2014). Performance outcomes for borderline cochlear implant recipients with substantial preoperative residual hearing. *Otol Neurotol, 35*(8), 1373–1384. https://doi.org/10.1097/MAO.0000000000000367

Hyde, M., Punch, R., & Komesaroff, L. (2010a). A comparison of the anticipated benefits and received outcomes of pediatric cochlear implantation: parental perspectives. *American Annals of the Deaf, 155*(3), 322–338. https://doi.org/10.1353/aad.2010.0020

Hyde, M., Punch, R., & Komesaroff, L. (2010b). Coming to a decision about cochlear implantation: Parents making choices for their deaf children. *J Deaf Stud Deaf Educ, 15*(2), 162–178. https://doi.org/10.1093/deafed/enq004

Kirk, K. I., Pisoni, D. B., & Osberger, M. J. (1995). Lexical effects on spoken word recognition by pediatric cochlear implant users. *Ear Hear, 16*(5), 470–481. https://doi.org/10.1038/jid.2014.371

Leigh, J., Dettman, S., Dowell, R., & Sarant, J. (2011). Evidence-based approach for making cochlear implant recommendations for infants with residual hearing. *Ear Hear, 32*(3), 313–322. https://doi.org/10.1097/AUD.0b013e3182008b1c

Leigh, J. R., Dettman, S. J., & Dowell, R. C. (2016). Evidence-based guidelines for recommending cochlear implantation for young children: Audiological criteria and optimizing age at implantation. *Int J Audiol, 55*(2), S9–S18. https://doi.org/10.3109/14992027.2016.1157268

Meredith, M. A., Rubinstein, J. T., Sie, K. C. Y., & Norton, S. J. (2017). Cochlear implantation

in children with postlingual progressive steeply sloping high-frequency hearing loss. *J Am Acad Audiol, 28*(10), 913–919. https://doi.org/10.3766/jaaa.16115

Michael, R., Attias, J., & Raveh, E. (2019). Cochlear implantation and social-emotional functioning of children with hearing loss. *J Deaf Stud Deaf Educ*, *24*(1), 25–31. https://doi.org/10.1093/deafed/eny034

Moog, J. S., & Geers, A. E. (1991). Educational management of children with cochlear implants. *Am Ann Deaf, 136*(2), 69–76. https://doi.org/10.1353/aad.2012.1348

Moog, J. S. (2002). Changing expectations for children with cochlear implants. *Ann Otol Rhinol Laryngol*, *111*(5 II), 138–142. https://doi.org/10.1177/00034894021110s527

Moteki, H., Nishio, S., Miyagawa, M., Tsukada, K., Iwasaki, S., & Usami, S. (2017). Long-term results of hearing preservation cochlear implant surgery in patients with residual low frequency hearing. *Acta Oto-Laryngologica, 137*(5), 516–521. https://doi.org/10.1080/00016489.2016.1252061

Nicholas, J. G., & Geers, A. E. (2007). Will they catch up? The role of age at cochlear implantation in the spoken language development of children with severe to profound hearing loss. *J Speech Lang Hear Res*, *50*(4), 1048–1062. https://doi.org/ 10.1044/1092-4388(2007/073)

Nilsson, M., Soli, S. D., & Sullivan, J. A. (1994). Development of the Hearing In Noise Test for the measurement of speech reception thresholds in quiet and in noise. *J Acoust Soc Am*. https://doi.org/10.1121/1.408469

Niparko, J. K., Tobey, E. A., Thal, D. J., Eisenberg, L. S., Wang, N.-Y., Quittner, A. L., & Fink, N. E. (2010). Spoken language development in children following cochlear implantation. *JAMA*, *303*(15), 1498–1506. https://doi.org/10.1001/jama.2010.451

O'Connor, A., Stacey, D., & Boland, L. (2015). *Ottawa Decision Support Tutorial*. The Ottawa Hospital Research Institute.

Osberger, M. J., Zimmerman-Phillips, S., & Koch, D. B. (2002). Cochlear implant candidacy and performance trends in children. *Ann Otol Rhinol Laryngol*. https://doi.org/10.1177/00034894021110s513

Raine, C. (2013). Cochlear implants in the United Kingdom: Awareness and utilization. *Cochlear Implants Int*. https://doi.org/10.1179/1467010013Z.00000000077

Ross, M., & Lerman, J. (1970). A picture identification test for hearing-impaired children. *J Speech Lang Hear Res*, *13*, 44–53. https://doi.org/10.1044/jshr.1301.44

Sarant, J. Z., Harris, D. C., & Bennet, L. A. (2015). Academic outcomes for school-aged children with severe–profound hearing loss and early unilateral and bilateral cochlear implants. *J Speech Lang Hear Res*, *58*, 1017–1032. https://doi.org/10.1044/2015_JSLHR-H-14-0075

Sarant, J. Z., Harris, D. C., Galvin, K. L., Bennet, L. A., Canagasabey, M., & Busby, P. A. (2018). Social development in children with early cochlear implants: Normative comparisons and predictive factors, including bilateral implantation. *Ear Hear*, *39*(4), 770–782. https://doi.org/10.1097/AUD.0000000000000533

Skarzynski, H. (2012). Ten years experience with a new strategy of partial deafness treatment. *J Hear Sci, 2*(2), 11–18.

Skarzynski, H., Lorens, A., Piotrowska, A., & Podskarbi-Fayette, R. (2009). Results of partial deafness cochlear implantation using various electrode designs. *Audiol Neurootol*, *14 Suppl 1*, 39–45. https://doi.org/https://dx.doi.org/10.1159/000206494

Sweeney, A. D., Hunter, J. B., Carlson, M. L., Rivas, A., Bennett, M. L., Gifford, R. H., Noble, J. H., Haynes, D. S., Labadie, R. F., & Wanna, G. B. (2016). Durability of hearing

preservation after cochlear implantation with conventional-length electrodes and scala tympani insertion. *Otolaryngol Head Neck Surg, 154*(5), 907–913. https://doi.org/10.1177/0194599816630545

Sweeney, A. D., Carlson, M. L., Zuniga, M. G., Bennett, M. L., Wanna, G. B., Haynes, D. S., & Rivas, A. (2015). Impact of perioperative oral steroid use on low-frequency hearing preservation after cochlear implantation. *Otol Neurotol, 36*(9), 1480–1485. https://doi.org/10.1097/MAO.0000000000000847

Teagle, H. F. B., Park, L. R., Brown, K. D., Zdanski, C., & Pillsbury, H. C. (2019). Pediatric cochlear implantation: A quarter century in review. *Cochlear Implants Int*, *20*(6), 288–298. https://doi.org/10.1080/14670100.2019.1655868

Thoutenhoofd, E. (2006). Cochlear implanted pupils in Scottish schools: 4-year school attainment data (2000-2004). *J Deaf Stud Deaf Educ*, *11*(2), 171–188. https://doi.org/10.1093/deafed/enj029

Trimble, K., Rosella, L. C., Propst, E., Gordon, K. A., Papaioannou, V., & Papsin, B. C. (2008). Speech perception outcome in multiply disabled children following cochlear implantation: Investigating a predictive score. *J Am Acad Audiol*, *19*(8), 602–611. https://doi.org/10.3766/jaaa.19.8.4

Wilson, K., Ambler, M., Hanvey, K., Jenkins, M., Jiang, D., Maggs, J., & Tzifa, K. (2016). Cochlear implant assessment and candidacy for children with partial hearing. *Cochlear Implants Int*, *17*(1), 66–69. https://doi.org/10.1080/14670100.2016.1152014

Yoshinaga-Itano, C., Sedey, A. L., Wiggin, M., & Mason, C. A. (2018). Language outcomes improved through early hearing detection and earlier cochlear implantation. *Otol Neurotol*. https://doi.org/10.1097/MAO.0000000000001976

Zanetti, D., Nassif, N., & Redaelli De Zinis, L. O. (2015). Factors affecting residual hearing preservation in cochlear implantation. *Acta Otorhinolaryngol Ital, 35*(6), 433–441. https://doi.org/10.14639/0392-100X-619

Zimmerman-Phillips, S., Osberger, M. J., & Robbins, A. M. (2001). Infant-Toddler Meaningful Auditory Integration Scale. In *Advanced Bionics Corp*.

Table 1. Assessment tools used in the clinical protocol in the CI program at CHEO

Speech perception measures *	
Open-set tests	
Phonologically Balanced Kindergarten [PBK]	Haskins, 1949
Hearing in Noise Test for Children [HINT-C]	Nilsson et al., 1994
Glendonald Auditory Screening Procedure [GASP]	Erber, 1982
Lexical Neighborhood Tests [LNT]	Kirk et al., 1995
Multisyllabic Lexical Neighborhood Tests [MLNT]	Kirk et al., 1995
Closed-set tests	
Early Speech Perception [ESP]	Moog & Geers, 1991
Word Intelligibility by Picture Identification [WIPI]	Ross & Lerman, 1970
Parent auditory performance questionnaires *	
Infant-Toddler Meaningful Auditory Integration Scale [IT-MAIS]	Zimmerman-Phillips et al., 2001
LittlEARS® Auditory Questionnaire [LittlEARS]	Coninx et al., 2009
Parents' Evaluation of Aural/Oral Performance of Children [PEACH]	Ching & Hill, 2007

* All scores are reported as percent (%) correct

Table 2. Modified Pediatric Ranked Order Speech Perception (PROSPER) Score

Modified Pediatric Ranked Order Speech Perception Score [PROSPER]		Modified PROSPER category score
Blank	Did not test	Blank
0	Could not test	0 (Could not test)
1	IT-MAIS[a] <50%	1 (Parent questionnaires)
2	IT-MAIS ≥50%	
3	LittlEARS <50%	
4	LittlEARS ≥50%	
5	ESP[b] standard monosyllable <50%	2 (Closed-set to easy open-set word tests)
6	ESP standard monosyllable ≥50%	
7	WIPI[c] <50%	
8	WIPI ≥50%	
9	GASP[d] word <50%	
10	GASP word ≥50%	
11	MLNT[d] word <50%	3 (LNT open-set word tests)
12	MLNT word ≥50%	
13	LNT[f] word <50%	
14	LNT word ≥50%	
15	PBK[g] word <50%	4 (PBK open-set word tests)
16	PBK word ≥50%	

[a] Infant-Toddler Meaningful Auditory Integration Scale
[b] Early Speech Perception
[c] Word Intelligibility by Picture Identification
[d] Glendonald Auditory Screening Procedure
[e] Multisyllabic Lexical Neighborhood Test
[f] Lexical Neighborhood Test
[g] Phonetically Balanced Kindergarten

Table 3. Clinical characteristics of children (n=389) with cochlear implant(s)

	Total (n=389)		Residual hearing (n=100)		Bilateral profound hearing loss (n=289)	
Sex (female), n (%)	167	(42.9)	42	(42.0)	125	(43.4)
Route to identification, n (%)						
Screened	126	(32.4)	52	(52.0)	74	(25.6)
Not screened	112	(28.8)	27	(27.0)	85	(29.4)
Unknown	151	(38.8)	21	(21.0)	130	(45.0)
Onset of hearing loss, n (%)						
Congenital	114	(29.3)	29	(29.0)	85	(29.4)
Early Onset	46	(11.8)	17	(17.0)	29	(10.0)
Late Onset	44	(11.3)	14	(14.0)	30	(10.4)
Acquired	30	(7.7)	3	(3.0)	27	(9.4)
Unknown	155	(39.8)	37	(37.0)	118	(40.8)
Etiology, n (%)						
Unknown	168	(43.2)	31	(31.0)	137	(47.4)
Known						
ENT malformation [a]	25	(6.4)	16	(16.0)	9	(3.1)
Familial/genetic	69	(17.7)	22	(22.0)	47	(16.3)
Syndromic	33	(8.5)	8	(8.0)	25	(8.7)
NICU admission [b]	52	(13.4)	19	(19.0)	33	(11.4)
Prenatal infection	14	(3.6)	1	(1.0)	13	(4.5)
Meningitis	25	(6.4)	3	(3.0)	22	(7.6)
Chemotherapy	3	(0.8)	0	(0.0)	3	(1.0)
Age at diagnosis (months), median (IQR)	12.2	(5.6, 22.0)	13.0	(3.6, 25.0)	12.2	(6.2, 21.7)
Age at hearing aid fitting (months), median (IQR)	15.1	(7.9, 25.9)	14.9	(6.6, 24.9)	15.6	(9.6, 26.0)
Age at CI candidacy (months), median (IQR)	21.7	(11.1, 46.2)	35.8	(16.6, 88.3)	18.7	(10.3, 37.5)
Age at CI surgery (months), median (IQR)	41.5	(23.0, 84.2)	46.2	(29.3, 94.0)	39.4	(22.1, 81.0)
Time diagnosis to surgery (months), median (IQR)	20.0	(8.9, 50.0)	29.6	(11.8, 61.4)	16.7	(7.8, 46.8)
Degree of hearing loss at diagnosis (Better ear), n (%)						

Within normal range (Unilateral hearing loss)	8	(2.1)	5	(5.0)	3	(1.0)
Mild	9	(2.3)	3	(3.0)	6	(2.1)
Moderate	25	(6.4)	18	(18.0)	7	(2.4)
Moderate-severe	34	(8.7)	17	(17.0)	17	(5.9)
Severe (PTA ≥70 and ≤90 dB HL)	72	(18.5)	38	(38.0)	34	(11.8)
Profound (PTA >90 dB HL) [c]	210	(54.0)	10	(10.0)	200	(69.2)
Unknown	31	(8.0)	9	(9.0)	22	(7.6)
Degree of hearing loss at 1st CI surgery (Better ear), n (%)						
Single-sided deafness	1	(0.3)	1	(1.0)	0	(0.0)
Moderate	7	(1.8)	7	(7.0)	0	(0.0)
Moderate-severe	10	(2.6)	10	(10.0)	0	(0.0)
Severe (PTA ≥70 and ≤90 dB HL)	82	(21.1)	82	(82.0)	0	(0.0)
Profound (PTA >90 dB HL)	289	(74.3)	0	(0.0)	289	(100.0)

N, number; ENT, ear nose throat; IQR, interquartile range; NICU, neonatal intensive care unit; PTA, pure-tone average

[a], cochlear anomalies, enlarged vestibular aqueduct syndrome (EVAS), and mondini malformation;

[b], NICU does not include children with syndromic hearing loss or ENT anomaly;

[c], Based on initial auditory brainstem response (ABR)

Table 4. Characteristics of 83 children by pre-CI Modified PROSPER category

Modified PROSPER category (n)	Total (n=83)	0 (n=15)	1 (n=21)	2 (n=11)	3 (n=2)	4 (n=34)
		Could not test	Parent Questionnaires	Closed-set /easy open-set	LNT open-set	PBK open-set
Age at Pre-CI assessment (months), median (IQR)	45.8 (22.7, 104.0)	23.5 (11.1, 34.8)	19.6 (13.8, 24.5)	45.8 (32.0, 103.6)	75.1 (39.5, 110.8)	104.1 (69.8, 150.1)
Age at CI surgery, (months), median (IQR)	46.2 (29.2, 93.6)	30.0 (12.1, 43.1)	25.6 (14.8, 30.9)	46.1 (36.7, 109.1)	18.6 (48.2, 115.0)	93.6 (58.3, 152.7)
Age at Post-CI assessment [a] (months), median (IQR)	115.2 (69.3, 161.5)	92.2 (56.1, 126.1)	80.0 (39.0, 95.2)	128.9 (71.1, 161.4)	196.5 (158.0, 235.0)	154.8 (108.9, 183.5)
Duration of CI use [b] (months), median (IQR)	34.3 (19.0, 75.3)	55.9 (30.3, 92.3)	25.9 (18.9, 73.6)	27.4 (6.8, 93.0)	114.9 (109.8, 120.1)	30.7 (14.0, 54.2)
PTA at surgery in better ear (dB HL), median (IQR)	81.6 (73.8, 86.7)	80.8 (76.3, 86.9)	85.0 (76.7, 86.7)	80.0 (54.2, 84.2)	90.0 (90.0, 90.0)	79.1 (73.3, 86.3)
PTA at surgery in CI ear (dB HL), median (IQR)	88.3 (81.7, 96.5)	85.8 (77.5, 96.7)	88.3 (85.0, 92.9)	93.3 (84.2, 93.3)	90.0 (90.0, 90.0)	87.5 (78.7, 97.7)
PTA at surgery in non-CI ear (dB HL), median (IQR)	81.7 (75.0, 88.3)	84.6 (79.2, 90.0)	86.7 (78.3, 89.6)	80.0 (55.8, 80.0)	90.0 (90.0, 91.7)	80.0 (73.3, 86.3)

[a], most recent result

[b], time CI surgery to post-CI assessment

[c], one child with missing data

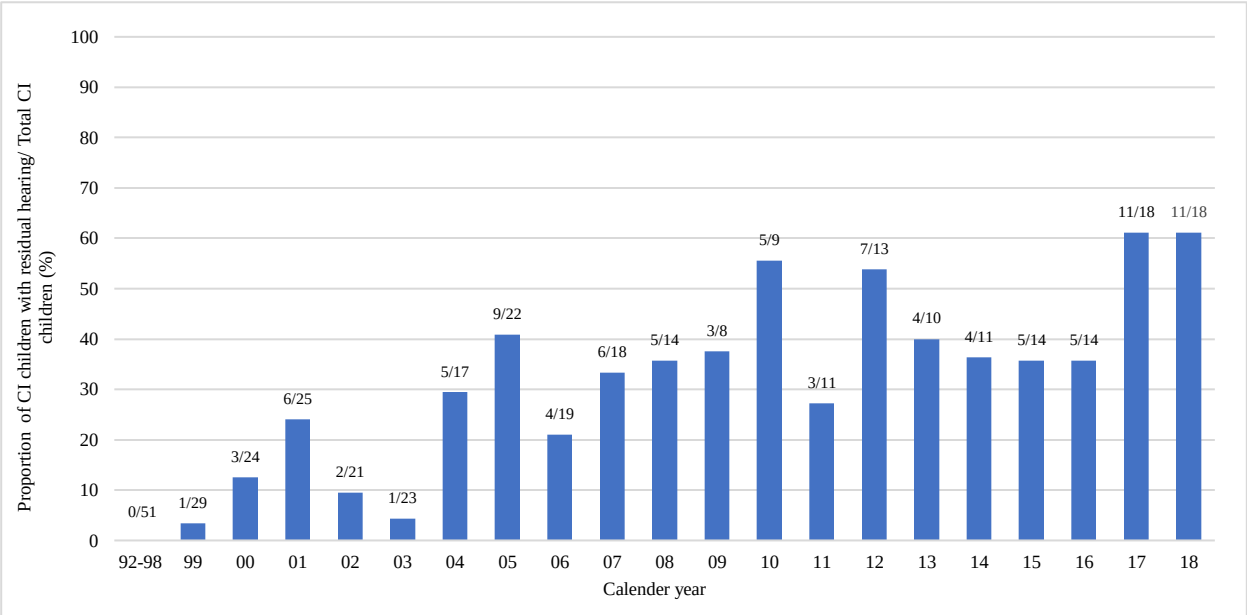
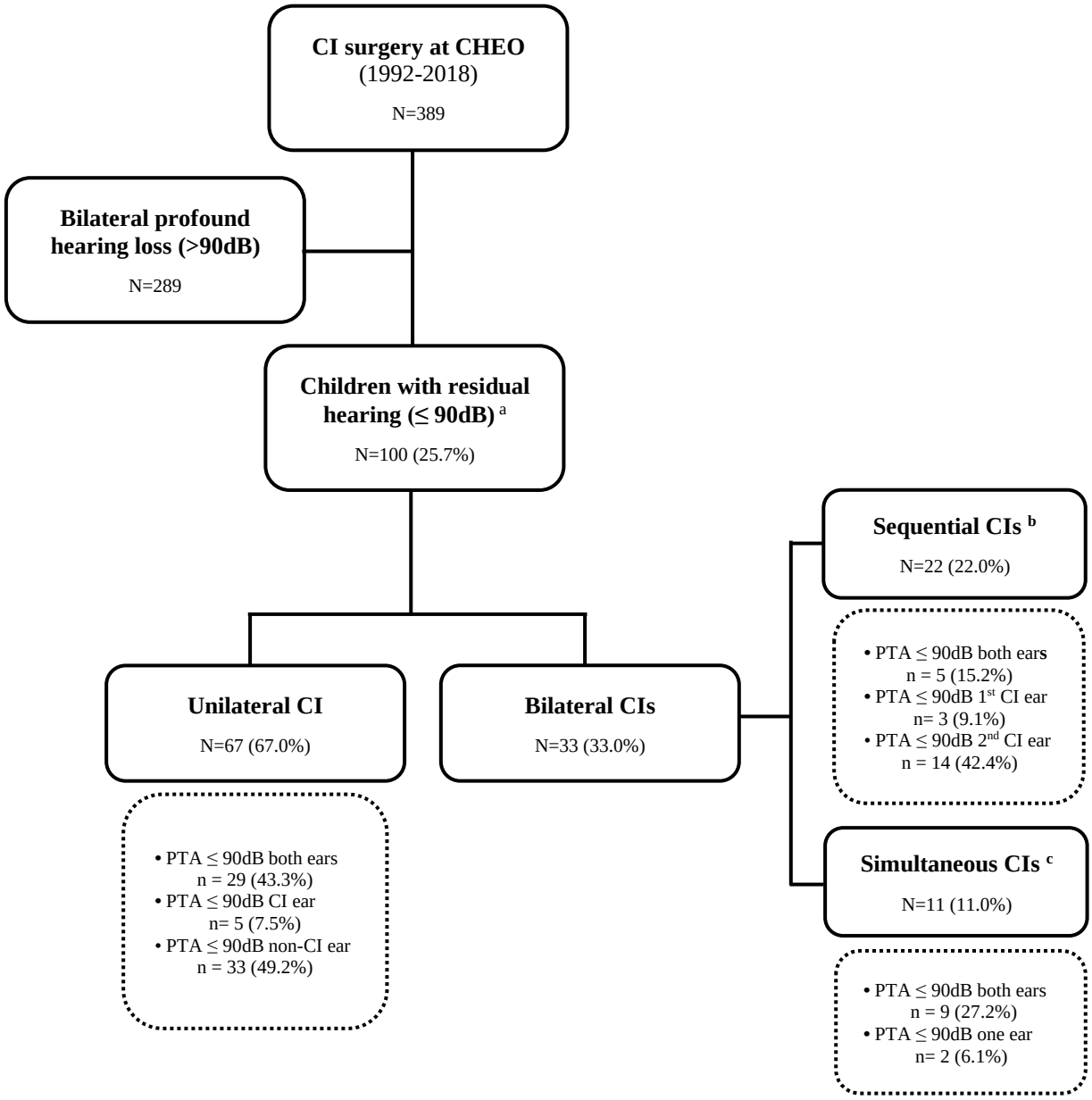
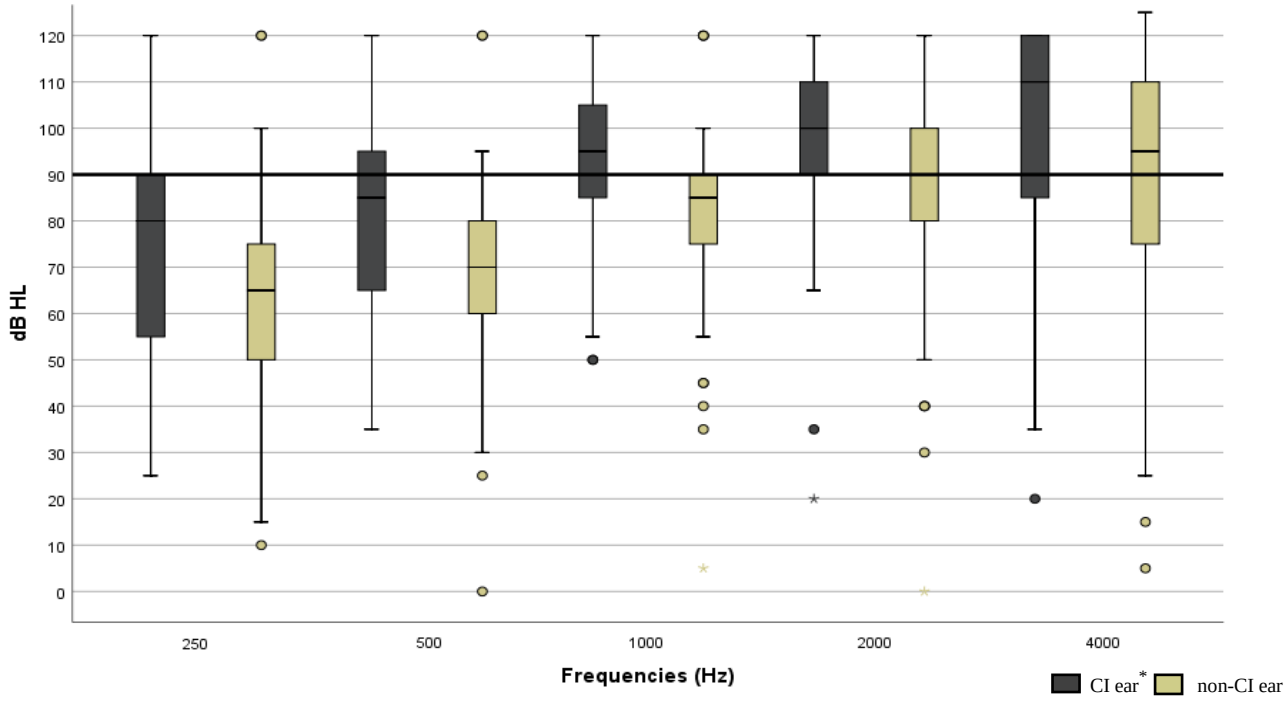


Figure 1. Proportion of children with residual hearing receiving CIs relative to total number of children implanted (1992-2018)



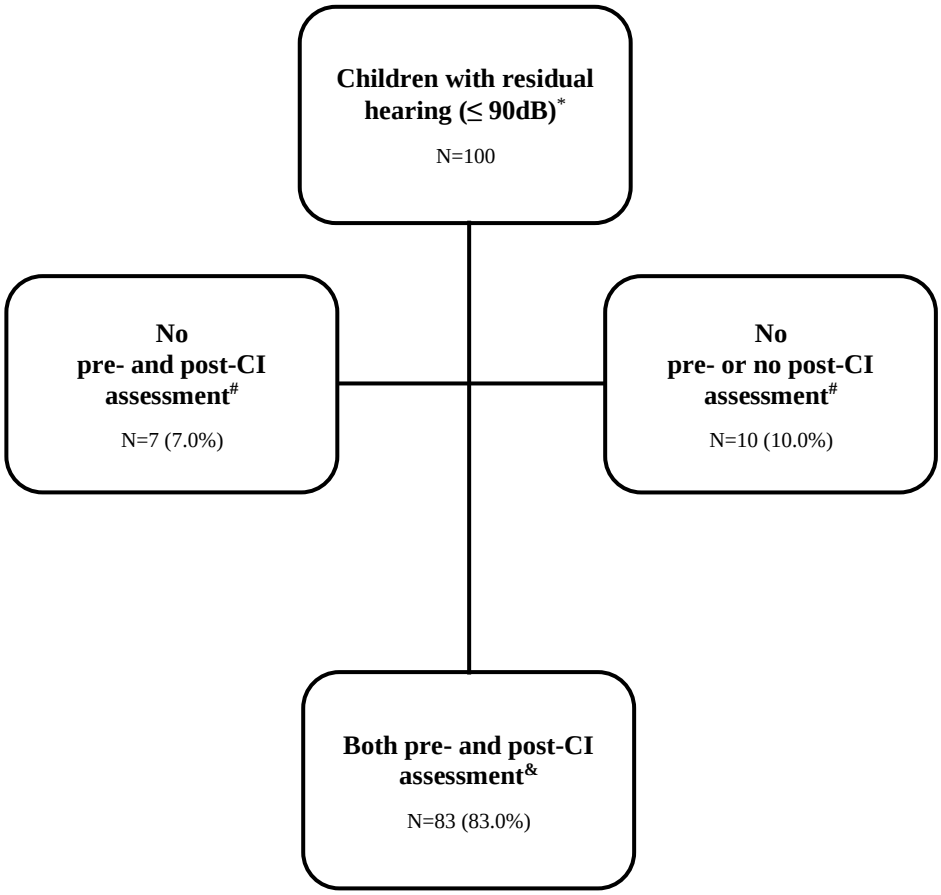
N, number; CI, cochlear implant
[a], in at least one ear, at 500, 1000, 2000 Hz
[b], each ear was implanted in two separate surgeries (over 6 months apart)
[c], both ears were implanted at the same time or less than6 months apart)

Figure 2. Selection of study participants



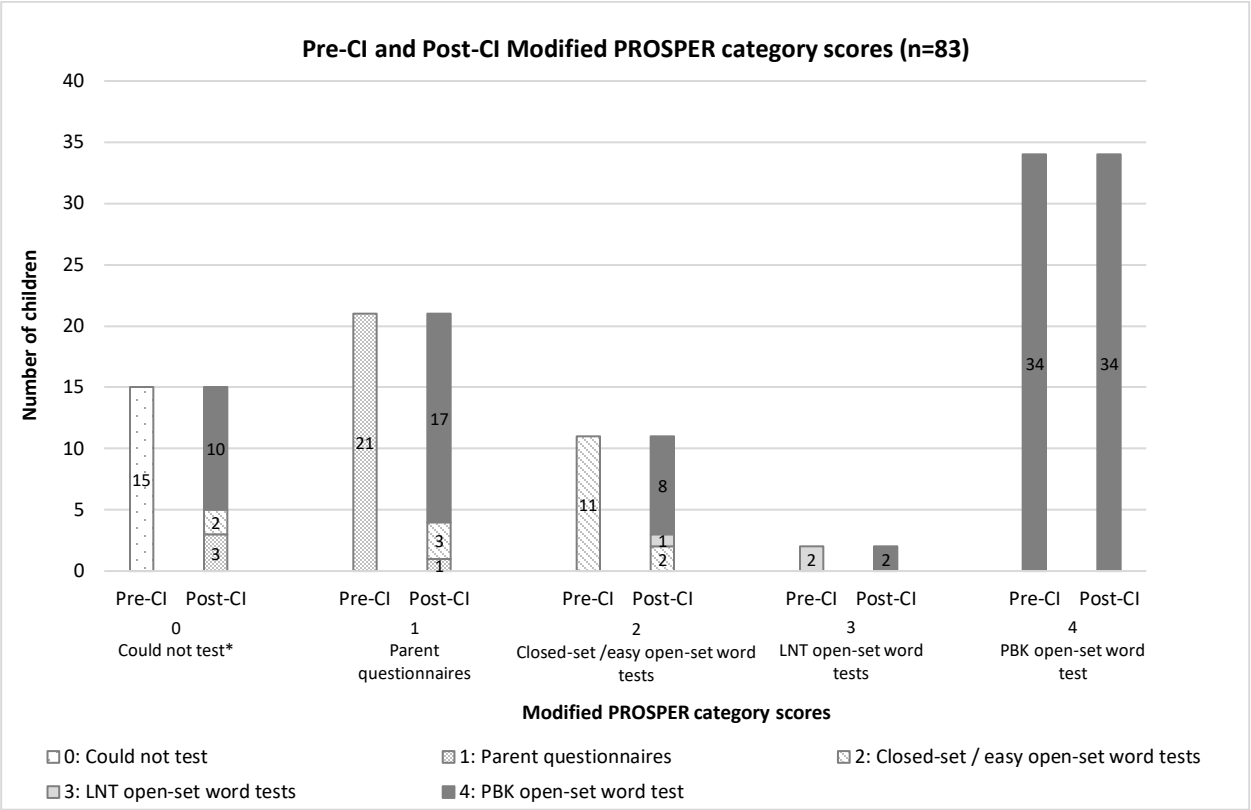
* For the 22 children who received sequential bilateral CIs, the first ear was selected as the CI ear for this calculation
Figure does not include 11 children with simultaneous bilateral CIs because CI and non-CI ears are not applicable
The boxplots show the distribution of individual thresholds for both ears, the box height represents the inter-quartile range with the median indicated by the line within the box. Outliners in the distribution are indicated by the circles

Figure 3. Preoperative thresholds for CI ears and non-CI ears (n=89 children) in children with unilateral CI (n=67) and children with sequential bilateral CIs (n=22)



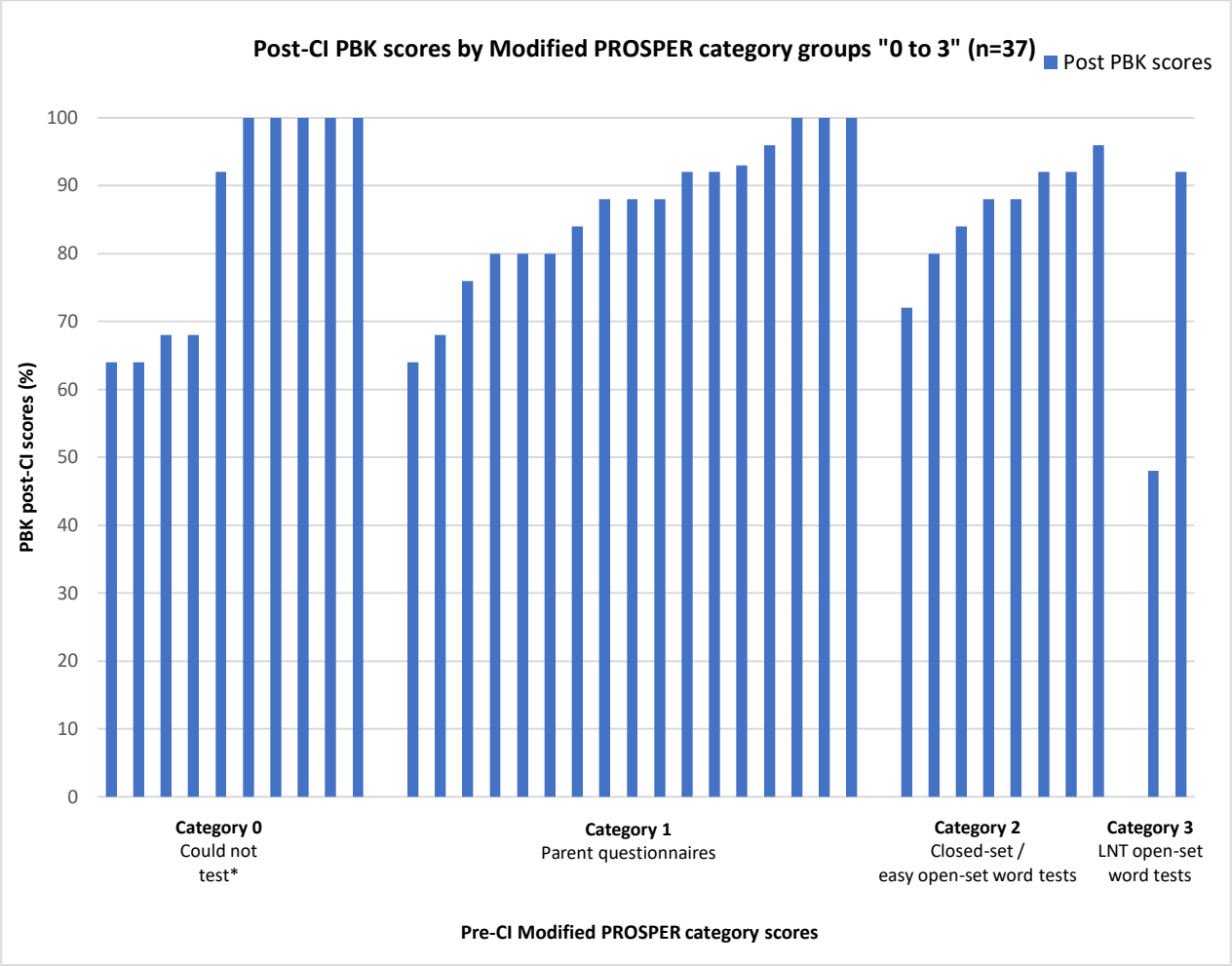
CI, cochlear implant
* PTA in at least one ear (average of thresholds at 500, 1000, 2000 Hz)
# Children had incomplete or missing tests
& Includes 15 children assigned a pre-CI score of 0%

Figure 4. Selection of children with speech perception outcomes



* Numbers on bar graphs refer to number of children with the modified PROSPER category score Children who could not complete pre-CI assessments and had clear documentation of no auditory abilities (n=15) were assigned a pre-CI speech perception score of 0%

Figure 5. Distribution of pre- and post-CI Modified PROSPER category scores



** Assigned score 0%*

Figure 6. Comparison of post-CI PBK test scores to pre-CI Modified PROSPER category scores (0-3) in 37 children

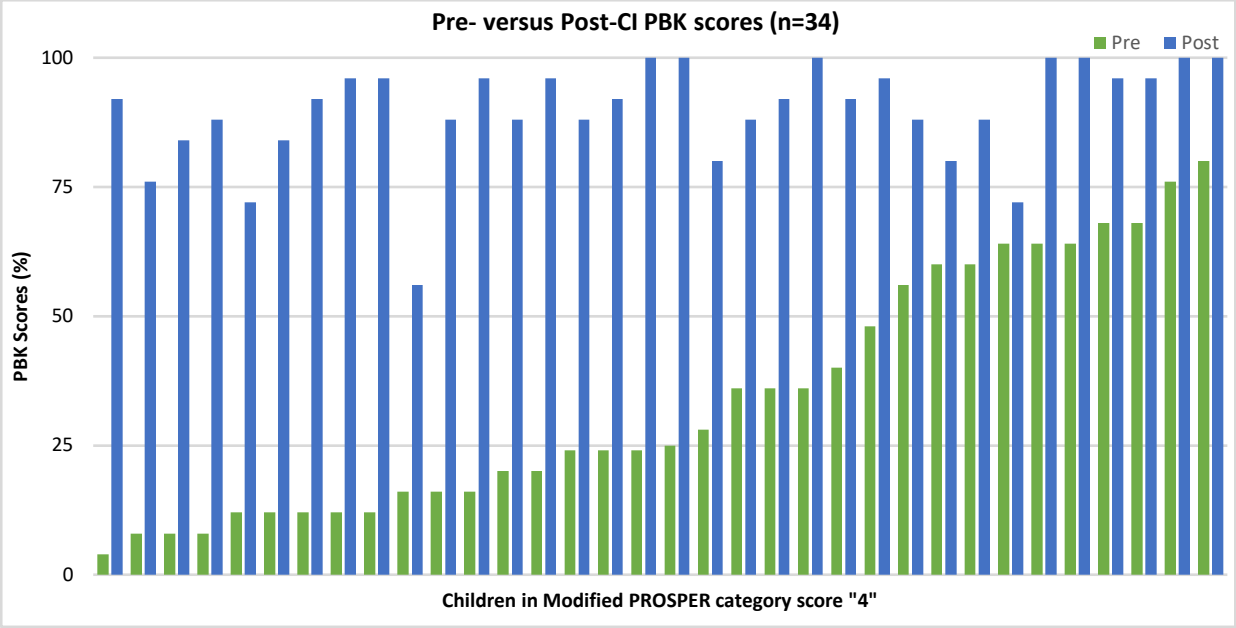


Figure 7. Pre-CI and post-CI PBK scores in 34 children

Chapter 3: Systematic review

Benefits and risks related to cochlear implantation for children with residual hearing: A systematic review

Manuscript submitted and formatted for the International Journal of Audiology

Abstract

Objective: This study aimed to synthesize information concerning the potential benefits and risks related to cochlear implants (CIs) versus hearing aids (HAs) in children with residual hearing.

Design: A systematic review of articles published from January 2003 to January 2019 was conducted.

Study sample: Our review included studies that compared the benefits and risks of CIs versus HAs in children (≤ 18years old) with residual hearing. A total of 3265 citations were identified; 8 studies met inclusion criteria.

Results: Children with CIs showed significantly better speech perception scores post-CI than pre-CI. There was limited evidence related to improvement in everyday auditory performance, and the results showed non-significant improvement in speech intelligibility. One study on social-emotional functioning suggested benefits from CIs. In four studies, 37.2% (16/43) of children showed loss of residual hearing and 14.0% (8/57) of children had discontinued or limited use of their CI or HA.

Conclusions: Overall, CIs provide improvement in speech perception outcomes for children with residual hearing but there is little evidence to guide decision-making related to other areas of development. It will be important to conduct further research of both benefits and risks of CIs in this specific population to facilitate decision-making.

Keywords: cochlear implantation; residual hearing; speech perception; pediatric cochlear implant; systematic review

Introduction

Cochlear implantation for children with severe-profound hearing loss has led to positive outcomes in speech and language acquisition, academic achievement, and social development (Ching et al., 2018; Dettman et al., 2016; Leigh et al., 2013; Thoutenhoofd, 2006). Since 1990 when FDA first approved cochlear implantation for children, the candidacy criteria have expanded to include children with substantially more hearing than FDA-indicated cochlear implant (CI) candidacy criteria (i.e., pure-tone average [PTA] ≤ 90 dB HL at 500, 1000, 2000 Hz). Several studies have also found positive outcomes of CIs for children with residual hearing, indicating that they can develop better speech recognition outcomes with CIs than with hearing aids (HAs) (Dettman et al., 2004; Fitzpatrick et al., 2006; Gratacap et al., 2015).

However, risks and negative consequences are associated with CI surgery. Device failure is one of the major risks following CI surgery and is the most common cause of revision surgery (Blanchard et al., 2015; Brown et al., 2009; Lassig et al., 2005). Children with residual hearing are also at risk of losing residual hearing, which complicates the decision for their parents (Bergeron, 2000; Zanetti et al., 2015). For these reasons, parents of this specific population may be uncertain about whether the potential benefits outweigh the risks (Greaver et al., 2017; Hardonk et al., 2010, 2011), often making CI decision-making for these families more challenging than for those of children with bilateral profound hearing loss (Fitzpatrick et al., 2009; Hyde et al., 2010).

According to a systematic review (Chiossi & Hyppolito, 2017), children with residual hearing obtain positive language-related outcomes after CIs. However, there is limited evidence in other areas such as social-emotional functioning and academic achievement. Providing comprehensive evidence including both the risks and benefits following CIs to families is

important. Furthermore, updated information considering newer device technology and surgical techniques is required because CI device technology and surgical techniques for preserving residual hearing have improved in the last few years (Moteki et al., 2017; Snels et al., 2019).

The aim of our systematic review was to synthesize the benefits and risks of CIs compared to those of HAs in children who have residual hearing. The research questions addressed in this review were: 1) What are the benefits (i.e., speech perception, speech intelligibility, social-emotional functioning, academic achievement, and psychosocial development) of children with residual hearing who received CIs, compared to their pre-implant outcomes or compared to children with HAs? and 2) What are the risks and negative outcomes (i.e., medical complications, device-related issues, loss of residual hearing) of CIs compared to HAs for children with residual hearing?

Materials and methods

The protocol for this systematic review was registered in the PROSPERO review registry (#CRD42018114134, https://www.crd.york.ac.uk/PROSPERO), an international database of prospectively registered systematic review protocols in health and social care (Page et al., 2018; Schiavo, 2019). The report for this systematic review was prepared according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) checklist (Moher et al., 2009).

Search strategy

A comprehensive search strategy was developed in collaboration with a librarian who has expertise in conducting systematic reviews. The strategy was reviewed by a second librarian

using the Peer Review of Electronic Search Strategies (PRESS) template (McGowan et al., 2010, 2016; Sampson et al., 2009) (Appendix B). The search strategy was developed for MEDLINE and adjusted for other databases (Appendix C). The electronic databases MEDLINE (via Ovid), Embase (via Ovid), CINAHL (via EBSCO Host), Linguistics and Language Behavior Abstracts (LLBA) (via Proquest), SpeechBITE, PsycINFO (via Ovid), and Cochrane Central Register of Controlled Trials (CENTRAL) (via Ovid) were searched. Articles published from 2003 to January 2019 were accessed.

Eligibility criteria

The criteria for inclusion in this review followed the Population, Intervention, Comparison, Outcomes, and Study Design (PICOS) format.

Population

Children with hearing loss up to age 18 years and usable residual hearing (i.e., pure-tone average [PTA] ≤ 90 dB HL at 500, 1000, 2000 Hz, or as defined in the articles) were included. Articles including children and adults were retained if the mean age of participants was ≤ 18 years or the age of the majority of participants was ≤ 18 years and the results for children were presented separately. Studies that included all degrees of hearing loss were included only if the majority of participants had residual hearing or if results were presented separately for children with residual hearing. We also included articles that described participants as having 'residual hearing, high-frequency, low-frequency, partial hearing, or asymmetrical hearing,' even if the average hearing was higher than 90 dB HL. Studies were excluded if they reported outcomes for

children with hearing loss with other disabilities that prevented the measurement of outcomes targeted for this review.

Interventions

We included children who received unilateral CI, bilateral CIs, or who were bimodal users (CI+HA).

Comparisons

This review included studies comparing outcomes before and after CI and studies comparing outcomes of children with HAs to those with CIs.

Outcomes

Benefits. Primary outcomes: The primary outcomes for this review were: 1) auditory performance, 2) speech perception, 3) receptive and expressive language, 4) vocabulary, and 5) other aspects of communicative competence such as appropriate use of language, grammar knowledge, and communication repair.

Secondary outcomes: Studies exploring other developmental outcomes including academic, behaviour, social, or cognitive function and other related consequences (e.g., self-esteem, quality of life) were included.

Risks. Risks were considered to be negative outcomes related to 1) loss of residual hearing, 2) surgery-specific complications, 3) medical-specific complications, 4) device issues, and 5) other consequences (e.g., lower self-esteem, lower quality of life).

Study designs

We included randomized controlled trials, controlled clinical trials, and other quasi-experimental studies comparing HAs to CIs. This review also included prospective and retrospective studies comparing pre- and post-CI outcomes and comparing HA to CI outcomes, cross-sectional studies with comparison groups, and case-control studies. Other study designs were excluded: case reports, case series, documents reporting expert opinions, conference abstracts, editorials and theses.

Time frame

Full-text literature published since 2003 was considered for inclusion. This time frame was selected because studies have emerged over the last 17 years on new CI electrode designs and new surgical techniques have been developed to preserve residual hearing (e.g., Adunka et al., 2004; Eshraghi et al., 2003; Stöver et al., 2005).

Language

Only studies published in English were included. As previous studies reported, there is no evidence of a systematic bias from the use of language restrictions in systematic review and meta-analyses in health-related studies (Moher et al., 2000; Morrison et al., 2009).

Study selection

Literature search results were compiled and stored in a reference manager software, Mendeley. The citations were then exported to systematic review software, Covidence

(covidence systematic review software, http://www.covidence.org) and duplicate records were eliminated.

Two researchers first piloted screening of titles and abstracts with 20 retrieved articles following the PICOS strategy. The titles and abstracts of all records were then screened to determine if they met inclusion criteria. All potentially relevant literature was retrieved for full-text screening using Covidence. Two researchers calibrated the screening by conducting full-text screening on 10% of the literature before independent full-text screening was begun. Disagreements between researchers were resolved through discussion or consultation with a third senior researcher as needed.

Data extraction

One researcher extracted all the information, and a trained research assistant verified the data. A data extraction form (Appendix D) was developed in Microsoft Excel to extract pre-determined data from each study. Extracted information included: 1) study characteristics (e.g., author names, year of publication, country, title of the study, title of the journal, and source of funding), 2) study design, 3) characteristics of the population (e.g., sample size, sex, age at diagnosis, and degree of hearing loss), 4) details of interventions, 5) details of comparison groups, 6) outcomes including speech perception, other areas of auditory performance, receptive and expressive language, vocabulary, communicative competence, loss of residual hearing, surgery-specific complications, medical-specific complications, device-related issues, academic achievement, behaviour, social integration, cognitive function, and other related consequences, and 7) author definitions of residual hearing.

Assessment of methodological quality

We assessed methodological quality for all selected studies using the Quality Assessment Tool for Quantitative Studies, developed by the Effective Public Health Practice Project at McMaster University (Effective Public Health Practice Project, 2010; Thomas et al., 2004). This tool was used to assess quasi-experimental, cohort, case-control, and cross-sectional studies and results in an overall methodological rating of studies based on eight elements: selection bias, study design, confounders, blinding, data collection methods, withdrawals and dropouts, the integrity of intervention, and study analysis. These eight elements are each rated as 1 (strong), 2 (moderate), or 3 (weak), and are combined to yield overall methodology ratings of strong, moderate, or weak evidence. One researcher independently assessed the methodological quality of each study, and a second reviewer verified the assessment. If there were any disagreements between ratings, decisions were resolved through discussion or by consulting a third researcher.

Data synthesis

A summary of the findings, including study characteristics and study data, was presented in tabular form using descriptive analysis. Data were presented as percentages, medians, interquartile ranges, and means and standard deviations, where possible. Data synthesis of included studies depended on the heterogeneity of studies retrieved. Due to the clinical heterogeneity related to outcome measures, ages of children, definitions of residual hearing, a meta-analysis was not possible (Ioannidis et al., 2008).

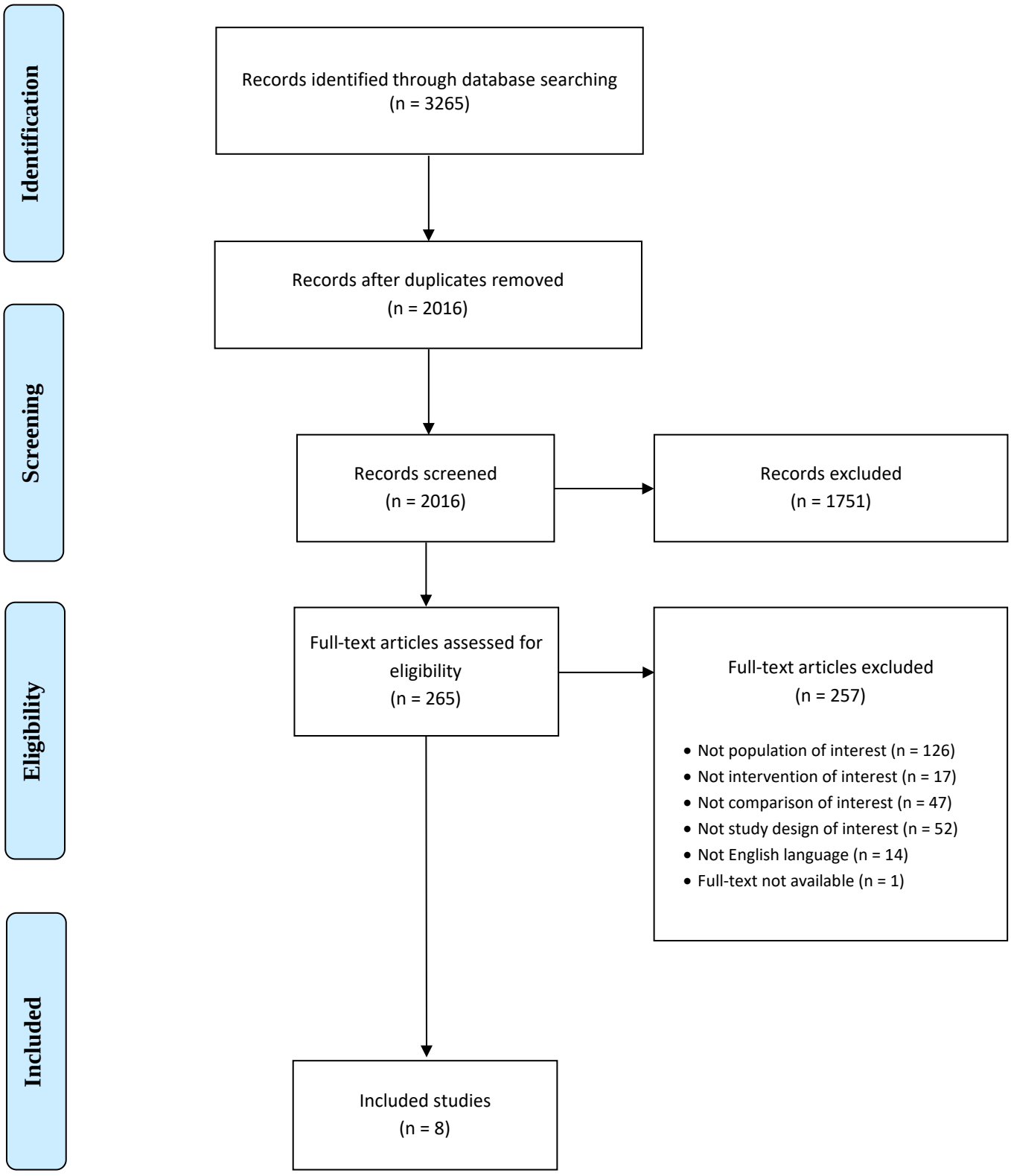


Figure 1. PRISMA Flow diagram of included studies

Table 1. Study characteristics

Study, year, country	Study design	Study objective(s)	Age at Dx Mean ± SD [range] (y)	Age at device fitting Mean ± SD [range] (y)	Duration of device use Mean ± SD [range] (y)	Threshold of Pre-CI better ear Mean ± SD [range] (dB HL)	Intervention (n)	Comparison (n)	Methodological Quality
Dettman et al., 2004, Australia	Retrospective cohort	To evaluate postoperative speech perception benefits in children whose speech perception scores exceeded conservative candidacy guidelines	NR [a]	10.3± 2.4 [6.4-16.7]	1.14± 0.5 [0.5-2.1]	94.5± 10.1* [73-110]	Bimodal (CI+HA) (16)	Pre-CI (16)	Moderate
Fitzpatrick et al., 2006, Canada	Retrospective cohort	To examine speech recognition outcomes in children who were regarded as borderline candidates for cochlear implantation	1.79±1.8* [0.1-4.8]	7.7± 3.2 [4.1-14.6]	2.7± 1.4* [1.0-5.4]	90.3± 7.2* [78-98]	Unilateral CI (10)	Pre-CI (10)	Moderate
Gratacap et al., 2015, France	Retrospective cohort	To review the outcomes of cochlear implantation in children with different residual hearing conditions	4.6±3.2 [0.8-14.0]	10.2± 4.9* [2.5-18.0]	5.4± 1.5 [1.0-5.9]	101.2 ± 9.3* [71-125]	Unilateral CI (53)	Pre-CI (53)	Moderate
Hughes et al., 2014, United States	Retrospective cohort	To examine whether children with substantial preoperative residual hearing obtained more benefit from a CI than from a HA	6.6±4.6* [0.5-14.3]	13.5± 3.3* [7.9-18.6]	2.6± 2.6* [0.5-9.7]	70.3± 21.8* [33.3-93.3]	Bimodal (CI+HA) or Unilateral CI (10)	Pre-CI (10)	Weak
Michael et al., 2019, Israel	Cross-sectional	To examine differences in social-emotional functioning among children with and without CIs	NR [b]	CI 3.9± 3.3 [1.0-15.0] HA 3.2±2.2 [0.2-7.5]	CI 6.7 ± 3.3 HA NR	CI [61-90] HA [40-90]	Unilateral CI (23) Bilateral CI (8)	HAs (32)	Weak

Rezaei et al., 2017, Iran	Cross-sectional	To evaluate and compare verbal comprehension in children with CIs and HAs	CI [c] 0.95 HA [d] 0.93	CI 2.4 HA 2.5	CI 6.8 HA 6.9 (*p*=.348)	CI 88.7 [70-90] HA 88.3 [70-90] (*p*=.052)	Unilateral CI (15)	HAs (15)	Weak
Sadadcharam et al., 2016, United Kingdom	Retrospective cohort	To investigate the benefit of unilateral CI in children currently outside of the CI audiological guidelines in the contralateral ear	NR [e]	5.2 [1.1-14.3]	[1.0-4.0] [f]	[50-90] at 2000 and 4000 Hz	Unilateral CI (47)	Pre-CI (47)	Moderate
Wilson et al., 2016, United Kingdom	Retrospective cohort	To examine and compare hearing preservation, auditory performance, and speech intelligibility in children with residual hearing with CI	NR	6.0 [1.1-12.0]	[0.5-7.0]	<65 dB HL at one or more frequencies between 250-1000 Hz [g]	Bilateral CI (5) Unilateral CI (23)	Pre-CI (28)	Weak

N, number; CI, cochlear implant; Dx, diagnosis; y, year; dB HL, decibels hearing level; Hz, hertz; HA, hearing aid; NR, not reported

[*]mean was calculated from individual data

[a], study only presented mean age at onset of profound hearing loss, which was 1.30±1.3 years

[b], age of hearing loss detection ranged between birth and 4 years among the CI users and between birth to 7 years among the HA users

[c], 75% congenital HL, 25% prelingual HL

[d], 70.8% congenital HL, 29.2% prelingual HL

[e], average age at referral was 4.1 years (range from 1 month to 11 years 9 months)

[f], post-operative CAP scores reported in the article reflect the most recent CAP scores at the annual review (ranging from 12 months to 4 years)

[g], all children in study had hearing thresholds of >90 dB HL at 2000 and 4000 Hz

Results

A PRISMA flow chart (Liberati et al., 2009; Moher et al., 2009) of included studies is presented in Figure 1. A total of 3265 citations were obtained through the initial search. After removing 1249 duplicates, titles and abstracts of the remaining 2016 articles were screened. After excluding 1751 articles that did not meet the inclusion criteria, the full texts of 265 articles were assessed, and of these, 257 documents were excluded. Three primary reasons for the exclusion of these articles were: 1) not the population of interest (n=126), 2) not the study design of interest (n=52), and 3) not the comparison of interest (n=47). A total of eight articles met the inclusion criteria for this study.

Study Characteristics

Table 1 summarizes the characteristics of the eight included studies. Six were retrospective cohort studies that were conducted specifically to compare pre- and post-CI outcomes, and two were cross-sectional studies that included a comparison group (HA). All eight articles reported benefits following CIs for children with residual hearing, including speech perception (Dettman et al., 2004; Fitzpatrick et al., 2006; Gratacap et al., 2015; Hughes et al., 2014), other area of auditory performance (Sadadcharam et al., 2016; Wilson et al., 2016), speech intelligibility (Rezaei et al., 2017; Wilson et al., 2016), and social-emotional functioning (Michael et al., 2019). One study each was conducted in Canada (Fitzpatrick et al., 2006), Australia (Dettman et al., 2004), France (Gratacap et al., 2015), the United States (Hughes et al., 2014), Iran (Rezaei et al., 2017), and Israel (Michael et al., 2019), and two were conducted in the United Kingdom (Sadadcharam et al., 2016; Wilson et al., 2016). The sample sizes ranged from 10 to 63 children.

As shown in Table 2, residual hearing was defined in six studies according to audiometric thresholds and/or speech perception abilities (Dettman et al., 2004; Fitzpatrick et al., 2006; Gratacap et al., 2015; Hughes et al., 2014; Sadadcharam et al., 2016; Wilson et al., 2016). The two remaining studies (Michael et al., 2019; Rezaei et al., 2017) only considered PTA thresholds. Specifically, in four studies (Dettman et al., 2004; Fitzpatrick et al., 2006; Gratacap et al., 2015; Hughes et al., 2014), candidacy selection was based on speech perception scores of greater than 30% in open-set word tests or greater than 50% in sentence test scores. The remaining four studies (Michael et al., 2019; Rezaei et al., 2017; Sadadcharam et al., 2016; Wilson et al., 2016) selected candidates with residual hearing based on their audiometric thresholds; two of these studies considered CI candidacy based on three frequency PTA $\leq$ 90 dB, and the other two considered the degree of hearing in individual frequencies or low frequency average (250, 500, 1000Hz).

Quality assessment

The results of the quality assessments are reported in Table 1. The Quality Assessment Tool for Quantitative Studies (Effective Public Health Practice Project, 2010) was used to assess the quality of all eight studies. Applying the rating criteria for this tool, if there was at least one "weak" rating among the above eight evaluation areas, the study was given an overall rating of moderate, and two or more "weak" evaluation areas resulted in a weak overall rating. Four studies (Dettman et al., 2004; Fitzpatrick et al., 2006; Gratacap et al., 2015; Sadadcharam et al., 2016) were rated as moderate and the other four (Hugh et al., 2014; Michael et al., 2019; Rezaei et al., 2017; Wilson et al., 2016) as weak. For all studies, the rating was lowered due to some information (i.e., selection bias, blinding, confounders, withdrawals and drop-outs) not being

explicitly reported. In particular, in all eight studies, the examiners scoring each child's performance were not blind to device use (HAs or CIs), thus reducing the quality rating.

Table 2. Definitions of residual hearing

Study, year	Hearing Thresholds	Speech perception
Dettman et al., 2004		Open-set sentence scores ≥ 30%
Fitzpatrick et al., 2006	Bilateral PTA < 90 dB HL at 500, 1000, 2000 Hz	Open-set word scores ≥ 20% Open-set sentence scores ≥ 50%
Hughes et al., 2014	Hearing threshold ≤ 70 dB HL at 2 or more frequencies	Open-set sentence scores > 50% in CI ear or >60% in the best-aided condition
Sadadcharam et al., 2016	Hearing threshold between 50 and 90 dB HL at 2000 and 4000 Hz in the non-CI ear	
Wilson et al., 2016	Hearing threshold < 65 dB HL at one or more frequencies between 250 and 1000 Hz	
Michael et al., 2019	PTA between 61 and 90 dB HL	
Rezaei et al., 2017	PTA between 70 and 90 dB HL	
Gratacap et al., 2015	1. Hearing thresholds < 60 dB HL at 250 to 500 Hz and > 70 dB HL at 2000 to 4000Hz 2. Mean aided PTA (500, 1000, 2000, and 4000 Hz) < 50 dB HL, and open-set word scores < 50% with a HA or > 30% differences in open-set word scores between silent and noise conditions 3. PTA in left/right ear difference > 30 dB HL with a HA 4. PTA drop > 40 dB HL in at least one ear since children's early auditory assessments 5. PTA changes > 15 dB HL at one or more frequencies between 500, 1000, or 2000 Hz at least 2 times within the 12 months before CI	

PTA, pure-tone average; dB HL, decibel hearing level; Hz, hertz; CI, cochlear implant; HA, hearing aid

Table 3. Benefits of CI in children with residual hearing

Study, year	Outcome Measures		Intervention – Comparison	Main Outcomes	Statistical Information (*p*-value)
Speech Perception					
Dettman et al., 2004	PBK_{ph} (mean %)		Post-CI – Pre-CI	79.8 vs. 54.2	Significant $p < .001$
	CNCw + PBKw (mean±SD %)			53.4 vs. 22.8	Significant $p < .001$
Fitzpatrick et al., 2006	PBKw (mean±SD %)		Post-CI – Pre-CI	76.0 vs. 16.0	Significant*
Gratacap et al., 2015			Post-CI – Pre-CI	80.7±23.0 vs. 47.9±33.8	Significant $p < .001$
Hughes et al., 2014	PBKph+PBKw+ HINT-C+AzBio (%)		Post-CI – Pre-CI	5/10 of participants improved (range, 15-55%) 4/10 of participants showed no changes or decreased their scores (range, -32-3%) 1/10 data inconclusive	NR
Dettman et al., 2004	BKB (mean %)	Live voice	Post-CI – Pre-CI	87.3 vs. 53.8	Significant $p < .001$
		Quiet		85.4 vs. 36.0	Significant $p < .005$
		+10 SNR		61.2 vs. 28.0	Significant $p < .050$
Fitzpatrick et al., 2006	HINT-C Live voice (mean %)		Post-CI – Pre-CI	93.5 vs. 58.0	Significant*
Auditory Performance					
Sadadcharam et al., 2016	CAP (mean±SD score) [a]		Post-CI – Pre-CI	7.22±1.1 vs. 5.02±1.1	Significant*
Wilson et al., 2016			Post-CI – Pre-CI	6.07±1.8 vs. 3.71±1.7	NR
Speech Intelligibility					

Rezaei et al., 2017	Test of speech intelligibility level (mean±SD %) [b]		CI – HA	72.31±23.42 vs. 68.94±17.58	Not significant $p = .901$
Wilson et al., 2016	SIR (mean score) [c]		Post-CI – Pre-CI	4.10±1.0 vs. 2.63±1.3	NR
Social-Emotional Functioning					
Michael et al., 2019	SDQ [d] (mean±SD score)	Total difficulties [e]	CI – HA	8.83±4.11 vs.10.25±4.68	Not significant p [f] = .210
		Conduct problems		1.30±1.21 vs. 1.56±1.52	Not significant p [f] = .450
		Emotional symptoms		3.03±2.52 vs. 2.40±2.01	Not significant p [f] = .320
		Hyperactivity /inattention		2.10±1.71 vs. 3.50±2.26	Significant p [f] < .010
		Peer problems		1.97±1.59 vs. 1.97±2.01	Not significant p [f] = .990
		Pro-social behavior		8.43±1.65 vs. 7.59±1.88	Significant p [f] < .050

PBK, Phonetically Balanced Kindergarten test; CNC, Consonant-Nucleus-Consonant test; Ph, phonemes; W, words; BKB, Bamford-Kowal-Bench Sentence Lists; NR, not reported; SNR, Signal-to-Noise Ratio; HINT-C, Hearing in Noise Test for Children; CAP, Categories of Auditory Performance; SIR, Speech Intelligibility Rating; SDQ, Strengths and Difficulties Questionnaire

[*] *p*-value not reported

[a], scores ranging from 1 to 9

[b], test was developed for use in Persian-speaking children aged 3 to 5 years with hearing loss

[c], scores ranging from 1 to 5

[d], smaller numbers correspond with more positive outcomes, except in pro-social behaviour / five items in each subtest

[e], scores for conduct problems, emotional symptoms, hyperactivity/inattention, and peer problems are summed

[f], p-values calculated from individual data

Outcomes

In the area of primary outcomes, speech perception was reported as an outcome in four studies (Dettman et al., 2004; Fitzpatrick et al., 2006; Gratacap et al., 2015; Hughes et al., 2014). Two studies each reported auditory performance (Sadadcharam et al., 2016; Wilson et al., 2016) and speech intelligibility (Wilson et al., 2016; Razaei et al., 2017). Risks following CIs, including loss of residual hearing and device non-use, were discussed in four of the eight studies (Gratacap et al., 2015; Wilson et al., 2016; Hughes et al., 2014; Sadadcharam et al., 2016). In the area of secondary outcomes, one study reported social-emotional functioning (Michael et al., 2019).

Benefits

The benefits of CI related to speech perception, auditory behaviour, and social-emotional functioning for children with residual hearing are shown in Table 3. No studies reported outcomes related to receptive and expressive language, vocabulary, communicative competence, or academic achievement.

Speech perception. Differences in speech perceptions pre- and post-CI were reported in four studies (Dettman et al., 2004; Fitzpatrick et al., 2006; Gratacap et al., 2015; Hughes et al., 2014). These studies used several assessment measures including the Phonetically Balanced Kindergarten Test (PBK) (Haskins, 1949), the Consonant-Nucleus-Consonant (CNC) words (Peterson & Lehiste, 1962), and the Fournier or Saussus-Boorsma lists (the French equivalent of the PBK test) (Fournier, 1951) for measuring open-set word recognition; and the Bamford-Kowal-Bench Sentence Lists (BKB) (Bench et al., 1979), the Hearing in Noise Test for Children

(HINT-C) (Nilsson et al., 1994), and the AzBio (Spahr et al., 2014) for open-set sentence recognition.

Three studies of moderate quality (Dettman et al., 2004; Gratacap et al., 2015; Fitzpatrick et al., 2006) reported that CIs provided significant benefits in speech perception. Post-CI speech perception scores showed improvement ranging from 25.6 to 60.0% compared to pre-CI scores. In particular, open-set word scores increased in post-CI (ranging from test scores of 53.4 to 80.7%) compared to pre-CI (ranging from 16.0 to 47.9%). Open-set sentence perception scores also showed an improvement post-CI (ranging from test scores of 87.3 to 93.5%) compared to pre-CI scores (ranging from 53.8 to 58.0%). One additional study of weak quality (Hughes et al., 2014) reported that 5 out of 10 participants showed improvement in the battery of speech perception assessments (PBKph, PBKw, HINT-C, AzBio), but no statistical information was provided.

Auditory performance. Two studies (one of moderate and one of weak quality) (Sadadcharam et al., 2016; Wilson et al., 2016) compared pre- and post-CI auditory performance on the Categories of Auditory Performance (CAP) test (Archbold et al., 1995), a measure that reflects everyday auditory performance. It consists of hierarchical scales ranging from 0: "No awareness of environmental sounds or voice" to 9: "Use of telephone with the unknown speaker in an unpredictable context." In both studies, post-CI scores (ranging from 6.07 to 7.22) showed an improvement compared to pre-CI scores (ranging from 3.71 to 5.02), but no statistical information was provided.

Speech Intelligibility. Two weak quality studies (Rezaei et al. 2017; Wilson et al., 2016) reported

speech intelligibility outcomes using the Test of Speech Intelligibility Level (Persian) and the Speech Intelligibility Rating (SIR) test (Cox & McDaniel, 1989). A cross-sectional study (Rezaei et al., 2017) reported that children with CIs showed scores that were 3.4% higher than those of children using HAs, but this improvement was not statistically significant *(p = .901).* The other pre-post comparison study (Wilson et al., 2016) used the SIR test to measure speech intelligibility scales ranging from 1: "Connected speech is unintelligible pre-recognizable words in the spoken language" to 5: "Connected speech is intelligible to all listeners and child is understood easily in everyday contexts". In this study, positive change was observed only for younger children, but statistical information was not reported.

Social-emotional Functioning. Social-emotional functioning was examined in one weak quality cross-sectional study (Michael et al., 2019) which used the Strengths and Difficulties Questionnaire (SDQ) test (Goodman, 1997). The SDQ consists of 25 items divided into five subtests, including conduct problems, emotional symptoms, hyperactivity/inattention, peer problems, and pro-social behaviour. Each item can be marked 0: "Not true", 1: "somewhat true", or 2: "certainly true". In this test, conduct problems, emotional symptoms, hyperactivity/inattention, and peer problems are all negative aspects of social-emotional functioning; therefore, smaller numbers correspond with more positive outcomes. In social-emotional functioning, children with CIs showed statistically lower levels of hyperactivity/inattention *(p <.010)* and significantly higher levels of pro-social behaviour *(p <.050)* compared with their peers with HAs. In addition, the total difficulties score of children with CIs was 8.83±4.11 (a score less than 10 means the child is unlikely to have social-emotional

problems) while the children with HAs scored a little over 10 (10.25±4.68). However, the difference in the total difficulties scores between the two groups was not statistically significant.

Risks

Table 4 presents the findings of two primary risks that were observed in four moderate and weak quality pre- and post-CI design studies.

Table 4. Risks of CI in children with residual hearing

Study, year	**Risks**	**Number (n)**	**Total number in study population (n)**	**Duration of CI use** Mean ± SD [range] (y)
Hearing Status				
Gratacap et al., 2015	Loss of residual hearing	3	5 [a]	5.4±1.5 [1.0-5.9]
Hughes et al., 2014	Complete loss of residual hearing	2	10	2.6±2.6 [b] [0.5-9.7]
	Partial loss of residual hearing	1		
Wilson et al., 2016	Partial loss of residual hearing [c]	7 [d]	28	[0.5-7.0]
	Almost complete loss of residual hearing [e]	3 [d]		
Device Use				
Hughes et al., 2014	CI non-use	2	10	2.6±2.6 [b] [0.5-9.7]
Sadadcharam et al., 2016	CI non-use	1	47	[1.0-4.0] [f]
	HA intermittent or non-use	5		

[a], 1 of 5 different groups; 5 of 53 children in study assessed for post-implant residual hearing
[b], mean calculated from individual data
[c], 25-75% hearing remains
[d], out of 22 children who had post-operative audiograms
[e], 1-25% hearing remains
[f], post-operative scores reported in the article reflect the most recent scores at the annual review (ranging from 12 months to 4 years)

Three studies (Gratacap et al., 2015; Hughes et al., 2014; Wilson et al., 2016) reported risks in hearing status; 16 of 43 (37.2%) children either partially or totally lost their residual hearing after the CI surgery. Negative outcomes related to device-use were reported in two studies (Hughes et al., 2014; Sadadcharam et al., 2016). A total of 3 of 57 (5.3%) children in these studies were reported to discontinue use of their CI device, and 5 of 57 (8.8%) children who used HAs in the non-implanted ear discontinued or only intermittently used their HA after surgery (Table 4). Reasons for discontinued use of CIs were not reported. Some children who became HA non-users indicated that they did not like the sound from their HAs. No decline in auditory functioning and no risks related to receptive and expressive language, vocabulary, communicative competence, academic achievement, or surgery-related complications were reported in these studies. No decline in auditory functioning and no risks related to receptive and expressive language, vocabulary, communicative competence, academic achievement, or surgery-related complications were reported in these studies.

Discussion

The primary goal of this review was to synthesize the benefits and risks related to CIs in children with residual hearing. A total of eight studies were included in this review. Benefits in speech perception and everyday auditory performance were documented in six studies of weak to moderate quality that compared pre-post CI performance (Dettman et al., 2004; Fitzpatrick et al., 2006; Gratacap et al., 2015; Hughes et al., 2014; Sadadcharam et al., 2016; Wilson et al., 2016). However, there remains relatively little information about other areas of development. Improvements in speech intelligibility were reported in two weak quality studies (Wilson et al., 2016; Razaei et al., 2017). In another weak quality study, two areas of social-emotional

functioning (hyperactivity/inattention and pro-social behaviour) showed better outcomes with CIs compared to HAs (Michael et al., 2019).

The findings from our systematic review are similar to those of a systematic review by Chiossi et al. (2017) on speech perception outcomes in children with residual hearing. Unlike the previous review, we only included studies that compared children with residual hearing pre-post implant or compared children with CIs to a comparison group using HAs. Using these comparisons provides a better estimate of how much improvement children with residual hearing obtain from CIs. In addition to speech perception, we found two studies that provided weak evidence for improvements in speech intelligibility (Wilson et al., 2016; Razaei et al., 2017). We also captured studies that included new surgical techniques and device technology that placed greater emphasis on the preservation of residual hearing, all of which were published after 2003.

Similar to Chiossi et al. (2017), we retrieved no studies related to cognitive outcomes in these children. Our review added only one study of weak quality on social-emotional development (Michael et al., 2019). The information from this study suggests that there may be better outcomes in some domains beyond speech perception for children with residual hearing. According to previous studies (Fitzpatrick et al., 2009; Zwolan & Sorkin, 2017; Holcomb & Smeal, 2020), the outcomes in multiple communication and developmental areas such as ease of listening, communication, and academic functioning are important for families of children with CIs and these may have added value for families of children with residual hearing. Overall, our review identified that there is some limited evidence to support the contribution of CIs to areas of development beyond speech perception.

Our review expands the previous systematic review (Chiossi et al., 2017) to include risks of CIs compared to HAs. There is extensive literature reporting the risks related to surgery for all

children who undergo cochlear implantation (e.g., loss of residual hearing, surgery-specific complications, medical-specific complications, and device-related issues) (Bhatia et al., 2004; Johnston et al., 2010; Loundon et al., 2010). However, our review provides specific information about deterioration in hearing and discontinued or limited use CI or HA in the non-implanted ear from four different moderate to weak quality studies. Total or partial loss of residual hearing was reported in approximately 40% (n=16) of 43 children in these studies (Gratacap et al., 2015; Hughes et al., 2014; Sadadcharam et al., 2016). Limited CI or HA use (in the non-implanted ear) was reported in 5.3% to 8.8% of children (Hughes et al., 2014; Sadadcharam et al., 2016). According to previous studies, 3 to 24% of children with profound hearing loss use their CIs less than full-time (Archbold et al., 2009; Contrera et al., 2014; Marnane & Ching, 2015; Özdemir et al., 2013). Our findings suggest that children with residual hearing on the contralateral side are not at greater risk of non-use or limited use of their hearing technology than children whose pre-implant hearing was in the severe to profound range bilaterally.

This comprehensive information about both benefits and risks from our review may assist parents in making informed CI decisions for children with residual hearing.

Implications

Our review, coupled with the findings of Chiossi et al. (2017) supports speech perception advantages of CIs over HAs and provides limited support for improvement in other areas of auditory performance in children with residual hearing. However, additional studies that investigate listening-related skills such as ease of listening, listening fatigue, cognitive load, and confidence in communicating through hearing would provide useful insights into other less frequently measured areas for this population of children. This review also contributes new

information about the loss of residual hearing and device use, which can assist in counselling parents of children with residual hearing who are faced with CI decisions. The lack of new information about the benefits of CIs for children with residual hearing in the areas of language, cognitive and academic development makes it difficult to provide any specific recommendations to assist practitioners in guiding families who raise questions about the advantage of CIs versus HAs in these areas. Future studies are required to provide higher quality evidence about the benefits and risks in multiple domains beyond speech perception that are of importance to parents of children with residual hearing.

Limitations

Our review included only studies published in English. There is variation in CI candidacy criteria and regulatory requirements in different countries. Given the interest in expanding CI candidacy criteria worldwide (Carlson et al., 2015; Fitzpatrick et al., 2009; Gifford, 2016; Leigh et al., 2011, 2016; Skarzynski et al., 2015; Zwolan et al., 2017), it could be useful to review studies conducted in other cultures and health care contexts which may have different candidacy criteria and different clinical practices. In addition, our review was restricted to only peer-reviewed studies, so there is a potential for results to be affected by publication bias because we did not include studies such as those presented at conferences or published in other languages.

Conclusion

To our knowledge, this is the first systematic review that considered both the benefits and risks of CIs compared to HAs in children with residual hearing. This review suggests that CIs provide more benefits in speech perception than HAs for children with residual hearing. These

children may also receive other benefits (e.g., in other areas of auditory performance, speech intelligibility, and social-emotional functioning) compared to HAs, although information in these areas is very limited. Our review also reported risks related to CIs for these children including the potential loss of residual hearing and discontinued and limited use of CI or HA. However, these findings are confined to a small number of studies or weak quality studies, making it difficult to draw conclusions about the results of CIs versus HAs. Given the increasing number of children with residual hearing who receive CIs, it will be critical to conduct more studies of high quality. Further examination of both the benefits and risks of CIs in this specific population can help inform evidence-based practices and facilitate decision-making for families of children with residual hearing who are considering CIs.

References

Adunka, O., Kiefer, J., Unkelbach, M. H., Lehnert, T., & Gstoettner, W. (2004). Development and evaluation of an improved cochlear implant electrode design for electric acoustic stimulation. *Laryngoscope, 114*(7), 1237–1241. https://doi.org/10.1097/00005537-200407000-00018

Archbold, S., Lutman, M. E., & Marshall, D. H. (1995). Categories of auditory performance. *Ann Otol Rhinol Laryngol, 166*, 312–314.

Archbold, S. M., Nikolopoulos, T. P., & Lloyg-Richmond, H. (2009). Long-term use of cochlear implant systems in paediatric recipients and factors contributing to non-use. *Cochlear Implants Int, 10*(1), 25–40. https://doi.org/10.1002/cii.363

Bench, J., Kowal, A., & Bamford, J. (1979). The BKB (Bamford-Kowal-Bench) sentence lists for partially-hearing children. *Br J Audiol, 13*(3). https://doi.org/10.3109/03005367909078884

Bergeron, F. (2000). Residual hearing following a cochlear implantation: Effect of time and device. *Adv Otorhinolaryngol, 57*, 389–392. https://doi.org/10.1159/000059188

Bhatia, K., Gibbin, K. P., Nikolopoulos, T. P., & O'Donoghue, G. M. (2004). Surgical complications and their management in a series of 300 consecutive pediatric cochlear implantations. *Otol Neurotol, 25*(5), 730–739.

Blanchard, M., Thierry, B., Glynn, F., De Lamaze, A., Garabedian, E. N., & Loundon, N. (2015). Cochlear implant failure and revision surgery in pediatric population. *Ann Otol Rhinol Laryngol, 124*(3), 227–231. https://doi.org/10.1177/0003489414551931

Brown, K. D., Connell, S. S., Balkany, T. J., Eshraghi, A. E., Telischi, F. F., & Angeli, S. A. (2009). Incidence and indications for revision cochlear implant surgery in adults and

children. *Laryngoscope*, *119*(1), 152–157. https://doi.org/10.1002/lary.20012

Carlson, M. L., Sladen, D. P., Haynes, D. S., Driscoll, C. L., DeJong, M. D., Erickson, H. C., Sunderhaus, L. W., Hedley-Williams, A., Rosenzweig, E. A., Davis, T. J., & Gifford, R. H. (2015). Evidence for the expansion of pediatric cochlear implant candidacy. *Otol Neurotol 36*(1), 43–50. https://doi.org/10.1097/MAO.0000000000000607

Ching, T. Y. C., Dillon, H., Leigh, G., Cupples, L., Ching, T. Y. C., Dillon, H., Leigh, G., & Cupples, L. (2018). Learning from the Longitudinal Outcomes of Children with Hearing Impairment (LOCHI) study: Summary of 5-year findings and implications. *Int J Audiol*, *57*(sup2), S105–S111. https://doi.org/10.1080/14992027.2017.1385865

Chiossi, J. S. C., & Hyppolito, M. A. (2017). Effects of residual hearing on cochlear implant outcomes in children: A systematic-review. *Int J Pediatr Otorhi*, *100*, 119–127. https://doi.org/10.1016/j.ijporl.2017.06.036

Contrera, K. J., Choi, J. S., Blake, C. R., Betz, J. F., Niparko, J. K., & Lin, F. R. (2014). Rates of long-term cochlear implant use in children. *Otol Neurotol*, *35*(3), 426–430. https://doi.org/10.1097/MAO.0000000000000243

Cox, R. M., & McDaniel, D. M. (1989). Development of the Speech Intelligibility Rating (SIR) test for hearing aid comparisons. *J Speech Lang Hear Res, 32*, 347–352. https://doi.org/10.1044/jshr.3202.347

Dettman, S. J., Dowell, R. C., Choo, D., Arnott, W., Abrahams, Y., Davis, A., Dornan, D., Leigh, J., Constantinescu, G., Cowan, R., & Briggs, R. J. (2016). Long-term communication outcomes for children receiving cochlear implants younger than 12 months: A multicenter study. *Otol Neurotol*, *37*(2),e82-95 https://doi.org/10.1097/MAO.0000000000000915

Dettman, S. J., Costa, W. A. D., Dowell, R. C., Winton, E. J., Hill, K. L., & Williams, S. S.

(2004). Cochlear implants for children with significant residual hearing. *Arch Otolaryngol Head Neck Surg.*, *130*(May), 612–618. https://doi.org/10.1001/archotol.130.5.612

Effective Public Health Practice Project. (2010). Quality assessment tool for quantitative studies. In *Effective Public Health Practice Project* (pp. 2–5).

Eshraghi, A. A., Yang, N. W., & Balkany, T. J. (2003). Comparative study of cochlear damage with three perimodiolar electrode designs. *Laryngoscope*, *113*(3), 415–419. https://doi.org/10.1097/00005537-200303000-00005

Fitzpatrick, E. M., McCrae, R., & Schramm, D. (2006). A retrospective study of cochlear implant outcomes in children with residual hearing. *BMC Ear Nose Throat Disord*, *6*(7), 1–6. https://doi.org/10.1186/1472-6815-6-7

Fitzpatrick, E., Olds, J., Durieux-Smith, A., McCrae, R., Schramm, D., & Gaboury, I. (2009). Pediatric cochlear implantation: How much hearing is too much? *Int J Audiol*, *48*(2), 91–97. https://doi.org/10.1080/14992020802516541

Fournier, J. (1951). Audiométrie vocale : les épreuves d'intelligibilité et leurs applications au diagnostic, à l'expertise et à la correction prothétique des surdités. In *Maloine*.

Gifford, R. H. (2016). Expansion of Pediatric Cochlear Implant Indications. *Hear J*, *December*, 8–10. https://doi.org/10.1097/01.HJ.0000511125.71672.3e

Goodman, R. (1997). The strengths and difficulties questionnaire: A research note. *J Child Psychol Psychiatry*, *38*, 581–586. https://doi.org/10.1111/j.1469-7610.1997.tb01545.x

Gratacap, M., Thierry, B., Rouillon, I., Marlin, S., Garabedian, N., & Loundon, N. (2015). Pediatric cochlear implantation in residual hearing candidates. *Ann Otol Rhinol Laryngol*, *124*(6), 443–451. https://doi.org/https://dx.doi.org/10.1177/0003489414566121

Greaver, L., Eskridge, H., & Teagle, H. F. B. (2017). Considerations for pediatric cochlear

implant recipients with unilateral or asymmetric hearing loss: Assessment, device fitting, and habilitation. *J Am Acad Audiol, 26*(2), 91–98. https://doi.org/10.1044/2016_AJA-16-0051

Hardonk, S., Bosteels, S., Desnerck, G., Loots, G., Van Hove, G., Van Kerschaver, E., Vanroelen, C., & Louckx, F. (2010). Pediatric cochlear implantation: A qualitative study of parental decision-making processes in Flanders, Belgium. *Am Ann Deaf, 155*(3), 339–352. https://doi.org/10.1353/aad.2010.0012

Hardonk, S., Daniels, S., Desnerck, G., Loots, G., Van Hove, G., Van Kerschaver, E., Sigurjónsdóttir, H. B., Vanroelen, C., & Louckx, F. (2011). Deaf parents and pediatric cochlear implantation: An exploration of the decision-making process. *Am Ann Deaf, 156*(3), 290–304. https://doi.org/10.1353/aad.2011.0027

Haskins H. (1949). *A phonetically balanced test of speech discrimination for children*. Northwestern University, Evanston, IL.

Holcomb, M., & Smeal, M. (2020). Pediatric Cochlear Implantation: Who is a Candidate in 2020? *Hear J, 73*(7), 8–9. https://doi.org/10.1097/01.HJ.0000689404.85842.2e

Hughes, M. L., Neff, D. L., Simmons, J. L., & Moeller, M. P. (2014). Performance outcomes for borderline cochlear implant recipients with substantial preoperative residual hearing. *Otol Neurotol, 35*(8), 1373–1384. https://doi.org/10.1097/MAO.0000000000000367

Hyde, M., Punch, R., & Komesaroff, L. (2010). Coming to a decision about cochlear implantation: Parents making choices for their deaf children. *J Deaf Stud Deaf Educ, 15*(2), 162–178. https://doi.org/10.1093/deafed/enq004

Ioannidis, J. P. A., Patsopoulos, N. A., & Rothstein, H. R. (2008). Reasons or excuses for avoiding meta-analysis in forest plots. *BMJ*, *336*(7658), 1413–1415.

https://doi.org/10.1136/bmj.a117

Johnston, J. C., Smith, A. D., Fitzpatrick, E., O'Connor, A., Angus, D., Benzies, K., & Schramm, D. (2010). Estimation of risks associated with paediatric cochlear implantation. *Cochlear Implants Int*, *11*(3), 146–169. https://doi.org/10.1002/cii.421

Lassig, A., Zwolan, T., & Telian, S. (2005). Cochlear implant failures and revision. *Otol Neurotol*, *26*(4), 624–634. https://doi.org/10.1097/01.mao.0000178123.35988.96

Leigh, J., Dettman, S., Dowell, R., & Sarant, J. (2011). Evidence-based approach for making cochlear implant recommendations for infants with residual hearing. *Ear Hear*, *32*(3), 313–322. https://doi.org/10.1097/AUD.0b013e3182008b1c

Leigh, J. R., Dettman, S. J., & Dowell, R. C. (2016). Evidence-based guidelines for recommending cochlear implantation for young children: Audiological criteria and optimizing age at implantation. *Int J Audiol*, *55 Suppl 2*, S9–S18. https://doi.org/https://dx.doi.org/10.3109/14992027.2016.1157268

Leigh, J. R., Dettman, S. J., Dowell, R. C., & Briggs, R. J. (2013). Communication development in children who receive a cochlear implant by 12 months of age. *Otol Neurotol*, *34*(3), 443–450. https://doi.org/10.1097/MAO.0b013e3182814d2c

Liberati, A., Altman, D., Tetzlaff, J., Mulrow, C., Gøtzsche, P., Ioannidis, J., Clarke, M., Devereaux, P., Kleijnen, J., & Moher, D. (2009). The PRISMA statement for reporting systematic reviews and meta-analyses of studies that evaluate healthcare interventions: explanation and elaboration. *BMJ (Clinical Research Ed.)*. https://doi.org/10.1136/bmj.b2700

Loundon, N., Blanchard, M., Roger, G., Denoyelle, F., & Garabedian, E. N. (2010). Medical and surgical complications in pediatric cochlear implantation. *Arch Otolaryngol Head Neck*

Surg, *136*(1), 12–15. https://doi.org/ 10.1001/archoto.2009.187

Marnane, V., & Ching, T. Y. C. (2015). Hearing aid and cochlear implant use in children with hearing loss at three years of age: Predictors of use and predictors of changes in use. *Int J Audiol*, *54*(8), 544–551. https://doi.org/10.3109/14992027.2015.1017660

McGowan, J., Sampson, M., & Lefebvre, C. (2010). An Evidence Based Checklist for the Peer Review of Electronic Search Strategies (PRESS EBC). *Evid Based Libr Inf Pract*, *5*(1), 149. https://doi.org/10.18438/B8SG8R

McGowan, J., Sampson, M., Salzwedel, D. M., Cogo, E., Foerster, V., & Lefebvre, C. (2016). PRESS Peer Review of Electronic Search Strategies: 2015 guideline statement. *Clin Epidemiol*, *75*, 40–46. https://doi.org/10.1016/j.jclinepi.2016.01.021

Michael, R., Attias, J., & Raveh, E. (2019). Cochlear implantation and social-emotional functioning of children with hearing loss. *J Deaf Stud Deaf Educ*, *24*(1), 25–31. https://doi.org/https://dx.doi.org/10.1093/deafed/eny034

Moher, D., Liberati, A., Tetzlaff, J., Altman, D. G., & The PRISMA Group. (2009). Preferred reporting items for systematic reviews and meta-analyses: The PRISMA statement. *PLoS Med*, *6*(7), e1000097. https://doi.org/10.1016/j.jclinepi.2009.06.005

Moher, D., Pham, B., Klassen, T. P., Schulz, K. F., Berlin, J. A., Jadad, A. R., & Liberati, A. (2000). What contributions do languages other than English make on the results of meta-analyses? *J Clin Epidemiol*, *53*(9), 964–972. https://doi.org/10.1016/S0895-4356(00)00188-8

Morrison, A., Moulton, K., Clark, M., Polisena, J., Fiander, M., Mierzwinski-Urban, M., Mensinkai, S., Clifford, T., & Hutton, B. (2009). English-Language restriction when conducting systematic review-based meta-analyses: Systematic review of published studies.

In *Canadian Agency for Drugs and Technologies in Health*. Canadian Agency for Drugs and Technologies in Health.

Moteki, H., Nishio, S., Miyagawa, M., Tsukada, K., Iwasaki, S., & Usami, S. (2017). Long-term results of hearing preservation cochlear implant surgery in patients with residual low frequency hearing. *Acta Oto-Laryngologica, 137*(5), 516–521. https://doi.org/10.1080/00016489.2016.1252061

Nilsson, M., Soli, S. D., & Sullivan, J. A. (1994). Development of the Hearing In Noise Test (HINT) for the measurement of speech reception thresholds in quiet and in noise. *J Acoust Soc Am, 95*(2),1085-1099. https://doi.org/10.1121/1.408469

Özdemir, S., Tuncer, Ü., Tarkan, Ö., Kiroĝlu, M., Çetik, F., & Akar, F. (2013). Factors contributing to limited or non-use in the cochlear implant systems in children: 11 years experience. *Int J Pediatr Otorhinolaryngol, 77*(3), 407–409. https://doi.org/10.1016/j.ijporl.2012.11.041

Page, M. J., Shamseer, L., & Tricco, A. C. (2018). Registration of systematic reviews in PROSPERO: 30,000 records and counting. *Syst Rev*, *32*(7). https://doi.org/10.1186/s13643-018-0699-4

Peterson, G. E., & Lehiste, I. (1962). Revised CNC lists for auditory tests. *J Speech Lang Hear Res, 27,* 62-70. https://doi.org/10.1044/jshd.2701.62

Rezaei, M., Emadi, M., Zamani, P., Farahani, F., & Lotfi, G. (2017). Speech intelligibility in Persian hearing impaired children with cochlear implants and hearing aids. *J Audiol Otol, 21*(1), 57–60. https://doi.org/https://dx.doi.org/10.7874/jao.2017.21.1.57

Sadadcharam, M., Warner, L., Henderson, L., Brown, N., & Bruce, I. A. (2016). Unilateral cochlear implantation in children with a potentially useable contralateral ear. *Cochlear*

Implants Int, 17 Suppl 1, 55–58. https://doi.org/https://dx.doi.org/10.1080/14670100.2016.1155832

Sampson, M., McGowan, J., Cogo, E., Grimshaw, J., Moher, D., & Lefebvre, C. (2009). An evidence-based practice guideline for the peer review of electronic search strategies. *Clin Epidemiol*, *62*(9), 944–952. https://doi.org/10.1016/j.jclinepi.2008.10.012

Schiavo, J. H. (2019). PROSPERO: An international register of systematic review protocols. *Med Ref Serv Q*, 171–180. https://doi.org/10.1080/02763869.2019.1588072

Skarzynski, H., Lorens, A., Dziendziel, B., & Skarzynski, P. H. (2015). Expanding pediatric cochlear implant candidacy: A case study of Electro-Natural Stimulation (ENS) in partial deafness treatment. *Int J Pediatr Otorhinolaryngol*, *79*(11), 1896–1900. https://doi.org/10.1016/j.ijporl.2015.08.040

Snels, C., Inthout, J., Mylanus, E., Huinck, W., & Dhooge, I. (2019). Hearing preservation in cochlear implant surgery: A meta-analysis. *Otol Neurotol*, *40*(2), 145–153. https://doi.org/10.1097/MAO.0000000000002083

Spahr, A. J., Dorman, M. F., Litvak, L. M., Cook, S. J., Loiselle, L. M., DeJong, M. D., Hedley-Williams, A., Sunderhaus, L. S., Hayes, C. A., & Gifford, R. H. (2014). Development and validation of the pediatric AzBio sentence lists. *Ear Hear*, *35*(4), 418–422. https://doi.org/https://dx.doi.org/10.1097/AUD.0000000000000031

Stöver, T., Issing, P., Graurock, G., Erfurt, P., ElBeltagy, Y., Paasche, G., & Lenarz, T. (2005). Evaluation of the advance off-stylet insertion technique and the cochlear insertion tool in temporal bones. *Otol Neurotol*, *26*(6), 1161–1170. https://doi.org/10.1097/01.mao.0000179527.17285.85

Thomas, B. H., Ciliska, D., Dobbins, M., & Micucci, S. (2004). A process for systematically

reviewing the literature: Providing the research evidence for public health nursing interventions. *Worldviews Evid Based Nurs*, *1*(3), 176–184. https://doi.org/10.1111/j.1524-475X.2004.04006.x

Thoutenhoofd, E. (2006). Cochlear implanted pupils in Scottish schools: 4-year school attainment data (2000-2004). *J Deaf Studies Deaf Educ*, *11*(2), 171–188. https://doi.org/10.1093/deafed/enj029

Wilson, K., Ambler, M., Hanvey, K., Jenkins, M., Jiang, D., Maggs, J., & Tzifa, K. (2016). Cochlear implant assessment and candidacy for children with partial hearing. *Cochlear Implants Int*, *17 Suppl 1*, 66–69. https://doi.org/https://dx.doi.org/10.1080/14670100.2016.1152014

Zanetti, D., Nassif, N., & Redaelli De Zinis, L. O. (2015). Factors affecting residual hearing preservation in cochlear implantation. *Acta Otorhinolaryngologica Italica*, *35*(6), 433–441. https://doi.org/10.14639/0392-100X-619

Zwolan, T. A., & Sorkin, D. L. (2017). Expanded cochlear implant candidacy. *ASHA Lead*, *22*(3), 14–15. https://doi.org/10.1044/leader.AEA.22032017.14

Chapter 4: Interviews with parents

Cochlear implant decision-making for children with residual hearing: Perspectives of parents

Abstract

Cochlear implant (CI) decision-making is particularly challenging for families of children who have residual hearing. Parents of these children may be uncertain about whether the potential benefits outweigh the risks related to CI. However, to our knowledge, there are very few studies that have reported how parents experience the CI decision-making process for this population. This study aimed to understand parents' decisional needs during the CI decision-making process for children with residual hearing.

Semi-structured interviews were conducted with parents of 11 children who had received CI(s). Open-ended questions were asked to encourage parents to share their experiences about the process of CI decision-making, their need for information, clarification of values/preferences, and decision support. The interviews were transcribed verbatim and analyzed using thematic analysis.

Three key themes emerged from the data: 1) Parents' decisional conflict, 2) values and preferences, and 3) decision support and parents' needs. We found that overall, parents were satisfied with their decision-making process and the decision support from practitioners. However, parents stressed the importance of receiving more personalized information that took into account their specific concerns, values, and preferences related to their child's and family's circumstances.

Our research provides additional evidence to guide the CI decision-making process for these children. Additional collaborative research with audiology and decision-making experts specifically on facilitating SDM is needed to provide better decision coaching for parents of these specific children with residual hearing.

Introduction

Parents of children with hearing loss are faced with many difficult decisions almost immediately after diagnosis. However, most parents require time to fully appraise the situation faced by their children and make decisions (Duncan, 2009; Mitchell & Karchmer, 2004). These decisions are mainly about children's communication approaches (i.e., oral communication, sign language, or total communication) and hearing technologies (i.e., hearing aid [HA] or cochlear implant [CI]) (Li et al., 2003; Duncan, 2009).

Although CI is considered the standard of care for children with bilateral severe to profound hearing loss who receive little benefit from HAs (Moog, 2002; Osberger et al., 2002; Raine, 2013; Skarzynski et al., 2006), some parents feel CI decision-making is one of the most difficult and stressful decision for their children with hearing loss (Duncan, 2009; Incesulu et al., 2003). More than 90% of children with hearing loss have parents with typical hearing (Mitchell & Karchmer, 2004). These parents often make a life-long decision for their children with a sense of urgency about their children's language development and consequences (Duncan, 2009; Hyde et al., 2010).

Decision-making regarding CI for children with residual hearing can be more challenging than for children with bilateral profound hearing loss because parents experience more uncertainty about a CI if their children have usable hearing and benefit from HAs (Fitzpatrick et al., 2009; Hyde et al., 2010). Unlike children with profound hearing loss, children with residual hearing are at risk of losing hearing, which complicates the decision for parents (Hyde et al., 2010; Duncan, 2009; Johnston et al., 2008; Zanetti et al., 2015). Although preserving residual hearing may be possible with recent developments in surgical techniques and device technologies (Eshraghi et al., 2017; Skarzynski, 2012; Sweeney et al., 2016), loss of residual

hearing remains an important risk following CI surgery (Moteki et al., 2017; Zanetti et al., 2015). In addition, like any typical CI surgery, other risks and negative consequences could occur after CI surgery (Duncan, 2009; Zanetti et al., 2015), including surgical complications, negative outcomes, and device-related complications, which may impact the children's future quality of life (Alegre-de la Rosa & Villar-Angulo, 2020; Chute & Nevins, 2002; Ganek et al., 2020). For these reasons, parents and practitioners may be uncertain as to whether the potential benefits outweigh the risks related to CI (Greaver et al., 2017; Hardonk et al., 2010; Li et al., 2004; Steinberg et al., 2000).

To assist CI decision-making for these families, comprehensive evidence about the CI decision-making process is important. During the CI decision-making process for children more generally, a CI team plays an important role in providing information and supporting families (Fitzpatrick et al., 2009; Hyde et al., 2010; Li et al., 2003; Roberts et al., 2015). Several studies have shown that information specifically related to the benefits and risks of CIs strongly impacted parents' CI decisions (Roberts et al., 2015; Hyde et al., 2010). Families of children who have already received CI are also considered an important information resource to parents who are in the CI decision-making process (Hyde et al., 2010; Johnston et al., 2008). However, to our knowledge, there are very few studies that have reported how parents experience the CI decision-making process for this population (Fitzpatrick et al., 2009; Porter et al., 2018).

Another important aspect of CI decision-making for these families is understanding their values and preferences (Kluwin & Stewart, 2000; Li et al., 2003, 2004). Shared decision making (SDM) is one way to arrive at informed and value-based clinical decisions regarding intervention plans with mutual understanding and agreement between patients and practitioners (Barry & Edgman-Levitan, 2012; Edwards & Elwyn, 2009). Since SDM considers personal values and

preferences, patients themselves can consider the benefits and risks based on their values and preferences (Charles et al., 1997). Applying SDM in healthcare provides the best information on each of the available options, based on the values and preferences of patients. However, preferences and values for decision-making involvement can vary among families and across decisions (Coyne et al., 2016; Lipstein et al., 2014). In addition, as several studies have reported, the preference of parents for information and their feelings about the sufficiency of information differ among families and across decisions (Harrison, 2004; Robertson et al., 2018).

Several studies have reported that it is important for parents and practitioners to work together in decision-making, which may improve decision satisfaction and reduce the risk of decisional conflict and regret (Mack et al., 2016; Mckenna et al., 2010; Sisk et al., 2019). Despite this, almost no information exists that focuses on either decision needs or decision involvement about CI decision-making for these children with residual hearing. As the first step to facilitating CI decision-making for these children, we captured parents' perspectives of their decisional support needs.

Methods

Study design

A qualitative research study design using interpretive description methodology (Thorne et al., 2004) was employed to understand the perspectives of parents during the CI decision-making process for their children with residual hearing. Semi-structured individual interviews were conducted with parents of 11 children. For one child both parents were interviewed separately. Parents were asked open-ended questions to obtain in-depth information about their experiences, values, preferences, and decisional needs.

Participants

Participants were recruited between March 2019 and August 2019. Parents of children receiving rehabilitation services were invited to participate in the study through the Audiology clinic at CHEO, a major Canadian pediatric audiology center, which serves a population of approximately one million people. The CI program is housed within the Audiology clinic and all children are implanted in a publicly funded health care setting. Parents were eligible to participate if their child had a CI in at least one ear, and they: a) had a chronological age ≤ 18 years at the time of CI surgery, b) had a preoperative degree of hearing loss better than profound (pure-tone average [PTA] ≤ 90 dB HL at 500, 1000, 2000 Hz) in at least one ear (either the CI ear or the non-CI ear, including children with single-sided deafness), and c) continued to be followed for regular CI audiological services. All interviews were conducted in English. For parents who had difficulty in English, one of parents who was more comfortable communicating in English participated in the interviews. A recruitment poster was placed on the posting board of the Audiology Clinic at CHEO. Eligible parents were informed about the study by CI audiologists and auditory-verbal therapists (AVTs) at the Audiology clinic at CHEO and signed a consent-to-contact form. For the interviews, the main researcher then directly contacted the parents who had indicated interest and arranged a time and place for the interview.

Ethical approval for this study was provided by the Children's Hospital of Eastern Ontario (CHEO)'s Research Ethics Board and the University of Ottawa Ethics Board (Appendix E). Parents signed an informed consent form prior to the interviews.

Procedure

An interview guide was developed (see Appendix F), guided by the Decisional Needs Assessment in Populations (Jacobsen et al., 2013), which is a workbook for assessing patients' and practitioners' decision-making needs based on the Ottawa Decision Support Framework (ODSF) (O'Connor et al., 1998). The ODSF was developed for health decisions such as a new diagnosis or new intervention that requires careful deliberation due to uncertainty in benefits and risks and that are of a value-sensitive nature (O'Connor et al., 1998). The interview guide was reviewed by two senior researchers who adapted the questions that were included for the appropriate subject content of the study. Individual interviews were conducted by the main researcher, who was unknown to the families. Parents had the choice to participate in in-person or phone interviews, depending on their preferences. The interviewer asked questions about the parents' experiences throughout the decision-making process, their values and preferences, and their needs for information and support. Consistent with qualitative methods, the interviewers also formulated subsequent questions in response to the answers. Field notes were taken during the interviews by the interviewer. Interviews were audio-recorded to facilitate subsequent detailed analysis and then were transcribed verbatim by two trained researchers, and the researcher who interviewed the parents verified the transcription, supplemented by field notes.

Data analysis

Conducting and transcribing interviews proceeded concurrently, since preliminary analysis can guide other data collection (DiCicco-Bloom & Crabtree, 2006). The interview data were coded and analyzed using NVivo (QSR International Pty Ltd. Version 12.0), a software program used for qualitative studies. A thematic analysis approach was applied to analyze the interview data.

The coding process involved a constant comparative method based on open, axial, and selective coding methods (Corbin & Strauss, 2008). The main researcher created initial codes, similar codes were grouped into categories, and then these categories were collapsed into themes. Themes and sub-themes were identified by comparing code frequencies, relationships, and similarities between codes. A trained researcher verified the codes, and if any of the coding was not clear, this was discussed between the two researchers and resolved.

Results

Participant characteristics

Parents of 11 children who had received CIs participated in the interviews. Interviews were conducted in person for five parents and by telephone for the remaining six. A description of the characteristics of the children is summarized in Table 1. Nine of the 11 children underwent newborn hearing screening and two were born in countries without screening. The children had a median age of confirmation of hearing loss of 20.5 months (interquartile range [IQR] 2.1, 47.5 months), and all had bilateral hearing loss at diagnosis except for one with single-sided deafness. The children received their CIs at a median age of 46.2 months (IQR 29.3, 94.0 months).

Table 1. Clinical characteristics of 11 CI children with residual hearing

Characteristics	Participants (n=11)	
Sex, n (%)		
Female	8	(72.7)
Route to identification, n (%)		
Screened	9	(81.8)
Non-screened	2	(18.2)
Onset of hearing loss, n (%)		
Congenital	5	(45.4)
Late onset	4	(36.4)
Unknown [a]	2	(18.2)
Etiology, n (%)		
Unknown	4	(36.4)
Known		
ENT malformation [b]	1	(9.0)
Familial/genetic	4	(36.4)
Syndromic	2	(18.2)
Age at diagnosis (months), median (IQR)	20.5	(2.1, 47.5)
Age at hearing aid fitting (months), median (IQR)	20.9	(3.8, 44.3)
Age at CI surgery (months), median (IQR)	43.1	(23.9, 57.7)
Age at interview (months), median (IQR)	49.4	(35.8, 56.1)
Degree of hearing loss at diagnosis (better ear), n (%)		
Within normal range (single-sided deafness)	1	(9.1)
Mild	1	(9.1)
Moderate	1	(9.1)
Moderate-severe	1	(9.1)
Severe	7	(63.6)
Degree of hearing loss at surgery (better ear), n (%)		
Moderate-severe	2	(18.2)
Severe	9	(81.8)
Type of hearing technology		
Unilateral CI		
CI in one ear, no HA use	1	(9.1)
Bimodal stimulation (CI and HA), HA in contralateral ear	4	(36.4)
Bilateral CIs		
Sequential [c]	1	(9.1)
Simultaneous [d]	5	(45.4)

N, number; ENT, ear nose throat; IQR, interquartile range; CI, cochlear implant; HA, hearing aid

[a], two children from other countries with limited diagnostic information

[b], cochlear anomalies, enlarged vestibular aqueduct syndrome (EVAS), and mondini malformation

[c], each ear was implanted in two separate surgeries (over 6 months apart)

[d], both ears were implanted at the same time or less than 6 months apart

Findings from parent interviews

The findings were categorized into three key themes that are aligned with major components of the ODSF. These themes and sub-themes are described in Table 2. The following section elaborates on each theme with examples of parents' comments. Numbers in parentheses refer to the study participant numbers.

Table 2. Themes and sub-themes from the interview data

Themes	Sub-themes
Decisional conflict	Parents' response to CI recommendation
	Parents' concerns about CI
	Considerations affecting CI decision-making
	Deciding factors leading to CI
Parents' values and preferences	Inclusion into hearing society
	Preservation of residual hearing
Decision support and parents' needs	Information and support
	Personalized information
	Decision support tools

CI, cochlear implant

Decisional conflict

Parents' response to CI recommendation. Based on the interviews, some parents reported that they had already considered a CI as an option when their practitioner first recommended consideration of a CI, because their children showed limited progress in auditory skills and communication.

> *We would expect some similarities between her and her sister. Her sister started talking at nine months … and then at 13 months, that is when it really took off and when we weren't seeing the same sort of development with her or anything close to her sister ...*

Then, I think we were pretty sure about the cochlear implant...we got used to the idea that our kid is going to have it. (Parent_06)

However, some other parents believed that their children were functioning well with HAs and that a CI was not necessary.

... They [CI team] told us, now, we will plan for the CI team ... We were still depending on her right ear. It was not a big hurry so we thought that we needed some time to make the decision … Let's see how it goes if she is picking up something on her right ear or not. (Parent_11)

Parents' concerns about CI. When a CI was recommended, parents who participated in this interview reported that the major reason they delayed the CI decision was the fear of their child losing residual hearing, and these concerns continued during the CI decision-making process. As shown in Table 3, parents also talked about the fear of surgery and negative consequences, such as device failure. Parents described uncertainty about how much additional communication benefit a CI could offer and whether the benefits for speech and language outweighed the risks related to surgery. For example, parents of three children recalled how they regretted their choice during the surgery.

The only doubt that I had was on the day of surgery. It [the surgery] was for 12 hours. I started to question myself. What am I doing to my son? Oh my gosh! What am I putting him through? And that was when I really, I had some doubts about the first time ... (Parent_05)

Additionally, a few parents talked about the long-term reliability of CI as their main preoccupation. They were convinced that their children would benefit from CI but were

concerned about the long-term benefits and whether they would experience disadvantages (e.g., restricted access to magnetic resonance imaging [MRI]) of CI during their lifetime.

Table 3. Parents' concerns and sample quotes

Parents' concerns	Sample quotes
Outcomes after CI	*It [CI decision-making] was hard because I saw some children, they improved after the surgery and also, I saw a few children that had no improvement at all after the surgery. Even after a couple of years they still can't understand, can't speak. So yes, I saw a lot of different results. Because of this, it was a hard decision. (Parent_01)*
Anxiety about surgery	*Because it [CI surgery] was a very 'looong' surgery so I think that was the biggest worry. There is still, like, on the decision I wasn't worried about making the wrong decision for her… I was more worried about the [surgery] procedure itself. (Parent_03)*
Anxiety about loss of residual hearing	*I was more worried that she actually might lose the hearing in her good ear. That's the only thing I was really concerned about that she actually might lose ... That's the most concerning for me is that there is that risk that she can lose her hearing ... (Parent_04)*
Long-term reliability of CI	*Frankly speaking, the only worry we have it's like for lifetime and the working of this CI, processor. How long it's going to work, it should not cause any inflammation in her brain, it should not cause her discomfort, and in the future if she gets an MRI or something how are we just going to remove it and put it back. These are the worries, which are like long-term worries but they are in [the] back of our mind. (Parent_11)*

CI, cochlear implant; MRI, Magnetic resonance imaging

Considerations affecting CI decision-making. Some parents described how they experienced pressure as they made decisions about CI for their young children because this decision is a life-long decision.

> *It was terrible, because he should live his, all his life because of my decision. And because of my decision, he will wear all his life two cochlear implants. Yes, it was terrible. I hope I made the right decision. For me I can decide, it's my decision. I can live*

with my decision, yes. But in this situation, he should live all his life, he should wear all his life two cochlear implants because of my decision … (Parent_01)

Additionally, two parents noted that they felt CI decision-making was complicated for them because their children had medical complexities or additional disabilities beyond hearing loss. For these children, since they still benefitted a little from HAs, parents said that they feared the possibilities of additional health issues for their children by choosing a CI, which could add ongoing risks because they had a CI.

My child is Down syndrome, so she has very difficult to learn language so we work a lot with sign, with images a lot ... We push to make her talk more, to make her sign more because we didn't know what happened ... The audiologist just told us that she could be a potential cochlear implant candidate ... She could hear a little, and I didn't want to play in. If it didn't work, it could be worse ... (Parent_10)

Deciding factors leading to CI. As shown in Table 4, there were three major factors culminating in the CI decision for these children from the perspectives of parents. Over half of the parents talked about the deterioration of their children's hearing and limited social integration as two major factors that led them to move forward with a CI. For other parents, their realization of their children's limited speech and auditory-related functions made it clear that a CI was required to continue progressing.

Table 4. Deciding factors affecting decision-making and sample quotes

Deciding factors leading to CI decision	Sample quotes
Deterioration of hearing	*She has had a right ear that was working well and the doctor had told us that usually kids if they have one working ear well, they pick up the speech, but if it keeps on working and if the hearing decreases or something then we cannot say anything. But in our case, unfortunately, the same thing happened that we just had to make that decision as soon as possible. (Parent_11)*
Limited social integration	*I would use the advances that technology offers in order to have the fullest experience possible and that's the same way that we looked at the decision to get her CI. Well, we wanted her to make sure that she would be able to integrate into the social realm where we live. (Parent_06)*
Limited speech and auditory function	*When I say some words, she always precedes another word, like, I say 'banana', she would say it 'janana'. If you say 'swimming' she would say 'raining'. She could not specifically identify them, especially, for the lower, the silence, and the quiet letters. So that she did not grow up with that and not able to speak properly, not able to pronounce her words correctly [with her HAs], so I decided to go with the CI. (Parent_12)*

CI, cochlear implant; HA, hearing aid

Parents' values and preferences

Inclusion into hearing society. Parents expressed their hopes for better hearing, speech, language, and social functioning through CI use because they wanted their children to be well-integrated into the hearing culture that is familiar to family and friends.

> *Any parent in the world, it's, they understand that our kids need to hear for his or her lifetime and hearing is a thing which is helpful for speech and we have to give our kid this thing so that she can live a good and normal standard life ... Our kid needs to listen, needs to talk, because this is a complicated world, and he or she needs to hear for the rest of their lifetime. (Parent_11)*

In particular, parents of children who previously showed speech and language development expected that a CI could restore children's previous hearing and language abilities. Some parents

expected that with a CI their children could go back to a 'normal life'.

> *The probability of her having a normal life or a more than normal life was increased with the CI. Right? ... We expect that it will get her closer to a normal type of life. (Parent_08)*

Parents talked about maximizing their children's overall development. About half of these parents described that they hoped a CI would help their children to avoid academic, social, and employment-related restrictions in their future life.

> *If she were to have been non-hearing with sign language, it would restrict certain opportunities for education and employment and other stuff. So, we are still of the attitude that we will teach her sign but that's more of a backup plan. Right? You know, we expect that she is going to learn multiple languages. (Parent_08)*

Preservation of residual hearing. Parents noted that preserving residual hearing was particularly important. Knowledge about new CI technology and surgical techniques to preserve residual hearing facilitated their decision-making and seemed to offer some reassurance. Some parents said that they sought out information about new CI technology and tended to choose manufacturers that emphasized hearing preservation technologies.

> *It was really important to me that she still keep her natural sound as much as she can. I wanted a CI could give her the opportunity to be able to hear more than what she would hear previously to maintain the spoken language at home … We went with [CI company] because we were looking for hybrid CI which is what we got on her right side because she had some residual hearing. (Parent_07)*

Decision support and parents' needs

Information and support. Parents noted they were satisfied with the decision-making process because they had sufficient information and support from CI practitioners. In describing the support that they received from their CI team (i.e. audiologists, AVTs, CI surgeons, the itinerant teacher of the deaf and hard of hearing, speech therapists, psychologists), they often referred to the kind of information that was useful, such as information about the CI surgery process, benefits and risks following CI surgery, and rehabilitation plans. In particular, audiologists, CI surgeons, and AVTs were major information resources and therefore major sources of support throughout the process. Parents described the process as ongoing and talked about meeting frequently with clinicians, sometimes almost from the time of diagnosis. Parents said that this ongoing access to professional support made it easy for them to ask questions and receive information throughout the CI decision-making process.

> *Mostly [AVT name] because we spend most of our time with her. In our case [AVT name] reassured us the most because we see her the most, once a week. She's really sweet, and we have a good relationship with her. She always reassured us. She showed us so many of her patients and their progress … she's really nice to us and we like her a lot ... It's like an open door of communication ... (Parent_02)*

Parents also frequently stressed that other families of children who had received CI were an important resource for them. Parents could meet them through Internet groups and the meetings were facilitated by clinicians. They highlighted that these families provided real-life information to them.

> *Speaking with other families was very helpful for us. We spoke with other families who were in the same situation as us. Actually, speaking with other families is very, very, very*

important. They actually helped us a lot, when it came to deciding where to go to seek support. They were the biggest factor and biggest support for us. (Parent_09)

According to the parents, the Internet was also an important information resource. Parents described the information and support resources that they accessed during the decision-making process, and for them, information and support went hand in hand.

We did a lot of research through the Internet before we made this decision. We were mindset that we had to collect as much information as we possibly could make the determination. I think we were pretty satisfied that finding some actual scientific information. (Parent_08)

Personalized information. Parents enumerated suggestions they believed could further enhance the decision-making process. One recommendation from some parents was for personalized information about the child's everyday functioning with a CI beyond the clinical information they received. For example, some parents felt it would be helpful to have more technical information about the noise attenuation of different devices and to conduct measures more customized to their specific values and preferences.

We just want to make sure that she is getting the best use of her equipment. So as time goes on and she ends up in more complex environments, it is going to be worth creating different maps in her CI and her hearing aid, to facilitate her best access to sound. She is entering school in September and ... what is going to reap the best benefits for her by using her technology to its fullest potential? … If the child is in a sports environment, say swimming, this is what we think you should consider. If the child is in a sports

environment, in an outside environment, maybe these are different considerations, right? (Parent_08)

Decision support tools. Some parents noted that they needed decision support tools to facilitate CI decision-making for them and their children. They suggested that these tools would be helpful if they included statistical information about the probabilities of the benefits and risks of CIs compared to HAs as well as specific perspectives and experiences including the everyday functioning of children with residual hearing and their families' experiences. In addition to tools, they noted that peer support programs could be highly valuable to families.

I think that it varies for each individual. I mean everyone makes their decision-making process differently. But tool-wise, if they gave you just a list of, like, the possible side effects and stuff and then versus the benefits that might make it a little easier because that's what I did with the pro and con list ... I think that the tool should definitely talk to you about the resources that are available and like the benefits that are available ... (Parent_07)

They also suggested that a family-friendly tool about CI could be helpful. For example, they talked about a range of visual information such as video clips and decision aids.

Maybe a visual thing to see what it actually looks like because when I first heard about the option, I searched through the internet, but it was not very helpful for us. So, I think most people are visual and tactile, so it would be helpful to the decision to see how the CI works for our kids... (Parent_06)

Discussion

This study explored the CI decision-making process through interviews with parents of children with residual hearing. Three key themes based on parents' perspectives were decisional conflict, values and preferences, and decision support and parents' needs.

When their practitioner first recommended consideration of a CI, some parents reported that they had already considered a CI as an intervention option because they felt their children showed limitations with HAs. These parents' responses to the recommendation were consistent with previous studies that reported parents' perspectives on decision-making for any child receiving a CI (Johnston et al., 2008; Hyde et al., 2010). Some other parents of children in our study tended to delay CI decision-making because they believed their children still received enough benefit from HAs to continue progressing in language development. This finding is similar to findings of previous studies that indicated that parents and clinicians may require more time to make a decision about CI for children with residual hearing than for children with bilateral profound hearing loss (Fitzpatrick et al., 2009; Hyde et al., 2010; Leigh et al., 2016).

Parents of children with hearing loss were concerned about losing residual hearing. Like parents making CI decisions for any child, parents of children with residual hearing also worried about the surgery and expressed uncertainty about whether the benefits of speech and language outweighed the risks related to surgery (Chang, 2017; Porter et al., 2018; Ravi & Gunjawate, 2020). They also talked about their concerns related to the long-term reliability of a CI and disadvantages of a CI (e.g., restrictions around MRI).

Parents in our study identified several considerations that affected their decision-making. In particular, parents sometimes felt pressure making decisions on behalf of their young children who were still receiving benefits from HAs. They talked about feeling conflicted about whether a

CI was the right decision and whether the child would blame them later. Although some research (Dettman et al., 2007, 2016; Holt & Svirsky, 2008) has concluded that implantation before 12 months of age is advantageous for language development, there is relatively little information to guide parents on the effectiveness of very early implantation specific to young children with residual hearing (Carlsson et al., 2011; Hoff et al., 2019). One of the reasons for the uncertainty around decision-making for young children is the difficulty in measuring speech perception abilities (Fitzpatrick et al., 2009; Leigh et al., 2016). Parents of children with residual hearing and additional disabilities also reported that other developmental issues were an important consideration for them. This is similar to the findings for children who are traditional candidates for CI and who have additional medical and developmental issues (Holcomb & Smeal, 2020; Oghalai et al., 2012; Wiley et al., 2012; Zaidman-Zait et al., 2015). In particular, Oghalai et al. (2012) reported that the timing of CI surgery can be delayed for children with additional disabilities due to their complex medical needs. In addition, Zaidman-Zait et al. (2015) noted that families of these children may also need more information and family support.

Parents identified several factors that led them to ultimately decide that a CI was the best option for their children. Hearing deterioration, limited auditory function, and limited social integration were the three major factors for these parents. Recent studies have also reported that the child's social-emotional well-being was an important deciding factor for parents (Chang, 2017; Porter et al., 2018). Our findings are also aligned with practitioners' perspectives on the factors they consider in CI decision-making for children with residual hearing (Fitzpatrick et al., 2009; Leigh et al., 2016). In particular, in these studies, practitioners highlighted the importance of evaluating overall auditory function, listening effort and fatigue, social behaviour, and progress in therapy. In our recent research with practitioners, they emphasized that these factors

continue to contribute to their recommendations for a CI for children with residual hearing (Na et al., submitted).

Central to decision-making for these parents was the value they placed on inclusion into their hearing community and society. These findings are consistent with previous studies on decision-making for more traditional CI pediatric candidates (Chang, 2017; Hardonk et al., 2010; Hyde et al., 2010; Johnston et al., 2008). In typical CI decisions, some parents also took into account Deaf culture or obtained information from the Deaf community (Hyde et al., 2010; Chang, 2017). However, according to Chang (2017), most parents considered CI because they hoped that their children would be part of their spoken language community after implantation. Spoken communication was a core value for the parents of children in our study, and some parents expressed high expectations that their children's hearing would be 'normal'.

Parents in our study reported that they were satisfied with the quality of their decision-making because they received sufficient information and support from the CI team. We found that they appreciated the team approach and viewed information and support as inseparable. Parents noted that other parents were also an important resource for them in the process. Similarly, in a recent systematic review that explored barriers and facilitators in the general CI decision-making process, Ravi & Gunjawate (2020) found that the CI team, parents of other implanted children, the Internet, and manufacturer materials were important resources.

Despite their positive perceptions of the information and support they received from various resources, parents still wanted more information that is personalized to their values and their children's specific needs. They specifically identified information about CI use related to children's everyday activities such as swimming, music, and language learning. Parents also hoped for better decision support intervention (e.g., video clips, peer support programs) to assist

them in accessing the information they need. Our findings that parents require evidence-based information that is personalized to their values and preferences is aligned with the SDM approach. Several studies in the decision-making sciences have shown that providing personalized information to patients contributes to higher quality decision-making and less decisional conflict (Downing et al., 2009; Drake et al., 2009).

Limitations

A limitation of our study is that all participants were recruited from a single region in Canada where children received services within a publicly funded health care setting and had access to a CI team located in one pediatric center. Parents from other types of health care settings may have different perspectives and experiences in reaching a CI decision for their children with residual hearing. Additionally, the CI program where this study was conducted focuses on the development of spoken language. Parents' perspectives may differ with different intervention models and cultural backgrounds. While our participants were from diverse cultures, we did not collect culture-specific information. Our interviews were limited to parents only, but inclusion of children's perspectives could add further insights in future studies.

Implications

This study is a part of a larger project that captured perspectives from both practitioners and parents. This study provided new insights into the perspectives of parents of children with residual hearing who underwent the decision-making process to receive a CI for their children. The findings have several important implications for practitioners who guide parents in making these decisions. Parents of children with residual hearing appreciated the information and

support from their CI team. It is recommended that these CI practitioners provide more evidence-based information about children's social functioning outcomes beyond hearing and language benefits, as parents highlighted their needs in this area. For these parents, their children's inclusion into the hearing culture with their family and friends was an important value and therefore is an important component of the counselling process. Parents would also appreciate more emphasis on personalized information tailored to their specific concerns, values, preferences and family circumstances during the pre-implant coaching period. Additionally, providing better decision support intervention may guide these parents of children with residual hearing to reduce the uncertainty in this decision-making process.

Conclusion

This study is one of the first to capture parents' experiences including their values, preferences, and decisional needs in the CI decision-making process for children with residual hearing. We found that parents were satisfied with the decision-making process overall and with the decision support from practitioners and other parents who provided information on practical aspects related to CI. However, parents stressed the importance of receiving more personalized information which takes into account their specific values and preferences related to their child and their family circumstances during the decision-making process. Our research provides additional evidence to guide the CI decision-making process for children with residual hearing. Additional collaborative research with audiology and decision-making experts specifically on facilitating SDM is needed to provide better decision coaching for parents of these specific children with residual hearing.

References

Alegre-de la Rosa, O. M., & Villar-Angulo, L. (2020). Health-related quality of life in children who use cochlear implants or hearing aids. *Heliyon*, *6*(1), e03114. https://doi.org/https://doi.org/10.1016/j.heliyon.2019.e03114

Barry, M. J., & Edgman-Levitan, S. (2012). Shared decision making — The pinnacle patient-centered care. *New Eng J Med*, *366*(9), 780-781. https://doi.org/10.1056/NEJMp1109283

Carlsson, P. I., Hall, M., Lind, K. J., & Danermark, B. (2011). Quality of life, psychosocial consequences, and audiological rehabilitation after sudden sensorineural hearing loss. *Int J Audiol*, *50*(2), 139–144. https://doi.org/10.3109/14992027.2010.533705

Chang, P. F. (2017). Patient education and counseling breaking the sound barrier: Exploring parents' decision-making process of cochlear implants for their children. *Patient Educ Couns*, *100*(8), 1544–1551. https://doi.org/10.1016/j.pec.2017.03.005

Charles, C., Gafni, A., & Whelan, T. (1997). Shared decision-making in the medical encounter: What does it mean? (or it takes at least two to tango). *Soc Sci Med*, *44*(5), 681–692.

Chute, P. M., & Nevins, M. E. (2002). *The parents 'guide to cochlear implants*. Gallaudet University Press.

Corbin, J., & Strauss, A. (2008). *Basics of qualitative research: Techniques and procedures for developing grounded theory*. Thousand Oaks, CA: Sage publications.

Coyne, I., O'Mathúna, D. P., Gibson, F., Shields, L., Leclercq, E., & Sheaf, G. (2016). Interventions for promoting participation in shared decision-making for children with cancer. *Cochrane Database Syst Rev*, *11*. https://doi.org/10.1002/14651858.CD008970.pub3

Dettman, S. J., Dowell, R. C., Choo, D., Arnott, W., Abrahams, Y., Davis, A., Dornan, D.,

Leigh, J., Constantinescu, G., Cowan, R., & Briggs, R. J. (2016). Long-term communication outcomes for children receiving cochlear implants younger than 12 months: A multicenter study. *Otol Neurotol*, *37*(2), e82-95. https://doi.org/10.1097/MAO.0000000000000915

Dettman, S., Pinder, D., Briggs, R., Dowell, R., & Leigh, J. (2007). Communication development in children who receive the cochlear implant younger than 12 Months: Risks versus benefits. *Ear Hear*, *28*(2), 11S-18S. https://doi.org/10.1097/AUD.0b013e31803153f8

DiCicco-Bloom, B., & Crabtree, B. F. (2006). The qualitative research interviews. *Med Educ*, *40*(4), 314–321. https://doi.org/10.1111/j.1365-2929.2006.02418.x

Downing, G. J., Boyle, S. N., Brinner, K. M., & Osheroff, J. A. (2009). Information management to enable personalized medicine: Stakeholder roles in building clinical decision support. *BMC Med Inform Decis Mak*, *9*, 44. https://doi.org/10.1186/1472-6947-9-44

Drake, R. E., Cimpean, D., & Torrey, W. C. (2009). Shared decision making in mental health: Prospects for personalized medicine. *Dialogues Clin Neurosci*, *11*(4), 455–463. https://pubmed.ncbi.nlm.nih.gov/20135903

Duncan, J. (2009). Parental readiness for cochlear implant decision-making. *Cochlear Implants Int*, *10*(1), 38–42. https://doi.org/10.1002/cii.384

Edwards, A., & Elwyn, G. (2009). Shared decision-making in health care: Achieving evidence-based patient choice. In A. Edwards & G. Elwyn (Eds.), *Oxford University Press* (illustrate). OUP Oxford, 2009. https://doi.org/10.1007/s13398-014-0173-7.2

Eshraghi, A. A., Ahmed, J., Krysiak, E., Ila, K., Ashman, P., Telischi, F. F., Angeli, S. A., Prentiss, S., Martinez, D., & Valendia, S. (2017). Clinical, surgical, and electrical factors impacting residual hearing in cochlear implant surgery. *Acta Oto-Laryngologica*, *137*(4),

384–388. https://doi.org/10.1080/00016489.2016.1256499

Fitzpatrick, E. M., Olds, J., Durieux-Smith, A., McCrae, R., Schramm, D., & Gaboury, I. (2009). Pediatric cochlear implantation: How much hearing is too much? *Int J Audiol*, *48*(2), 91–97. https://doi.org/10.1080/14992020802516541

Ganek, H. V., Feness, M. L., Goulding, G., Liberman, G. M., Steel, M. M., Ruderman, L. A., Papsin, B. C., Cushing, S. L., & Gordon, K. A. (2020). A survey of pediatric cochlear implant recipients as young adults. *Int J Pediatr Otorhinolaryngol*, *132*, 109902. https://doi.org/10.1016/j.ijporl.2020.109902

Greaver, L., Eskridge, H., & Teagle, H. F. B. (2017). Considerations for pediatric cochlear implant recipients with unilateral or asymmetric hearing loss: Assessment, device fitting, and habilitation. *J Am Acad Audiol*, *26*(2), 91–98. https://doi.org/10.1044/2016_AJA-16-0051

Hardonk, S., Bosteels, S., Desnerck, G., Loots, G., Van Hove, G., Van Kerschaver, E., Vanroelen, C., & Louckx, F. (2010). Pediatric cochlear implantation: A qualitative study of parental decision-making processes in Flanders, Belgium. *Am Ann Deaf*, *155*(3), 339–352. https://doi.org/10.1353/aad.2010.0012

Harrison, C. (2004). Treatment decisions regarding infants, children and adolescents. *J Paediatr Child Health*, *9*(2), 99–103. https://doi.org/10.1093/pch/9.2.99

Hoff, S., Ryan, M., Thomas, D., Tournis, E., Kenny, H., Hajduk, J., & Young, N. M. (2019). Safety and effectiveness of cochlear implantation of young children, including those with complicating conditions. *Otol Neurotol*, *40*(4), 454–463. https://doi.org/10.1097/MAO.0000000000002156

Holcomb, M., & Smeal, M. (2020). Pediatric cochlear implantation: Who is a candidate in 2020?

Hear J, *73*(7), 8–9. https://doi.org/http://bit.ly/HJcurrent

Holt, R. F., & Svirsky, M. A. (2008). An exploratory look at pediatric cochlear implantation: Is earliest always best? *Ear Hear*, *29*, 492–511. https://doi.org/10.1097/AUD.0b013e31816c409f

Hyde, M., Punch, R., & Komesaroff, L. (2010). Coming to a decision about cochlear implantation: Parents making choices for their deaf children. *J Deaf Stud Deaf Educ*, *15*(2), 162–178. https://doi.org/10.1093/deafed/enq004

Incesulu, A., Vural, M., & Erkam, U. (2003). Children with cochlear implants: Parental perspective. *Otol Neurotol*, *24*, 605–611. https://doi.org/10.1097/00129492-200307000-00013

Jacobsen, M. J., O'Connor, A., & Stacey, D. (2013). Decisional Needs Assessment in Populations: A workbook for assessing patients 'and practitioners 'decision-making needs. *Decisional Needs Assessment in Populations*.

Johnston, J. C., Durieux-Smith, A., Fitzpatrick, E., O'Connor, A., Benzies, K., & Douglas, A. (2008). An assessment of parents 'decision-making regarding paediatric cochlear implants. *Can J Speech-Lang Pathol Audiol*, *32*(4), 169–182.

Kluwin, T. N., & Stewart, D. A. (2000). Cochlear implants for younger children: A preliminary description of the parental decision process and outcomes. *Am Ann Deaf*, *145*(1), 26–32. https://doi.org/10.1353/aad.2012.0247

Leigh, J. R., Dettman, S. J., & Dowell, R. C. (2016). Evidence-based guidelines for recommending cochlear implantation for young children: Audiological criteria and optimizing age at implantation. *Int J Audiol*, *55 Suppl 2*, S9–S18. https://doi.org/https://dx.doi.org/10.3109/14992027.2016.1157268

Li, Y., Bain, L., & Steinberg, A. G. (2003). Parental decision making and the choice of communication modality for the child who is deaf. *Arch Pediatr Adolesc Med*, *157*(2), 162–168. https://doi.org/10.1001/archpedi.157.2.162

Li, Y., Bain, L., & Steinberg, A. G. (2004). Parental decision-making in considering cochlear implant technology for a deaf child. *Int J Pediatr Otorhinolaryngol*, *68*(8), 1027–1038. https://doi.org/10.1016/j.ijporl.2004.03.010

Lipstein, E. A., Dodds, C. M., & Britto, M. T. (2014). Real life clinic visits do not match the ideals of shared decision making. *J Pediatr*, *165*(1), 178–183. https://doi.org/10.1016/j.jpeds.2014.03.042

Mack, J. W., Cronin, A. M., & Kang, T. I. (2016). Decisional regret among parents of children with cancer. *J Clin Oncol*, *34*(33), 4023–4029. https://doi.org/10.1200/JCO.2016.69.1634

Mckenna, K., Collier, J., Hewitt, M., & Blake, H. (2010). Parental involvement in paediatric cancer treatment decisions. *Eur J Cancer Care*, *19*(5), 621–630. https://doi.org/10.1111/j.1365-2354.2009.01116.x

Mitchell, R. E., & Karchmer, M. A. (2004). Chasing the mythical ten percent: Parental hearing status of deaf and hard of hearing students in the United States. *Sign Lang Stud*, *4*(2), 138–163. https://doi.org/10.1353/sls.2004.0005

Moog, J. S. (2002). Changing expectations for children with cochlear implants. *Ann Otol Rhinol Laryngol*, *111*(5 II), 138–142. https://doi.org/10.1177/00034894021110s527

Moteki, H., Nishio, S., Miyagawa, M., Tsukada, K., Iwasaki, S., & Usami, S. (2017). Long-term results of hearing preservation cochlear implant surgery in patients with residual low frequency hearing. *Acta Oto-Laryngologica*, *137*(5), 516–521. https://doi.org/10.1080/00016489.2016.1252061

O'Connor, A. M., Tugwell, P., Wells, G. A., Elmslie, T., Jolly, E., Hollingworth, G., McPherson, R., Bunn, H., Graham, I., & Drake, E. (1998). A decision aid for women considering hormone therapy after menopause: Decision support framework and evaluation. *Patient Educ Couns*, *33*(3), 267–279. https://doi.org/10.1016/S0738-3991(98)00026-3

Oghalai, J. S., Caudle, S. E., Bentley, B., Abaya, H., Lin, J., Baker, D., Emery, C., Bortfeld, H., & Winzelberg, J. (2012). Cognitive outcomes and familial stress after cochlear implantation in deaf children with and without developmental delays. *Otol Neurotol*. https://doi.org/10.1097/MAO.0b013e318259b72b

Osberger, M. J., Zimmerman-Phillips, S., & Koch, D. B. (2002). Cochlear implant candidacy and performance trends in children. *Ann Otol Rhinol Laryngol*, *111*(5 II), 62–65.

Porter, A., Creed, P., Hood, M., & Ching, T. Y. C. (2018). Parental decision-making and deaf children: A systematic literature review. *J Deaf Stud Deaf Educ*, *23*(4), 295–306. https://doi.org/10.1093/deafed/eny019

Raine, C. (2013). Cochlear implants in the United Kingdom: Awareness and utilization. *Cochlear Implants Int*. https://doi.org/10.1179/1467010013Z.00000000077

Ravi, R., & Gunjawate, D. R. (2020). Parent reported barriers and facilitators towards cochlear implantation – A systematic review: Barriers and facilitators in cochlear implantation. *Int J Pediatr Otorhinolaryngol*. https://doi.org/10.1016/j.ijporl.2020.110163

Roberts, R. M., Sands, F., Gannoni, A., & Marciano, T. (2015). Perceptions of the support that mothers and fathers of children with cochlear implants receive in South Australia: A qualitative study. *Int J Audiol*, *54*(12), 942–950. https://doi.org/10.3109/14992027.2015.1060641

Robertson, E. G., Wakefield, C. E., Signorelli, C., Cohn, R. J., Patenaude, A., Foster, C., Pettit,

T., & Fardell, J. (2018). Strategies to facilitate shared decision-making about pediatric oncology clinical trial enrollment: A systematic review. *Patient Educ Couns, 101*(7), 1157–1174. https://doi.org/10.1016/j.pec.2018.02.001

Sisk, B. A., Kang, T. I., Goldstein, R., DuBois, J. M., & Mack, J. W. (2019). Decisional burden among parents of children with cancer. *Cancer, 125*(8), 1365–1372. https://doi.org/10.1002/cncr.31939

Skarzynski, H. (2012). Ten years experience with a new strategy of partial deafness treatment. *J Hear Sci, 2*(2), 11–18.

Skarzynski, H., Lorens, A., Piotrowska, A., & Anderson, I. (2006). Partial deafness cochlear implantation provides benefit to a new population of individuals with hearing loss. *Acta Oto-Laryngologica*, *126*(9), 934–940. https://doi.org/10.1080/00016480600606632

Steinberg, Annie, Brainsky, A., Bain, L., Montoya, L., Indenbaum, M., & Potsic, W. (2000). Parental values in the decision about cochlear implantation. *Int J Pediatr Otorhinolaryngol*, *55*(2), 99–107. https://doi.org/10.1016/S0165-5876(00)00373-6

Sweeney, A. D., Hunter, J. B., Carlson, M. L., Rivas, A., Bennett, M. L., Gifford, R. H., Noble, J. H., Haynes, D. S., Labadie, R. F., & Wanna, G. B. (2016). Durability of hearing preservation after cochlear implantation with conventional-length electrodes and scala tympani insertion. *Otolaryngol Head Neck Surg, 154*(5), 907–913. https://doi.org/10.1177/0194599816630545

Thorne, S., Kirkham, S. R., & O'Flynn-Magee, K. (2004). The analytic challenge in interpretive description. *Int J Qual Methods, 3*(1), 1–11. https://doi.org/10.1177/160940690400300101

Wiley, S., Meinzen-Derr, J., Grether, S., Choo, D. I., & Hughes, M. L. (2012). Longitudinal

functional performance among children with cochlear implants and disabilities: A prospective study using the Pediatric Evaluation of Disability Inventory. *Int J Pediatr Otorhinolaryngol*, *76*(5), 693–697. https://doi.org/10.1016/j.ijporl.2012.02.022

Zaidman-Zait, A., Curle, D., Jamieson, J. R., Chia, R., & Kozak, F. K. (2015). Cochlear implantation among deaf children with additional disabilities: Parental perceptions of benefits, challenges, and service provision. *J Deaf Stud Deaf Educ*, *20*(1), 41–50. https://doi.org/10.1093/deafed/enu030

Zanetti, D., Nassif, N., & Redaelli De Zinis, L. O. (2015). Factors affecting residual hearing preservation in cochlear implantation. *Acta Otorhinolaryngol Ital*, *35*(6), 433–441. https://doi.org/10.14639/0392-100X-619

Chapter 5: Interviews with practitioners

Cochlear implant decision-making for children with residual hearing: Perspectives of practitioners

Formatted for the Journal of Deaf Studies and Deaf Education

Abstract

Children with residual hearing are eligible for consideration as cochlear implant (CI) candidates. However, the decision-making process for families and practitioners of these children is particularly challenging because there is no clear audiological cut-point for CI candidacy. This study aimed to understand practitioners' perspectives of the CI decision-making process for families of children with residual hearing. A qualitative research design was employed. Four focus groups and one individual interview were conducted with a total of 17 practitioners. Data were organized into six broad domains: candidacy issues for children with residual hearing, parents' expectations and concerns, practitioner's roles in decision support, additional considerations affecting decision-making, factors facilitating decision-making, and practitioners' needs. We learned that practitioners' confidence in determining candidacy and supporting parents has increased due to their experiences with positive outcomes for these children. They indicated that there was a need for more research to guide the decision-making process for parents.

Key Words: *cochlear implantation; residual hearing; decision-making; decision support; pediatric; qualitative study*

Introduction

There are an increasing number of studies that report positive speech, language, and auditory outcomes in children with residual hearing who receive cochlear implants (CIs) (Dettman et al., 2004; Fitzpatrick et al., 2006; Gratacap et al., 2015; Michael et al., 2019). There is considerable interest in expanding audiometric CI candidacy criteria to include children with residual hearing (Carlson et al., 2015; Leigh et al., 2016; Skarzynski et al., 2015; Vincenti et al., 2014). However, there remains uncertainty in recommending CIs for children who have residual hearing because there is no clear audiological cut-point for this population of children, and because the evidence informing clinical practices for these children is very limited (Dettman et al., 2004; Fitzpatrick et al., 2009; Hyde et al., 2010).

Furthermore, as with all CI surgery, risks and negative consequences may occur after surgery for children with residual hearing, including surgical complications, negative outcomes with CI, device-related complications, and loss of residual hearing, which may impact the children's health and future quality of life (Alegre-de la Rosa & Villar-Angulo, 2020; Chute & Nevins, 2002; Ganek et al., 2020). For these reasons, parents and practitioners may be uncertain as to whether the potential benefits outweigh the risks related to CIs (Greaver et al., 2017; Hardonk et al., 2010; Li et al., 2004; Steinberg et al., 2000). Therefore, decision-making about cochlear implantation for children with residual hearing who already benefit from HAs is less straightforward than for children with bilateral profound hearing loss. Several studies related to CI decision-making have reported that parents of children who had more preoperative residual hearing took more decision-making time and experienced more stress than parents of children with bilateral profound hearing loss (Burger et al., 2005; Fitzpatrick et al., 2009; Hyde et al., 2010a). Parents of these children may experience uncertainty and decisional conflict, which can

lead to delays in decision-making (Anmyr et al., 2016; Hardonk et al., 2010, 2011; O'Connor, 2010).

Shared decision making (SDM) is one way to arrive at informed and value-based clinical decisions regarding intervention plans with mutual understanding and agreement between patients and practitioners (Barry & Edgman-Levitan, 2012; Edwards & Elwyn, 2009). SDM is a collaborative decision-making process where patients and practitioners make decisions together using the best available information about the likely benefits and risks of available interventions, and where patients are supported to arrive at informed preferences (Gabe et al., 2004; Makoul & Clayman, 2006). Other healthcare decision-making studies have reported that by applying SDM, patients' knowledge improved, both patients and practitioners felt respected and satisfied, and they were more comfortably invested in the outcomes (Sepucha & Mulley, 2009; Whitney et al., 2008; Whitney & McCullough, 2007). In healthcare decision-making, many families and practitioners preferred to apply SDM, but preferences and values for decision-making involvement can vary among families and across decisions (Coyne et al., 2016; Lipstein et al., 2014). In addition, as several studies have reported, the preference of parents for information and their feelings about the sufficiency of information differs among families and across decisions (Harrison, 2004; Robertson et al., 2018).

Several studies have noted that research specifically on SDM in audiology clinical practice is required (Johnston et al., 2008; Porter et al., 2018; Pryce & Hall, 2014; Steinberg et al., 2003). However, very few studies have explored how SDM could be used by parents and practitioners in decision-making regarding children with hearing loss. It is important for parents and practitioners to work together in decision-making so that they can improve decision satisfaction and reduce the risk of decisional conflict and regret (Mack et al., 2016; Mckenna et

al., 2010; Sisk et al., 2019). There is virtually no research focused on either decision needs or parental involvement in CI decision-making for children with residual hearing.

Therefore, this study explored practitioners' views during the CI decision-making process as the first step to understanding decision-making for these children. With a better understanding of practitioners' experiences, this information can help practitioners assist in best-practice decision-making interventions for the parents of these children. This study aimed to explore practitioners' views and perceptions and their needs in supporting CI decision-making for parents of children with residual hearing.

Methods

Study design

A qualitative research study design using interpretive description methodology (Thorne et al., 2004). was employed to understand the perspectives of CI practitioners during the CI decision-making process for parents of children with preoperative residual hearing. Semi-structured focus groups (n=4) and one individual interview were conducted with open-ended questions to encourage participants to share their perspectives. Ethical approval for the research was obtained from the Children's Hospital of Eastern Ontario (CHEO) Research Institute, the Ottawa-Carleton Research and Evaluation Advisory Committee, and the University of Ottawa Ethics Boards (Appendix E).

Participants

Participants were recruited for this study between November 2018 and May 2019 in Ottawa, Ontario. Participants included two groups of practitioners, one group from a hospital-based CI

program and the other from two local school boards.

Clinicians

Clinicians from a hospital-based CI program were recruited through the Audiology clinic at CHEO, a major Canadian pediatric audiology center, which serves a population of approximately one million people. At this CI program, all children received CI in a publicly funded health care setting and were provided with pre-implant CI candidacy assessments, surgery and audiological follow-up services. All audiologists, auditory-verbal therapists (AVTs), CI surgeons, and psychologists providing CI services were invited to participate in the interview.

Specialized teachers of the deaf and hard of hearing

Teachers of the deaf and hard of hearing who had experience in referring children and guiding parents in exploring CI were recruited from two main local school districts in Ottawa. With the school boards' ethics approval, a recruitment poster, information letter, and consent form were sent to the local school boards.

Characteristics of participants

A total of 17 practitioners, including 8 CI clinicians (three audiologists, three AVTs, a CI surgeon, and a psychologist) and 9 teachers who had experience working with children with CI, accepted to participate in the interviews. All participants completed a consent form before the interviews. A description of participant characteristics is summarized in Table 1.

Procedure

An interview guide was developed based on the Decisional Needs Assessment in Populations (Jacobsen et al., 2013) (see Appendix F). A semi-structured format was selected to guide the interviews. Interviews were conducted between February 2019 and May 2019 with a total of four focus groups (three teacher groups and one clinician group) and one individual interview with a CI surgeon. Before the interview, practitioners were asked to complete a questionnaire to provide basic demographic information (i.e., discipline, work experience). The interviews were conducted in person by the main researcher in meeting rooms of the hospital or the school boards. Using an interview guide, the interviewer asked questions to encourage practitioners to share their experiences about the process of supporting decision-making for parents and children, as well as their own needs in supporting parents. Consistent with qualitative methods, the interviewers also formulated subsequent questions in response to the answers. For each interview, two researchers were present and field notes were taken during the interviews by one experienced researcher. Two trained research assistants listened to all the recordings of the focus group interviews and transcribed them, supplemented by field notes. The researcher who conducted the interviews verified the transcriptions.

Data analysis

Conducting and transcribing interviews proceeded concurrently since preliminary analysis can guide subsequent data collection (DiCicco-Bloom & Crabtree, 2006). The interview data were coded and analyzed using NVivo (QSR International Pty Ltd. Version 12.0), a software program used for qualitative studies. A thematic analysis approach was applied to analyze the interview data. The coding process involved a constant comparative method based on open, axial, and

selective coding methods (Strauss & Corbin, 2008). The main researcher created initial codes, similar codes were grouped into categories, and then these categories were collapsed into themes. Themes and sub-themes were identified by comparing code frequencies, relationships, and similarities between codes. A trained researcher coded and a second researcher verified the codes, and if any of the codings were unclear, this was discussed between the two researchers and resolved.

Results

Six major themes emerged from the interview data: candidacy issues for children with residual hearing, parents' expectations and concerns related to CI, practitioner's roles in decision support, additional considerations affecting decision-making, factors facilitating the decision-making process, and practitioners' needs in supporting the decision-making process. The themes and sub-themes are described in Table 2. The following section elaborates on each theme with examples of practitioners' comments. Numbers in parentheses refer to the study participant numbers.

Candidacy issues for children with residual hearing

Challenges in CI candidacy decision-making

Practitioners noted that there were two sets of challenges: one in deciding that a CI might be of benefit and then recommending it to the parents, and the second in supporting the parents' decision-making. Practitioners frequently mentioned that the absence of clear audiometric CI candidacy criteria for these children in a clinical setting was a major difficulty during the CI candidacy process.

> *You can think of a flat line, you know, a 75, 80, 80, 80, 80 dB which is like, still a lot of hearing. I think they're borderline because they're not really in the 85-90 dB. Like, the 50, 50, 70 and then, 90, 95. Not a lot of highs [high frequencies] there, but they have a lot of hearing, too. They're the kids usually that don't want the implant because they're not hearing 's' sounds but they know it's there … When they have that much hearing, you know, CI might help, but I don't know ... (Clinician_01, FG 01)*

Speech recognition ability is a common measure to help determine CI candidacy, and according to the practitioners, it is even more important to consider when children have residual hearing. This greater reliance on speech recognition for these children is problematic when children have limited skills. In these interviews, it was particularly difficult for the clinicians when they had to consider the speech recognition abilities of younger children whose linguistic capacities were not sufficient to objectively assess speech understanding.

> *I think it's harder for the younger kids when they've got residual hearing because you don't have speech perception testing. Sometimes you don't really know some of what the entire picture's going to be. (Clinician_04, FG 01)*

Factors affecting the recommendation of CI

As shown in Table 3, both practitioners mentioned several factors beyond children's audiometric thresholds and speech recognition abilities that influenced CI decision-making. Similar to typical CI recommendations, practitioners reported auditory functioning and speech recognition ability as factors affecting their recommendations for children with residual hearing. However, practitioners highlighted that the evaluation for CI should include assessments beyond hearing thresholds and stressed the importance of evaluating broader hearing configuration and overall

auditory function, such as listening effort and fatigue, social behaviour, and progress in therapy. In particular, teachers were focused on classroom functioning when recommending CI for these children.

Parents' expectations and concerns related to CI

Practitioners talked about parental expectations and concerns that were observed during the CI decision-making process.

Parents refused CI or needed time for CI decision-making

Parents of children with residual hearing sometimes refused CI or needed considerable time to work through the process of decision-making because they believed their children still received enough benefit from hearing aids (HAs). According to the teachers, parents tended to sometimes refuse CIs or delay decision-making due to the uncertainty of whether the potential benefits outweigh the negative outcomes related to CIs. In particular, CI clinicians perceived that parents needed time to understand their children's language gaps and communication difficulties in different environments with their HAs.

> *She is still managing well with the hearing aids. Could she be doing better with the cochlear implant? Possibly? But parents are on the fence still ... If her hearing is stable, which is hard to say. I think there is going to be more of a drop in the future. They're probably just going to stay on the fence until that does happen. And maybe they will look at it then ... (Teacher_04, FG 02)*
>
> *They're still benefitting, you know, somewhat with their hearing aids, or they appear to be. The parents think they're doing well with their hearing aids, so it's kind of hard to get*

them to understand that children are still missing out on a lot of information, especially the high-frequency information. (Clinician_05, FG 01)

Practitioners also reported that parents of these children tended to wait to proceed with CIs because they were looking for the right timing for their children. For example, when a child started school or changed schools, parents sometimes delayed the decision until their child adapted to the new environment.

Sometimes because these children are also of varying ages and we know that there is a fair commitment with new learning to listen, challenges and getting used to the CI. Sometimes where they are school age, parents may say, 'I don't want for it to coincide with the entry of kindergarten'. You know, 'I want the child to be settled in that environment, so let see how they adapt in that first year'... I think parents do look at, like, when is the right time ... (Clinician_06, FG 01)

Parents' expectations

According to the interviews, parents expected positive CI outcomes related to language development and spoken communication. In particular, CI clinicians stated that parents often identified specific areas where they hoped to see improvement, such as speech articulation.

They care about speech articulation errors that might happen with the borderline [candidates] with the English language, it affects also grammar, [and] voice quality ... and that was influencing the parents' decisions to perhaps move forward because this is important for some families. (Clinician_03, FG 01)

Additionally, clinicians commented that the social isolation of these specific children also affected parental CI decision-making, and these parents hoped using CI could address the social isolation of children.

> *I think that the kind of situation that the little one in a daycare setting where she was just really isolated and that was obvious to the parents ... She had lost hearing, and there she was all by herself in her daycare setting, actually not really wanting people in her little bubble because it was hard, so that influenced those parents' decision to move forward [with] a cochlear implant because they did notice that she had become increasingly socially isolated. (Clinician_02, FG 01)*

Practitioners in both groups described that some parents of children with hearing loss who had been developing satisfactory speech and language skills and then experienced deterioration in their hearing expected that CI could restore their children's previous abilities. Sometimes, this expectation seemed too high to the practitioners. Some parents expected that CI could 'cure' or 'fix' the child's hearing loss, as illustrated in the quote below:

> *They have a hope that putting an implant in will be a miracle solution and bring back normal hearing even though they know that's not likely ... (Clinician_01, ID 01)*
>
> *They're gonna fix his hearing, there was no other discussion, there was no consideration of anything, and was very adamant that that was going to happen. Um, for the other two, parents thought, CI is going to make their child hear normally, this would restore their hearing. They were very unrealistic in their expectations, even though I reminded them, it doesn't work exactly as you think. ... (Teacher_01, FG 04)*

Parents' concerns about CI

As shown in Table 4, several practitioners reported that families of children with residual hearing expressed various concerns during the decision-making process. The major parental concerns during the process included fear of surgery, negative consequences including loss of residual hearing, and outcomes after CIs.

Practitioners' roles in decision support

Through the interviews, CI practitioners described how they supported the parents throughout the decision process. Practitioners respected parents as the final decision-makers, and assisted parents largely by providing information and support.

> *Sometimes, like, the team will decide, sure, like, they're a good candidate, but then the parents will again decide to put things on hold. They're not ready, especially for those kids who are more borderline ... well, she's still very borderline, like, at this point, again, the motivation has to come from the parents. (Clinician_05, FG 01)*

Providing information to the families

Practitioners stressed that information from different resources was important for families to guide them in making an informed decision.

> *We try to provide the parents with as much information as possible so that they can make an informed decision (Clinician_06, FG 01)*

Some practitioners noted that parents of children with residual hearing often choose CI with unrealistic expectations of outcomes following CI and they emphasized the importance of providing realistic information. One clinician said that realistic information means that parents

are educated about all the risks and complications, as well as the benefits of CI surgery. In addition, the clinician reported that they explain the pros and cons of various types of CI devices from each manufacturer, so that parents can make informed decisions.

> *We do talk about all the risks and complications to try to give them a realistic picture of what's going to happen while the child is here at CHEO with the surgery and the aftercare. We talk about the various device types and then advantages and disadvantages or benefits of one manufacturer over the other. But you know, it is not that one manufacturer is better than the others. It's just that each will have different approaches in terms of pairing with a contralateral hearing aid or assist hearing devices … I give them some guidance along with that. We have long discussions … basically trading off the residual hearing. (Clinician_01, ID 01)*

Practitioners reported that for these families, considerations about surgery were particularly important. CI clinicians reported that it was essential to discuss hearing preservation with these families, particularly new CI technology and surgical techniques to preserve residual hearing. For example, one CI clinician reported providing comprehensive information about CI technology from different manufacturers to parents in order to assist them in choosing one according to their values.

> *If there's a lot of residual hearing then we might have had this occasionally where the family is really concerned about saving the residual hearing. What you can counsel them on [is] the various types of electrodes between the manufacturers. Over the years, the electrodes have changed to become thinner and less traumatic, some of them now come pre-coiled, they tend to coil as opposed to being straight. So, we have a long discussion about, you know, the pros and cons of the various manufacturers on that. I would say that*

most of the time you have a discussion with families, most of them will end up saying well, they'll pick one, another one of the manufacturers where there's a little bit more of an opportunity to either pair with a contralateral hearing aid, or then pair with a contralateral CI, or better wireless. (Clinician_01, ID 01)

Teachers provided information about outcomes in the school environment for children with residual hearing who received CIs. They particularly shared with parents their observations about changes in children's social behaviour and classroom functioning.

I share my previous students' experiences. I've had both positive and not as positive cases with the CI. Getting an implant and then your kid hears, there's a lot of other factors. I never try to say yes or no, because I'm not an audiologist or on this panel and there are so many other considerations, but I think my role is just to share my experiences, both positive and negative cases. (Teacher_03, FG 02)

Practitioners highlighted that families of children who had already received a CI were also an important resource for the families during the CI process. They made efforts to connect parents since some practitioners expected that these experienced families could provide practical information to families during the decision-making process.

And making sure that parents are connected with other parents because that was their biggest thing throughout the whole process. I don't know how much chance they get to talk to other parents going through it, but that could help them get better realistic information and expectations. (Teacher_01, FG 03)

Support for the families

Both CI clinicians and teachers described that they met families several times during the CI

decision-making process because they wanted parents to make the decision as comfortably as possible.

> *I don't find that we rush the consult in any way. We spend a lot of time going over that, and some families need to meet two or three times before they're comfortable making a decision. (Clinician_01, ID 02)*

In particular, some teachers encouraged older children to be involved in the decision-making process.

> *Well, in this case, because it was an older student, I counselled her directly. Not so much to parent, and just encouraging her to be open-minded to this and to go back and talk to the audiologist more to get her fears relieved ... (Teacher_05, FG 02)*

Additional considerations affecting decision-making

CI for children with additional disabilities and medical complexity

Practitioners noted that CI decision-making for children with residual hearing was more complicated if they had additional disabilities beyond hearing loss or if they had medical complexity.

> *I do have one right now that also has Usher [syndrome] but he got a stable hearing loss that's kind of moderate to moderately severe, kind of in that range. I think if the vision goes later, if you [the kids] lose more hearing, then it could be late [for CIs]. I think, he might be one that they consider, because what else are you going to do, especially if you lose more hearing. (Teacher_01, FG 03)*

CI for adolescents with residual hearing

The teachers talked about the difficulties that they faced in supporting the CI decisions for adolescents with residual hearing. Some adolescents were satisfied with their HAs because they had developed their own coping strategies to compensate for difficulties in hearing and communication. The teachers provided examples of adolescents having discontinued using their CIs, regretting their decision, or needing long-term rehabilitation after CIs.

> *...We have the later implanted kids who LOVED their hearing aids. And this used to happen more with the power hearing aids versus the newer digital hearing aids. There was really a depression almost when they got implanted, of 'this doesn't sound the same, doesn't sound the same, I want my hearing aids back.' … And it was weeks of tears with parents thinking, you know, 'what have I done', and there's no going back. As a clinician, it's hard to coach the parents through that and the child who's 'why'd you make me do this, I don't like this, I don't'. Usually, that's just a bump...They try, try, try and then they don't use it after a year or two… (Teacher_02, FG 04)*

Factors facilitating the decision-making process

Positive post-CI outcomes

CI clinicians stated that clinical examples of positive outcomes were useful in helping parents through the CI decision-making process. Clinicians indicated that based on their clinical experiences, they had observed many positive outcomes after Cis in children with residual hearing, and they had rarely seen kids perform worse with Cis.

> *In general, with the implant, they won't do worse than with the hearing aids, it's very rare that they do worse, but you'll probably do better. 'We can't guarantee you, but it'll*

be a success'. I've rarely, rarely, rarely seen a kid do much worse with the implants, especially with the borderlines, they've got so much going for them, you know all this residual hearing means like the nerves are still stimmed and they've had hearing aids. (Clinician_01, FG 01)

Multidisciplinary team approach

The CI clinicians, in particular, reported that a multidisciplinary approach was very helpful in supporting parents' decision-making. They commented that CI team meetings allowed them to share information, so that they could assist families in making more informed decisions. By sharing information and professional experiences among practitioners, they indicated that they felt more confident in presenting CI as an option for children with residual hearing and in supporting parents' decision-making process.

I think nothing beats spending a fair bit of time with them. I think what we do by having a regular comprehensive team meeting, I think is very helpful, I think that everybody's on the same page. So that's not directly with the family but that's when the CI team gets together [and] has a discussion on the cases, at a big round table. (Clinician_02, FG 01)

New developments in CI technology and surgical techniques

The development of new CI technology and surgical techniques to preserve residual hearing with CI has allowed CI clinicians to be more comfortable in recommending a CI. From the practitioners' viewpoint, parents also tended to derive some comfort from this additional information when making a decision about CI for their children.

We've done an electro-acoustic stimulation implant. Those are very thin electrodes and

shorter, and it preserves more [hearing]. It is particularly preserved low-frequency hearing ... I think there is a lot of comfort for the families that are maybe possible to save the residual hearing … So, does it change our decision that much? Perhaps a bit, because as I say the electrodes are a little less traumatic. (Clinician_01, ID 01)

Practitioners' needs in supporting the decision-making process

Different needs were expressed by different practitioner groups. Teachers stressed the importance of sharing their perspectives on the child's functioning in school and everyday functioning. In addition, they suggested some ideas on how to put this information into action.

Collaboration between CI clinicians and teachers

CI clinicians reported that sharing information through the CI clinicians' team meetings facilitated support for parents during the decision process. However, they indicated that they need more discussion with teachers related to CI candidacy recommendations and parental decision-making process. The teachers also frequently indicated that more active communication with CI clinicians is necessary.

I begged to be part of it. Because I think that the CI committee was meeting about my student and really, I'm the only one who's had her for three months, so I've had this intensive relationship with her and I just had to send an email hoping that somebody might mention what my input would be … If they're considering one of my students at a CI meeting, I will, you know, share what I've observed, these are my clinical notes, these are my diagnostics. I would say that functionally. (Teacher_07, FG 02)

Additional information

All practitioners mentioned that clinical examples of positive outcomes helped them to support the decision-making process; however, practitioners noted that evidence-based information for this particular group of children is still lacking. Both groups of practitioners stressed the need for quantitative and qualitative research among children with residual hearing to guide decision-making.

> *I don't even bring that it is one of the main things that, I look at with residual hearing and I don't think I even bring that up with the parents enough when I'm talking to them. There is no scale to measure the effort of listening. It's very subjective. [We need] research and studies. (Clinician_01, FG 01)*

More specifically, practitioners highlighted the need for clinical studies that show the benefits and risks of CI versus HAs, how CI technology works for children with residual hearing, advanced CI technology for residual hearing preservation, and the experiences of families of other CI recipients.

> *I wish we could quantitatively measure the cost to that child of trying to listen with hearing aid versus the ease we know that kids listen with cochlear implants. If there was a definitive quantitative way to express that to parents, we all know this ski-slope hearing loss, and often kids won't even wear the hearing aids they're so bad, right? (Teacher_01, FG 02)*

Decision support tools

Practitioners suggested that decision support tools such as decision coaching or decision aids for parents could be helpful in supporting CI decision-making. They felt that these tools could

translate information to make it very accessible for parents and children. All practitioners recommended that it would be helpful for these tools to contain statistical information about the probabilities of benefits and risks of CI versus HAs, the experiences of other children with residual hearing, and children's actual hearing function in real-life auditory environments. These tools could be presented to parents through easily accessible formats, (e.g., using visual materials), via seminars, forums, peer support programs, questionnaires, pictograms, or video clips. Practitioners also commented that decision support interventions may be needed (e.g., decision aids, decision coaching, health provider training on SDM). Some practitioners mentioned culturally diverse families (e.g., English as a second language [ESL] families) and noted that visual materials and tools using appropriate language-levels would help to support these families in their decision-making.

> *Like having the visual there, explaining your likelihood of facial nerve paralysis is X percent or whatever. Maybe if they had a little picture that showed the likelihood of their kids improving. The majority of families would find that helpful. You do get a few families that are really kind of unsure where to go. (Clinician_01, ID 01)*

> *Decision aids … They will do things, like make it very visual. Like I have one-hundred happy faces and colour the percentage where there are complications. So, you could see how small it is … That would be good. Yeah, especially for English as a second language. People would like to see in terms of pictorial ... that would be the long-term goal. (Teacher_01, FG 04)*

Discussion

The aim of this study was to explore the experiences and perspectives of practitioners in supporting CI decision-making for parents of children with residual hearing. Practitioners provided comprehensive information about the decision process for children with residual hearing throughout the interviews. We identified factors affecting practitioners' recommendations, parents' expectations and concerns related to CI, practitioners' roles and considerations in parent support, practitioners' perceptions of facilitating factors, and their needs in supporting the CI decision-making process.

Practitioners reported that CI recommendations for these children were challenging because of the variability in audiometric candidacy criteria and incomplete speech recognition or auditory functioning test results with young children. Our finding is consistent with previous studies which indicated that it was more challenging for practitioners to recommend CI for these children than for those with profound hearing loss due to lack of clear audiological cut-points for CI surgery (Dowell et al., 2004; Fitzpatrick et al., 2009; Hyde et al., 2010). As previous studies have reported (Fitzpatrick et al., 2009; Leigh et al., 2011, 2016), our study also found that practitioners found it particularly difficult to make CI candidacy decisions for children whose linguistic capabilities were not sufficient to objectively assess speech understanding. Due to these difficulties, practitioners stressed the need to consider children's auditory functioning, speech recognition abilities, listening effort and fatigue, social behaviour, classroom functioning, and progress in therapy.

The importance accorded to children's everyday functioning in social and classroom settings has also been highlighted in multiple studies on typical CI candidates (De Raeve, 2016; Fitzpatrick et al., 2009; O'Brien et al., 2012). In particular, previous research (Fitzpatrick et al.,

2009) that was conducted at the same pediatric CI program as the present study reported that very similar factors including spoken language abilities and academic achievement were important considerations. Taken together, these findings highlight the need for considering functioning in multiple domains, beyond those that are typically measured for CI, for these children. Considering multiple factors, beyond measurable assessments for these children, appeared to somewhat compensate for practitioners' uncertainty during the decision-making process, given the variability of audiometric candidacy criteria in these children.

Throughout the interviews, practitioners talked extensively about parents' expectations and concerns in making decisions for their children with residual hearing. In their experience, they perceived that some parents declined to proceed with a CI or waited on decision-making because they believed that their children received sufficient benefits from HAs. As previous research has indicated (Leigh et al., 2016; Hyde et al., 2010; Fitzpatrick et al., 2009), CI decision-making for these children was even more challenging compared to decision-making for children with more severe hearing loss because they showed progress in speech and language development with HAs. Practitioners were concerned that some parents had such unrealistic expectations for progress in speech and language development that these hopes needed to be carefully managed. Our study found that, particularly for children who had developed good speech and language abilities and had experienced progressive hearing loss, parents had high expectations, sometimes expecting that CI could restore their children's previous abilities.

In our findings, practitioners stressed that they play important roles in providing up-to-date information and support to parents. This finding is similar to practitioners' roles in the CI decision-making process for pediatric CI candidacy more generally (Hyde et al., 2010; Johnston et al., 2008; Roberts et al., 2015). However, because of the special considerations associated with

residual hearing, practitioners focused more on discussing hearing preservation including new technology and surgical techniques for children with residual hearing, stressing that surgery and loss of residual hearing were major concerns for parents.

Practitioners also talked about additional considerations affecting the decision-making process. Children with residual hearing who have additional disabilities and medical complexities were viewed as particularly complicated cases, due to insufficient language abilities for assessment and limited methods for assessing these children's auditory functioning. Previous studies exploring parental CI decision-making (Wiley et al., 2012; Zaidman-Zait et al., 2015) have also noted the unique challenges in pre- and post-CI assessment and rehabilitation in this heterogeneous population with additional disabilities or medical issues. A previous study in general medical decision-making also found that families from various cultural backgrounds experience the decision-making process less positively than do parents from the majority culture (Hawley et al., 2017). Parents' knowledge regarding hearing loss, its treatment, different CI candidacy criteria, and parents' involvement in their children's rehabilitation differ among cultures, and practitioners may need to consider these factors to support parental decision-making in a sensitive manner.

Practitioners also indicated that supporting CI decision-making for adolescents with residual hearing requires a different approach. As Hyde et al. (2010) reported, CI decision-making for adolescents, who are at a particularly sensitive time in their social and emotional development is difficult because adolescents are not confident in weighing the pros and cons of implantation. As other healthcare studies have indicated, adolescents may also be uncertain because of a lack of experience in decision-making (Decker et al., 2004; Robertson et al., 2019). In our study, one of the major reasons that practitioners found adolescents to be more

challenging was that they had developed their own coping strategies (e.g., lip-reading, sign language) to compensate for communication difficulties and therefore often perceived that they did not need CIs. These findings suggest that better strategies for assessing pre-and post-CI abilities depending on different clinical profiles, cultures, and ages may be needed to help support parents and adolescents through the decision-making process.

Practitioners reflected on several factors that assist them in guiding parental decision-making: their own experiences with positive post-CI outcomes in children with residual hearing, new developments in technology and surgical techniques, and a multidisciplinary team approach to decision-making. In a study that investigated barriers and facilitators to CI decision-making for both children and adults in Australia and the United Kingdom (Bierbaum et al., 2020), audiologists reported that sharing CI recipients' experiences with candidates played an important role in supporting decision-making. Audiologists who had these experiences felt more confident in selecting eligible candidates for CI assessments than audiologists who had fewer cases of implanted children with residual hearing. It is clear from our interviews that CI decision-making for children with residual hearing continues to create a certain discomfort, similar to conclusions from a previous study that was conducted 10 years ago with practitioners across Canada (Fitzpatrick et al., 2009). Since that time, there has been relatively little examination of practitioners' perspectives related to decision-making for this population. However, practitioners in our study specifically indicated that they now have more confidence in recommending CIs and supporting parents due to their increased experience with positive clinical outcomes in children with residual hearing and with applying a multidisciplinary approach. Practitioners' views about improvements in CI devices, electrode design, and surgical techniques are consistent with other reports that these developments have propelled the expansion of CI candidacy criteria to include

children with more residual hearing (Carlson et al., 2015, 2018; Gifford, 2016).

Although CI clinicians in this study reported that a multidisciplinary approach gave them more confidence in recommending CI for these children, teachers reported that teachers need a greater exchange of information with the clinic-based CI program and more involvement in the decision-making process. One previous study that explored practitioners' perspectives about the functioning of school-aged children with CI in the same region (Fitzpatrick et al., 2009) also found that teachers wanted to be more involved in pre-and post-implant meetings at the clinical program. For all practitioners, consideration of a child's social functioning is one of the most important factors in recommending CI; therefore, greater inclusion of teachers who observe and interact with these children in school settings may assist parents in decision-making as they work through the process.

In our study, practitioners highlighted the need for additional information in supporting the CI decision-making process for parents of children with residual hearing. Three major types of information were identified: 1) outcomes that extend beyond speech recognition to include benefits related to everyday functioning; 2) more up-to-date evidence about hearing preservation; and 3) experiences of families of other children with residual hearing who received CI. In addition, we found that there is a need to transfer this information to the parents in a family-friendly format. Several studies in the decision-making sciences have discussed the effectiveness of using plain language and visual aids for guiding health-related decision-making processes (Dansereau et al., 2013; Dansereau & Simpson, 2009; Houts et al., 2006; Langford et al., 2020; Westermann et al., 2013). Consistent with this research, in our study, practitioners felt that decision support tools including statistical information in plain language could help to support CI decision-making for these children. For example, practitioners discussed the use of decision

support interventions, such as decision aids or decision coaching that can more clearly present benefits and risks of CIs related to everyday functioning, taking into account parents' values and preferences, concerns, and decisional needs.

Limitations

We obtained valuable information from the findings of this study; however, one limitation of our research is that all participants were recruited from a single region in Canada where children received CI within a publicly funded health care setting. Future studies are needed to explore CI decision-making for these children in different healthcare settings, both private and public. In addition, CI programs vary in how they apply audiometric CI candidacy criteria among these children, and intervention models may differ with location and different cultural backgrounds. Therefore, it is anticipated that due to variation in CI candidacy decisions and experiences, the characteristics of children with residual hearing who receive CI may differ among programs.

Conclusion

This study allowed us to identify key aspects of the CI decision-making process for children with residual hearing from the perspectives of both hospital- and school-based practitioners who follow these children and families closely. Practitioners still find it quite challenging when considering this subpopulation of children for CI, both with respect to recommendations and decision support. However, their confidence in determining candidacy and supporting parents has increased largely due to their experiences with positive outcomes for these children. We found that practitioners primarily support parental decision-making by providing information on the practical aspects of the benefits and risks of CI. Despite not using the terms related to SDM

(e.g., shared decision-making, values and preferences) during the interviews, the discussions suggested that practitioners engage in SDM for these children because they consider parents' expectations, preferences, concerns, and decisional needs during the process. However, practitioners noted that more research among children with residual hearing is needed to guide decision-making. Additional studies with a focus specifically on SDM are required to examine the extent to which the approach is used and its effectiveness in supporting decision-making for children with residual hearing.

Table 1. Characteristics of 17 participants

	N	(%)
Sex (female), n (%)	15	(88.2)
Discipline, n (%)		
Clinicians	8	(47.1)
Audiologist	3	(17.6)
AVT	3	(17.6)
CI surgeon	1	(5.9)
Psychologist	1	(5.9)
Specialized teachers of the deaf and hard of hearing	9	(52.9)
Work experience, n (%)		
0-5 years	2	(11.8)
6-10 years	1	(5.8)
11-15 years	2	(11.8)
16-20 years	6	(35.3)
More than 20 years	6	(35.3)

n, number; CI, cochlear implant; AVT, auditory verbal therapist

Table 2. Themes and sub-themes from the interview data

Themes	Sub-themes
Candidacy issues for children with residual hearing	• Challenges in CI candidacy decision-making • Factors affecting the recommendation of CI
Parents' expectations and concerns related to CI	• Parents refused CI or needed time for decision-making • Parents' expectations • Parents' concerns about CI
Practitioner's roles in decision support	• Providing information to the families • Support for the families
Additional considerations affecting decision-making	• CI for children with additional disabilities and medical complexity • CI for adolescents with residual hearing
Factors facilitating the decision-making process	• Positive post-CI outcomes • Multidisciplinary team approach • New developments in CI technology and surgical techniques
Practitioners' needs in supporting the decision-making process	• Collaboration between CI clinicians and teachers • Additional information • Decision support tools

CI, cochlear implant

Table 3. Factors affecting the recommendation of CI and sample quotes

Factors affecting the recommendation of CI	**Sample quotes**
Auditory functioning	*Well, I think the one thing is that we don't look at the pure-tone average so much anymore. I think that's a big key [difference] from when we last looked at it, we're not looking specifically at PTA … or high-frequency PTA. (Clinician_04, FG 01)*
Speech recognition abilities	*Speech recognition is very important. A kid who gets like 76% in noise, it's very tricky and I understand there are other factors, like, you know, fatigue, but at the end of the day, these parents and these kids aren't necessarily the ones who are pushing implantation. (Clinician_01, FG 01)*
Listening effort and fatigue	*I think it was starting to be more things like more effort to listen. I think the child, too, would sort of initiating kind of controlled conversations to always be sort of the one in charge. They have difficulty when topics would change quickly. (Clinician_06, FG 01)*
Social behaviour	*They've stopped sort of joining in with things, they're becoming more of a wallflower, especially those progressive kids that are entering into that sort of that residual are you really going to implant them. (Clinician_04, FG 01)*
Classroom functioning	*A factor would be like how they're functioning in the classroom. If CHEO clinicians are considering one of my students at a CI meeting, I share what I've observed. These are my clinical notes and my diagnostics. I would say that functionally, like, she's missing 85% in the classroom, with an FM, with a powerful HA. (Teacher_02, FG 03)*
Progress in therapy	*If there's like, lack of progress. We test them repeatedly and see them not progress, you know, if they're not gonna get there then we look at the audiogram and the audiogram is in sort of around 80 dB or anything kind of severe, we just wish they'd get a CI. (Clinician_02, FG 01)*

CI, cochlear implant; PTA, pure-tone average; ID, individual interview; FG, focus group; FM, frequency modulation; HA, hearing aid

Table 4. Examples of parents' concerns and sample quotes

Parents' concerns about CI	Sample quotes
Anxiety about surgery	*I had a student who was really struggling with her hearing. She was an older student around the age of 17. And she actually was a cochlear implant candidate. CHEO audiologists suggested multiple times … Parents investigated this particularly when she was still pediatrics, but they were so afraid of the surgery, so afraid of pain, scarring, the process. So, they declined, but she struggled so much academically and socially. (Teacher_05, FG 02)*
Concern about the loss of residual hearing	*One of my main things is the parents' concern over losing that residual hearing ... Most of the kids with residual hearing, they can call them and at least have the awareness of sound. They could be without the sounds after the implant ... (Clinician_04, FG 01)*
Concern about the outcomes after CI	*The other thing they're looking for is that guarantee that it's going to be better than before, you know, can you guarantee that we're gonna make more progress on the language assessment next year? Can you guarantee that he's gonna be able to get through a day of school without an FM system? No, we don't make those kinds of guarantees but I think they're looking at, making a bargaining deal helps to somehow come to grips with the decision, so they're looking for guarantees, they're looking for reassurances. (Teacher_02, FG 04)*

CI, cochlear implant; FG, focus group; FM, frequency modulation

References

Alegre-de la Rosa, O. M., & Villar-Angulo, L. (2020). Health-related quality of life in children who use cochlear implants or hearing aids. *Heliyon*, *6*(1), e03114. https://doi.org/https://doi.org/10.1016/j.heliyon.2019.e03114

Barry, M. J., & Edgman-Levitan, S. (2012). Shared decision making - The pinnacle of patient-centered care. *N Engl J Med*, *366*(9), 780–781. https://doi.org/10.1056/NEJMp1109283

Bierbaum, M., McMahon, C. M., Hughes, S., Boisvert, I., Lau, A. Y. S., Braithwaite, J., & Rapport, F. (2020). Barriers and facilitators to cochlear implant uptake in Australia and the United Kingdom. *Ear Hear*, *41*(2), 374–385. https://doi.org/10.1097/AUD.0000000000000762

Carlson, M. L., O'Connell, B. P., Lohse, C. M., Driscoll, C. L., & Sweeney, A. D. (2018). Survey of the American Neurotology Society on cochlear implantation. *Otol Neurotol*, *39*(1), e12–e19. https://doi.org/10.1097/MAO.0000000000001631

Carlson, M. L., Sladen, D. P., Haynes, D. S., Driscoll, C. L., DeJong, M. D., Erickson, H. C., Sunderhaus, L. W., Hedley-Williams, A., Rosenzweig, E. A., Davis, T. J., & Gifford, R. H. (2015). Evidence for the expansion of pediatric cochlear implant candidacy. *Otol Neurotol*, *36*(1), 43–50. https://doi.org/10.1097/MAO.0000000000000607

Chute, P. M., & Nevins, M. E. (2002). *The parents 'guide to cochlear implants*. Gallaudet University Press.

Corbin, J., & Strauss, A. (2008). *Basics of qualitative research: Techniques and procedures for developing grounded theory*. Thousand Oaks, CA: Sage publications.

Coyne, I., O'Mathúna, D. P., Gibson, F., Shields, L., Leclercq, E., & Sheaf, G. (2016). Interventions for promoting participation in shared decision-making for children with

cancer. *Cochrane Database Syst Rev*, *11*. https://doi.org/10.1002/14651858.CD008970.pub3

Dansereau, D. F., Knight, D. K., & Flynn, P. M. (2013). Improving adolescent judgment and decision making. *Prof Psychol Res Pr*, *44*(4), 274. https://doi.org/10.1037/a0032495

Dansereau, D. F., & Simpson, D. D. (2009). A picture is worth a thousand words: The case for graphic representations. *Prof Psychol Res Pr*, *40*(1), 104. https://doi.org/10.1037/a0011827

De Raeve, L. (2016). Cochlear implants in Belgium: Prevalence in paediatric and adult cochlear implantation. *Eur Ann Otorhinolaryngol Head Neck Dis 133*, S57–S60. https://doi.org/10.1016/j.anorl.2016.04.018

Decker, C., Phillips, C. R., & Haase, J. E. (2004). Information needs of adolescents with cancer. *J Pediatr Oncol Nurs*, *21*(6), 327–334. https://doi.org/10.1177/1043454204269606

Dettman, S. J., Costa, W. A. D., Dowell, R. C., Winton, E. J., Hill, K. L., & Williams, S. S. (2004). Cochlear implants for children with significant residual hearing. *Arch Otolaryngol Head Neck Surg*, *130*(May), 612–618. https://doi.org/10.1001/archotol.130.5.612

DiCicco-Bloom, B., & Crabtree, B. F. (2006). The qualitative research interview. *Med Educ*, *40*(4), 314–321. https://doi.org/10.1111/j.1365-2929.2006.02418.x

Dowell, C., Hollow, R., & Winton, E. (2004). Outcomes for cochlear implant users with significant residual hearing. *Arch Otolaryngol Head Neck Surg*, *130*(May), 575–581. https://doi.org/10.1001/archotol.130.5.575

Duncan, J. (2009). Parental readiness for cochlear implant decision-making. *Cochlear Implants Int*, *10*(1), 38–42. https://doi.org/10.1002/cii.384

Edwards, A., & Elwyn, G. (2009). Shared decision-making in health care: Achieving evidence-based patient Choice. In A. Edwards & G. Elwyn (Eds.), *Oxford University Press*

(illustrate). OUP Oxford, 2009. https://doi.org/10.1007/s13398-014-0173-7.2

Fitzpatrick, E. M., McCrae, R., & Schramm, D. (2006). A retrospective study of cochlear implant outcomes in children with residual hearing. *BMC Ear Nose Throat Disord*, *6*(7), 1–6. https://doi.org/10.1186/1472-6815-6-7

Fitzpatrick, E. M., Olds, J., Durieux-Smith, A., McCrae, R., Schramm, D., & Gaboury, I. (2009). Pediatric cochlear implantation: How much hearing is too much? *Int J Audiol*, *48*(2), 91–97. https://doi.org/10.1080/14992020802516541

Gabe, J., Olumide, G., & Bury, M. (2004). "It takes three to tango": A framework for understanding patient partnership in paediatric clinics. *Soc Sci Med*, *59*(5), 1071–1079. https://doi.org/10.1016/j.socscimed.2003.09.035

Ganek, H. V., Feness, M. L., Goulding, G., Liberman, G. M., Steel, M. M., Ruderman, L. A., Papsin, B. C., Cushing, S. L., & Gordon, K. A. (2020). A survey of pediatric cochlear implant recipients as young adults. *Int J Pediatr Otorhinolaryngol*, *132*, 109902. https://doi.org/10.1016/j.ijporl.2020.109902

Gifford, R. H. (2016). Expansion of pediatric cochlear implant indications. *Hear J*, *December*, 8–10. https://doi.org/10.1097/01.HJ.0000511125.71672.3e

Gratacap, M., Thierry, B., Rouillon, I., Marlin, S., Garabedian, N., & Loundon, N. (2015). Pediatric cochlear implantation in residual hearing candidates. *Ann Otol Rhinol Laryngol*, *124*(6), 443–451. https://doi.org/10.1177/0003489414566121

Greaver, L., Eskridge, H., & Teagle, H. F. B. (2017). Considerations for pediatric cochlear implant recipients with unilateral or asymmetric hearing loss: Assessment, device fitting, and habilitation. *J Am Acad Audiol*, *26*(2), 91–98. https://doi.org/10.1044/2016_AJA-16-0051

Hardonk, S., Bosteels, S., Desnerck, G., Loots, G., Van Hove, G., Van Kerschaver, E., Vanroelen, C., & Louckx, F. (2010). Pediatric cochlear implantation: A qualitative study of parental decision-making processes in Flanders, Belgium. *Am Ann Deaf*, *155*(3), 339–352. https://doi.org/10.1353/aad.2010.0012

Harrison, C. (2004). Treatment decisions regarding infants, children and adolescents. *J Paediatr Child Health*, *9*(2), 99–103. https://doi.org/10.1093/pch/9.2.99

Hawley, K. A., Goldberg, D. M., & Anne, S. (2017). Utility of a multidisciplinary approach to pediatric hearing loss. *Am J Otolaryngol 38*(5), 547–550. https://doi.org/10.1016/j.amjoto.2017.05.008

Houts, P. S., Doak, C. C., Doak, L. G., & Loscalzo, M. J. (2006). The role of pictures in improving health communication: A review of research on attention, comprehension, recall, and adherence. *Patient Educ Couns*, *61*(2), 173–190. https://doi.org/10.1016/j.pec.2005.05.004

Hyde, M., Punch, R., & Komesaroff, L. (2010). Coming to a decision about cochlear implantation: Parents making choices for their deaf children. *J Deaf Stud Deaf Educ*, *15*(2), 162–178. https://doi.org/10.1093/deafed/enq004

Jacobsen, M. J., O'Connor, A., & Stacey, D. (2013). Decisional Needs Assessment in Populations: A workbook for assessing patients' and practitioners' decision-making needs. *Decisional Needs Assessment in Populations*.

Johnston, J. C., Durieux-Smith, A., Fitzpatrick, E., O'Connor, A., Benzies, K., & Douglas, A. (2008). An assessment of parents 'decision-making regarding paediatric cochlear implants. *Can J Speech-Lang Pathol Audiol*, *32*(4), 169–182.

Langford, A. T., Hawley, S. T., Stableford, S., Studts, J. L., & Byrne, M. M. (2020).

Development of a plain language decision support tool for cancer clinical trials: Blending health literacy, academic research, and minority patient perspectives. *J Cancer Educ*, *35*, 454–461. https://doi.org/10.1007/s13187-019-1482-5

Leigh, J., Dettman, S., Dowell, R., & Sarant, J. (2011). Evidence-based approach for making cochlear implant recommendations for infants with residual hearing. *Ear Hear*, *32*(3), 313–322. https://doi.org/10.1097/AUD.0b013e3182008b1c

Leigh, J. R., Dettman, S. J., & Dowell, R. C. (2016). Evidence-based guidelines for recommending cochlear implantation for young children: Audiological criteria and optimizing age at implantation. *Int J Audiol*, *55 Suppl 2*, S9–S18. https://doi.org/https://dx.doi.org/10.3109/14992027.2016.1157268

Li, Y., Bain, L., & Steinberg, A. G. (2004). Parental decision-making in considering cochlear implant technology for a deaf child. *Int J Pediatr Otorhinolaryngol*, *68*(8), 1027–1038. https://doi.org/10.1016/j.ijporl.2004.03.010

Lipstein, E. A., Dodds, C. M., & Britto, M. T. (2014). Real life clinic visits do not match the ideals of shared decision making. *J Pediatr*, *165*(1), 178–183. https://doi.org/10.1016/j.jpeds.2014.03.042

Mack, J. W., Cronin, A. M., & Kang, T. I. (2016). Decisional regret among parents of children with cancer. *J Clin Oncol*, *34*(33), 4023–4029. https://doi.org/10.1200/JCO.2016.69.1634

Makoul, G., & Clayman, M. L. (2006). An integrative model of shared decision making in medical encounters. *Patient Educ Couns*, *60*(3), 301–312. https://doi.org/10.1016/j.pec.2005.06.010

Mckenna, K., Collier, J., Hewitt, M., & Blake, H. (2010). Parental involvement in paediatric cancer treatment decisions. *Eur J Cancer Care*, *19*(5), 621–630.

https://doi.org/10.1111/j.1365-2354.2009.01116.x

Michael, R., Attias, J., & Raveh, E. (2019). Cochlear implantation and social-emotional functioning of children with hearing loss. *J Deaf Stud Deaf Educ*, *24*(1), 25–31. https://doi.org/https://dx.doi.org/10.1093/deafed/eny034

O'Brien, L. C. G., Valim, C., Neault, M., Kammerer, B., Clark, T., Johnston, J., Culver, S., Zhou, J., Kenna, M. A., & Licameli, G. R. (2012). Prognosis tool based on a modified children's implant profile for use in pediatric cochlear implant candidacy evaluation. *Ann Otol Rhinol Laryngol*, *121*(2), 73–84. https://doi.org/10.1177/000348941212100201

Porter, A., Creed, P., Hood, M., & Ching, T. Y. C. (2018). Parental decision-making and deaf children: A Systematic literature review. *J Deaf Stud Deaf Educ*, *23*(4), 295–306. https://doi.org/10.1093/

Pryce, H., & Hall, A. (2014). The role of shared decision-making in audiologic rehabilitation. *Perspectives on Aural Rehabilitation and Its Instrumentation*, *21*(1), 15–23. https://doi.org/10.1044/arri21.1.15

Roberts, R. M., Sands, F., Gannoni, A., & Marciano, T. (2015). Perceptions of the support that mothers and fathers of children with cochlear implants receive in South Australia: A qualitative study. *Int J Audiol*, *54*(12), 942–950. https://doi.org/10.3109/14992027.2015.1060641.

Robertson, E. G., Wakefield, C. E., Shaw, J., Darlington, A. S., McGill, B. C., Cohn, R. J., & Fardell, J. E. (2019). Decision-making in childhood cancer: parents 'and adolescents 'views and perceptions. *Supportive Care in Cancer*, *27*(11), 4331–4340. https://doi.org/10.1007/s00520-019-04728-x

Robertson, E. G., Wakefield, C. E., Signorelli, C., Cohn, R. J., Patenaude, A., Foster, C., Pettit,

T., & Fardell, J. (2018). Strategies to facilitate shared decision-making about pediatric oncology clinical trial enrollment: A systematic review. *Patient Educ Couns*, *101*(7), 1157–1174. https://doi.org/10.1016/j.pec.2018.02.001

Sepucha, K., & Mulley, A. G. (2009). A perspective on the patient's role in treatment decisions. *Med Care Res Rev*, *66*(1), 53S-74S. https://doi.org/10.1177/1077558708325511deafed/eny019

Sisk, B. A., Kang, T. I., Goldstein, R., DuBois, J. M., & Mack, J. W. (2019). Decisional burden among parents of children with cancer. *Cancer*, *125*(8), 1365–1372. https://doi.org/10.1002/cncr.31939

Skarzynski, H., Lorens, A., Dziendziel, B., & Skarzynski, P. H. (2015). Expanding pediatric cochlear implant candidacy: A case study of electro-natural stimulation (ENS) in partial deafness treatment. *Int J Pediatr Otorhinolaryngol*, *79*(11), 1896–1900. https://doi.org/https://dx.doi.org/10.1016/j.ijporl.2015.08.040

Steinberg, A., Bain, L., Li, Y., Delgado, G., & Ruperto, V. (2003). Decisions Hispanic families make after the identification of deafness. *J Deaf Stud Deaf Educ*, *8*(3), 291–314. https://doi.org/10.1093/deafed/eng016

Steinberg, Annie, Brainsky, A., Bain, L., Montoya, L., Indenbaum, M., & Potsic, W. (2000). Parental values in the decision about cochlear implantation. *Int J Pediatr Otorhinolaryngol*, *55*(2), 99–107. https://doi.org/10.1016/S0165-5876(00)00373-6

Thorne, S., Kirkham, S. R., & O'Flynn-Magee, K. (2004). The analytic challenge in interpretive description. *Int J Qual Methods*, *3*(1), 1–11. https://doi.org/10.1177/160940690400300101

Vincenti, V., Bacciu, A., Guida, M., Marra, F., Bertoldi, B., Bacciu, S., & Pasanisi, E. (2014).

Pediatric cochlear implantation: An update. *Ital J Pediatr*, *40*, 72. https://doi.org/https://dx.doi.org/10.1186/s13052-014-0072-8

Westermann, G. M. A., Verheij, F., Winkens, B., Verhulst, F. C., & Van Oort, F. V. A. (2013). Structured shared decision-making using dialogue and visualization: A randomized controlled trial. *Patient Educ Couns*, *90*(1), 74–81. https://doi.org/10.1016/j.pec.2012.09.014

Whitney, S. N., Holmes-Rovner, M., Brody, H., Schneider, C., McCullough, L. B., Volk, R. J., & McGuire, A. L. (2008). Beyond shared decision making: An expanded typology of medical decisions. *Med Decis Making*, *28*(5), 699–705. https://doi.org/10.1177/0272989X08318465

Whitney, S. N., & McCullough, L. B. (2007). Physicians 'silent decisions: Because patient autonomy does not always come first. *Am J Bioeth*, *7*(7), 33–38. https://doi.org/10.1080/15265160701399735

Wiley, S., Meinzen-Derr, J., Grether, S., Choo, D. I., & Hughes, M. L. (2012). Longitudinal functional performance among children with cochlear implants and disabilities: A prospective study using the Pediatric Evaluation of Disability Inventory. *Int J Pediatr Otorhinolaryngol*, *76*(5), 693–697. https://doi.org/10.1016/j.ijporl.2012.02.022

Zaidman-Zait, A., Curle, D., Jamieson, J. R., Chia, R., & Kozak, F. K. (2015). Cochlear implantation among deaf children with additional disabilities: Parental perceptions of benefits, challenges, and service provision. *J Deaf Stud Deaf Educ*, *20*(1), 41–50. https://doi.org/10.1093/deafed/enu030

Chapter 6: Integrated Discussion

CHAPTER 6: Integrated Discussion

Background

Cochlear implants (CIs) have gradually become more popular as an intervention option for children with residual hearing. Despite this trend, clinicians may still be uncomfortable recommending CIs for these children due to the variability in audiometric candidacy criteria in individual clinical practice (de Kleijn et al., 2018; Fitzpatrick et al., 2009; Hyde et al., 2010). This variation in determining CI candidacy for children with residual hearing may result in healthcare inequities in terms of access to optimal hearing technology (Carlson et al., 2018). It is generally agreed that during the CI assessment process, families and practitioners need to consider individual characteristics and environmental factors rather than simply adhering to audiometric criteria, due to the range of auditory and language performance in children (Chundu & Flynn, 2014; Fitzpatrick et al., 2009).

Decision-making about CIs among parents of children with residual hearing is difficult because they experience more uncertainty than parents of children with bilateral profound hearing loss (Burger et al., 2005; Duncan, 2009; Hyde et al., 2010). There is very limited information about the CI decision-making process and the needs of parents of children with residual hearing from both the parents' and practitioners' points of view (Porter et al., 2018). Therefore, we conducted a comprehensive study to examine the characteristics and outcomes of children with residual hearing who received CIs and to better understand the decision-making experiences and needs of families and practitioners.

Objectives of the research

Our study involved three inquiries: 1) the exploration of clinical characteristics and outcomes of children with residual hearing (Chapter 2); 2) a summary of the evidence of the benefits and risks of CIs compared to HAs in children with residual hearing (Chapter 3); and 3) the exploration of the decision-making process and needs from the perspectives of parents and practitioners (Chapters 4 and 5).

Conceptual Framework

The Ottawa Decision Support Framework (ODSF, O'Connor et al., 1998) guided this thesis project (see Figure 1). This conceptual framework depicts a family's decisional needs and how decision-making support can be used to address the family's needs to improve decision quality. This framework guided this thesis project to conceptualize decisional needs during the CI decision-making process for children with residual hearing.

The first project (Objective 1, Chapter 2) focused on exploring the clinical characteristics of children who were considered to have residual hearing (pure-tone average [PTA] < 90 dB HL). These "personal /clinical characteristics", described in the *decisional needs* component of the ODSF, are presented in Chapter 2. The second project (Objective 2, Chapter 3) involved a systematic review that synthesized the literature on CI in children with residual hearing. The findings of this review contribute to the "facts and probabilities" in the *decision support* component of ODSF. The third project (Objective 3, Chapters 4 and 5) also focused on understanding both parents' and practitioners' decisional needs including "decisional conflict, knowledge and expectations, values, support and resources, and decision: type, timing, stage, and leaning".

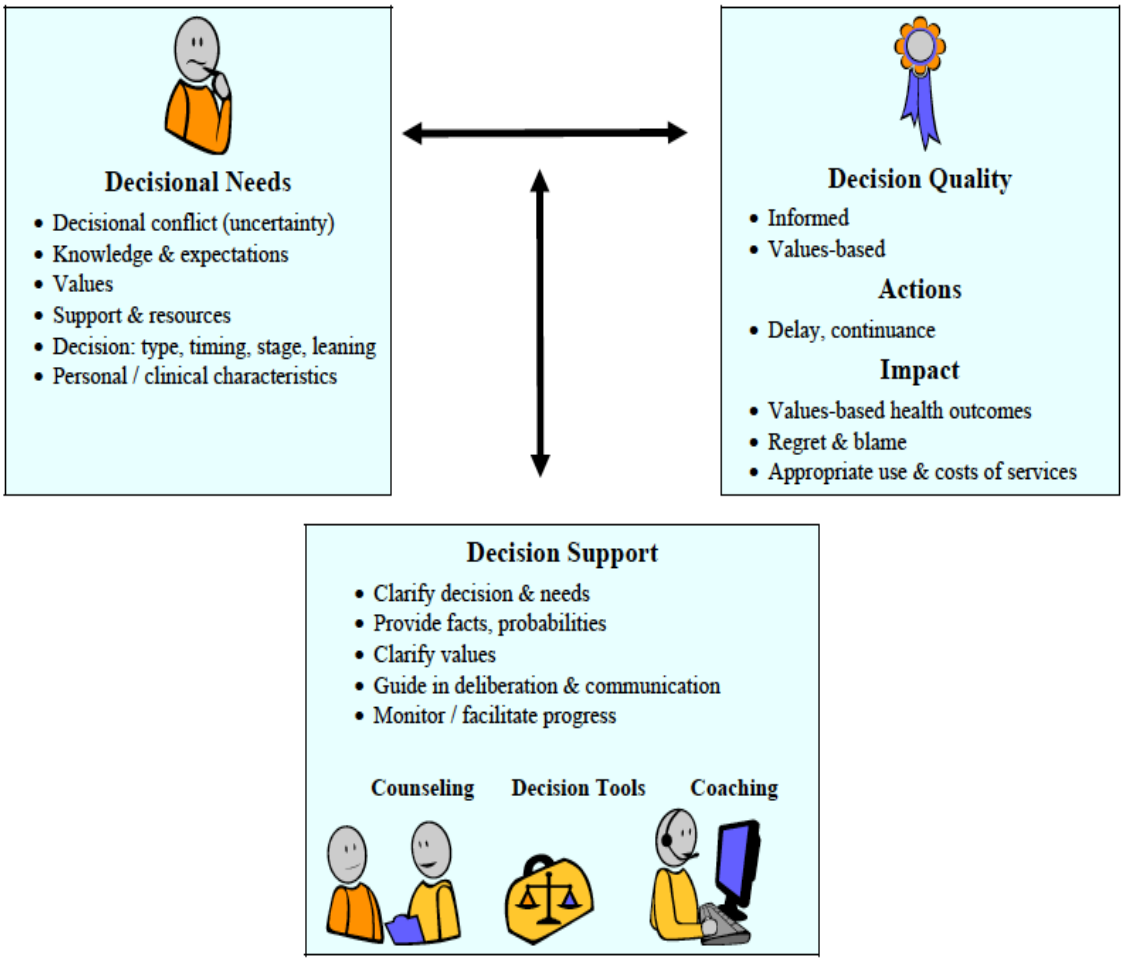


Figure 1. Ottawa Decision Support Framework

Method

A mixed-methods approach involving both qualitative and quantitative research methods was used to investigate the study objectives. A retrospective chart review was conducted to collect audiological and clinical information on children who had residual hearing before CI surgery (Chapter 2). A systematic review was completed according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) to summarize the benefits and risks related

to CIs compared to HAs for children with residual hearing (Chapter 3). Individual qualitative interviews were conducted with parents of 11 children and focus group interviews (and 1 individual interview) were carried out with 17 practitioners to understand the decision-making process and the needs of parents according to parents (Chapter 4) and practitioners (Chapter 5).

Summary of findings

Inquiry 1: Retrospective chart review

In our study, a total of 100 of 389 (25.7%) children who received CIs from 1992 to 2018 at the Children's Hospital of Eastern Ontario (CHEO) had residual hearing, representing more than half the children who were implanted in the last two years covered by the study. Children with residual hearing took longer to receive CIs compared to children with bilateral profound hearing loss (median time of 29.6 months [interquartile range-IQR: 11.8, 61.4] vs. 16.7 months [IQR: 7.8, 46.8]) ($p < 0.0001$), suggesting that decision-making may take longer due to uncertainty. Children had a median preoperative PTA threshold of 77.6 dB HL, indicating that audiometric thresholds for pediatric CI candidacy applied in clinical practice are lower than the FDA-approved standard CI candidacy criteria (PTA $\leq$ 90 dB HL at 500, 1000, 2000 Hz). As documented in our study, overall, these children showed positive auditory-related outcomes. Based on data available for 83 (83.0%) children, our findings showed that children with residual hearing demonstrated benefits in auditory functioning following cochlear implantation. Approximately 70% of these children achieved open-set word perception scores of 80% or more post-CI.

The limitation of this inquiry was that the data were collected from only one CI program. In addition, the data were limited to audiological and clinical characteristics. Other areas (e.g.,

language, social functioning, academic achievement, and listening fatigue) that are important to families and practitioners were not examined. Despite the limitations, this chart review provides comprehensive population-based information about hearing outcomes in a large cohort of children. These data can provide additional insights into the profiles of children who receive CIs. This information can assist in providing families with evidence-based information about how children can be expected to function post-implant.

Inquiry 2: Systematic review

The second inquiry involved a systematic review that included eight studies, published from 2003 to 2019, on children with residual hearing who received CIs. Children with CIs showed improvement in speech perception and other areas of auditory performance scores compared to those with HAs based on evidence from six studies of weak to moderate quality. However, there remains relatively little information about other areas of functioning. Two studies of weak quality suggested some improvement in speech intelligibility (results not statistically significant). Two aspects of social-emotional functioning (hyperactivity/inattention and pro-social behaviour) showed significant benefit from CI in one weak quality study. Our review, coupled with the findings of a previous review (Chiossi, 2017), provides reasonably good evidence for the advantages of CIs over HAs in improving speech perception and provides promising benefits in auditory performance. We had hoped to add new knowledge in the areas of language, cognitive and academic development, and quality of life; however, we found very limited information on domains beyond speech perception. Although we found information related to speech intelligibility and social-emotional functioning through the systematic review, these findings are confined to a small number of studies, making the results inconclusive. Our review included

information on the risks of CIs compared to HAs from four moderate to weak quality studies. A total of 16 of 43 children (37.2%) showed loss of residual hearing and a total of 3 of 57 (5.3%) children in these studies were reported to discontinue use of their CI device, and 5 of 57 (8.8%) children who used HAs in the non-implanted ear discontinued or only intermittently used their HA after surgery. There is extensive literature reporting the risks related to surgery for all children who undergo cochlear implantation (e.g., loss of residual hearing, surgery-specific complications, medical-specific complications, and device-related issues). However, our review provides specific information about the deterioration in hearing and discontinued or limited use of CIs for children with residual hearing.

This systematic review provides the first account to consider both the benefits and risks of CIs compared to HAs in children with residual hearing. However, our review included only studies published in English and the results of these findings cannot be globally representative because there is variation in CI candidacy criteria and regulatory requirements in different countries. In addition, there is a potential for results to be affected by publication bias because we did not include studies such as those presented at conferences. Despite these limitations, this systematic review helps to strengthen the evidence for families as they work through the process. This review confirms the benefits in speech perception and also contributes new information about the loss of residual hearing and device use, which can assist in counselling parents of children with residual hearing.

Inquiry 3: Qualitative interview

Interviews with parents

Parents reported experiencing decisional conflict when their CI practitioners first recommended

consideration of a CI. Some parents in our study tended to delay decision-making because they believed their children still received enough benefits from HAs to continue progressing in language development. The major concern for parents was the fear of their child losing residual hearing. In addition, they also worried about the surgery and expressed uncertainty about whether the benefits of speech-language outweighed the risks related to surgery. Parents also described concerns related to the long-term reliability of CI and the disadvantages of CI (e.g., restrictions around magnetic resonance imaging [MRI]). They also talked about their feelings during the decision-making process. They felt conflicted about whether a CI was the right decision and whether their children would blame them later when deciding on behalf of their young children. For the parents, the presence of additional disabilities was also an important consideration. Parents who participated in our study ultimately decided on a CI for their children, and the three major reasons were hearing deterioration, limited auditory function, and limited social integration. For these parents, inclusion into their hearing community and preservation of residual hearing were the key values. Spoken communication was a core value for these parents and some expressed high expectations that their children's hearing would be 'normal'. Overall, parents were satisfied with their decision-making process and the decision support from practitioners. However, parents stressed the importance of receiving more personalized information that took into account their specific concerns, values and preferences related to their child's and family's circumstances.

This study is the first of its kind to explore parents' perspectives and decisional needs in the CI decision-making process. Findings from this study provide valuable insights into the parents' decisional conflict, values, preferences, and decisional needs during the decision-making process for choosing CI for children with residual hearing. Although the participants of

this study were recruited from a single region in Canada and were limited to parents only, our research provides additional evidence to guide the CI decision-making process for children with residual hearing. Additional collaborative research with audiology and decision-making experts specifically on facilitating shared decision-making (SDM) is needed to provide better personalized information that could be potentially provided by decision support interventions for parents of children with residual hearing.

Interviews with practitioners

We found that CI recommendations for children with residual hearing were challenging for practitioners because of the variability of audiometric candidacy criteria. Practitioners therefore highlighted the need to consider children's functioning in multiple domains, beyond audiometric thresholds and speech perception scores. Based on their experiences, they observed that some parents declined to proceed with a CI or waited to decide because they believed that their children received sufficient benefits from HAs. Practitioners also reported that some parents had high expectations and sometimes expected that a CI could 'cure' or 'fix' the child's hearing loss. Practitioners provided realistic information and support to the parents and indicated they focused on discussing hearing preservation because they knew that surgery and loss of residual hearing were major concerns for the parents of children with residual hearing. Practitioners also talked about additional considerations for children with residual hearing who have additional medical complexities and for adolescents with residual hearing. In our discussions with practitioners about the decision-making process, we also learned that their confidence in determining candidacy and supporting parents has increased largely due to their experiences with positive outcomes for this population of children. They indicated that there was a need for more research

to guide parents' decision-making process.

This study allowed us to identify key aspects of the CI decision-making process for children with residual hearing from the perspectives of both hospital- and school-based practitioners who follow these children and families closely. Although all practitioners were recruited from a single region in Canada where children received CI within a publicly funded healthcare setting, interviews with both clinicians and specialized teachers permitted us to collect multidisciplinary perspectives, which strengthened the study. The findings of this study provide a better understanding of practitioners' experiences and are a first step in assisting in the development of optimal decision support interventions (e.g., decision aids, decision coaching) for the parents of these children.

Integrated discussion

The number of implants in children with lesser degrees of hearing loss continues to increase (Carlson et al., 2018; Teagle et al., 2019). CI practitioners need to acquire information and strategies that can be applied to coaching parents in decision-making for children with residual hearing. In our introduction chapter (Chapter 1), we found that CI clinicians may be uncomfortable recommending a CI for these children due to the variability in audiometric candidacy criteria in individual clinical practice. Although our data were collected from only one CI program, the findings of the first inquiry (Chapter 2) provided a better understanding of the clinical characteristics and outcomes of children with residual hearing who received CIs. The systematic review confirms the benefits in speech perception and also contributes new information about the loss of residual hearing and device use, which can assist in counselling parents of children with residual hearing (Chapter 3). The findings from the qualitative studies

with parents (Chapter 4) and practitioners (Chapter 5) provided a better understanding of the decision-making process and the needs of parents and practitioners as they consider CIs for their children with residual hearing.

Our retrospective chart review (Chapter 2) provided the important finding that a substantial proportion of children who received CIs in Eastern Ontario, Canada, had residual hearing. Findings from the chart review showed that children with residual hearing took longer to receive CIs compared to children with bilateral profound hearing loss. A possible explanation was revealed in our qualitative studies (Chapter 4 and Chapter 5). Both parents and practitioners indicated that some parents needed time to choose CIs because they believed their children still received substantial benefits from HAs and could continue to progress in language development. These parents mainly had concerns about the surgery and the loss of residual hearing, as well as uncertainty about whether the benefits of speech and language outweighed the risks related to surgery. Although the systematic review (Chapter 3) provided specific information that children with residual hearing may be at risk of non-use or limited use of their CIs, this information is still inconclusive. Our qualitative studies (Chapter 4 and Chapter 5) found that surgery and loss of residual hearing are important concerns for these parents; therefore, additional research about the risks of CIs is needed. The findings of the chart review (Chapter 2) also demonstrated benefits in speech perception and auditory behaviour outcomes following cochlear implantation. These findings are aligned with information from the qualitative study (Chapter 5) in which clinicians expressed confidence in recommending CIs and supporting parents of children with residual hearing because of their experiences with positive outcomes after surgery.

Our systematic review (Chapter 3) provided evidence for the advantages of CIs over HAs in improving speech perception and auditory performance. These findings help to strengthen the

evidence for families as they work through the decision-making process; however, we confirmed that there is a lack of evidence about outcomes that influence decision-making for both families and practitioners. For parents (Chapter 4), children's social functioning was an important factor in their decision-making. Practitioners (Chapter 5) also highlighted the importance of evaluating overall auditory function, listening effort and fatigue, social behaviour, and progress in therapy. These findings suggest the need for more research to explore the overall functioning (e.g. social functioning, listening effort and fatigue, and progress in therapy) of children with residual hearing beyond commonly measured outcomes.

Practitioners stressed the importance of further research (Chapter 5). Research related to outcomes that extend beyond speech perception to including benefits related to everyday functioning, more up-to-date evidence about hearing preservation, and information about the experiences of families of other children with residual hearing who received CIs may help to decrease parents' decisional conflict. The findings from the parents' interviews (Chapter 4) also suggest that more personalized information tailored to the family's specific concerns, values and preferences may help reduce the uncertainty in the decision-making process for parents of children with residual hearing. This study supports the need to incorporate parents' and practitioners' perspectives into the decision-making process. Practitioners and parents expressed similar needs, which can be used as a starting point for evidence to develop family-centered decision support interventions, such as decision aids or decision coaching for these families of children with residual hearing.

Limitations of the research

The main limitation of our study is that data and participants were recruited from a single region in Canada with a specific model of intervention. Since CI programs have different definitions and practices related to residual hearing, CI candidacy decisions and consequently the characteristics of children with residual hearing who receive CIs may differ among programs. For example, in this research, we defined usable residual hearing as an average PTA threshold ≤ 90 dB HL at 500, 1000 and 2000 kHz; however, many clinics might not consider 90 dB HL to be usable hearing. While participants in our study may experience CI decision-making differently depending on their backgrounds and cultures (Hawley & Morris, 2017; Jull et al., 2015), we did not collect culture-specific information. Moreover, despite increasing interest in supporting children's participation in health decision making, our interviews were limited to parents and practitioners.

Future research

Additional studies in decision-making about CIs for children with residual hearing are still required. In particular, to assist families in decision-making, we still need a better understanding of these children's everyday functioning in communication and their participation in classroom and social settings. From the qualitative studies (Chapter 4 and Chapter 5), we learned that parents need personalized information tailored to their specific concerns, values and preferences that can guide them within the context of their own family's circumstances. Research with families in diverse cultures can also help better inform personalized decision-making support. Furthermore, additional collaborative research with audiology and decision-making researchers is needed. Specifically, applying decision support interventions that include personalized

information in pediatric CI may help families and practitioners minimize decisional conflict as demonstrated in other areas of health care (LeBlanc et al., 2015; Légaré et al., 2018). According to previous studies, it is important to consider how decision-making might best be facilitated among multiple stakeholders (children, parents, and practitioners) and consider employing interventions that meet the needs of all those involved (Coyne, 2008; Moore & Kirk, 2010; Feenstra et al., 2015). Therefore, decision support interventions should include children's perspectives on decisions related to their own health (Coyne et al., 2014; Feenstra et al., 2014; Lipstein et al., 2015). Although interventions to support decision-making in adult health care have been well reported (Stacey et al., 2017), such research in pediatrics is sparse (Feenstra et al., 2014). Thus, inclusion of children's perspectives in future studies could add further insights into what is important for them and lead to more personalized care.

Conclusion

An important component of rehabilitation for children with hearing loss involves guiding parents in decision-making related to hearing technology. To our knowledge, this is the first study that examined decision-making for children with residual hearing in the Canadian context. Our study contributes new information about the characteristics of children receiving CIs, the potential benefits and risks for children with residual hearing, and decision-making needs from the perspectives of both families and health practitioners. The proportion of children with residual hearing who receive CIs is increasing across Canada and worldwide. Our research is a useful first step in providing evidence to understand this complex CI decision-making process for these children.

Knowledge Translation

Several knowledge translation activities took place throughout this thesis project. Two of the manuscripts (Chapter 2 and Chapter 3) have been submitted and are awaiting a decision regarding publication. The other two qualitative manuscripts (Chapter 4 and Chapter 5) will be submitted shortly. The research has been presented to parents, CI practitioners and researchers in the field at scientific conferences and meetings.

Presentations

- [Speak] "Cochlear implants for children with residual hearing: Supporting family decision-making". CHEO CI clinicians' team meeting, Ottawa, Ontario, Canada, 07 February 2021.
- [Poster] "Cochlear implantation in children with borderline hearing loss". 2018 Clinical Research Symposium, Ottawa Pediatric Rehabilitation Research Alliance (OPRRA), Ottawa, Ontario, Canada, 31 May 2018.
- [Poster] "Cochlear implantation in children outside typical candidacy criteria". 2018 Symposium in Rehabilitation Sciences, University of Ottawa, Ottawa, ON, Canada, 17 April 2018.
- [Poster] "Cochlear implantation in children with residual hearing". Canadian Academy of Audiology 2017 Conference and Exhibition, Ottawa, ON, Canada, 11-14 October 2017
- [Speak & Poster] "Cochlear implantation in children with borderline hearing loss". CI 2017 Pediatric 15th Symposium on Cochlear Implants in Children, San Francisco, California, U.S.A., 26-29 July 2017.

Publications

- Na, E., Toupin-April, K., Olds, J., Whittingham, J., & Fitzpatrick, E. M. (2021). Clinical characteristics and outcomes of children with cochlear implants who had preoperative residual hearing, International Journal of Audiology, https://doi.org/10.1080/14992027.2021.1893841
- Na, E., Toupin-April, K., Olds, J., Chen, J., & Fitzpatrick, E. M. Benefits and risks related to cochlear implantation for children with usable residual hearing: A systematic review. (Submitted) 2020
- Na, E., Fitzpatrick, E. M., Toupin-April, K., & Olds, J. Cochlear implant decision-making for children with residual hearing. Canadian Audiologist, 7(5).

References

Burger, T., Spahn, C., Richter, B., Eissele, S., Lohle, E., & Bengel, J. (2005). Parental distress: The initial phase of hearing aid and cochlear implant fitting. *Am Ann Deaf*, *150*(1), 5–10. https://doi.org/10.1353/aad.2005.0017

Carlson, M. L., O'Connell, B. P., Lohse, C. M., Driscoll, C. L., & Sweeney, A. D. (2018). Survey of the American Neurotology Society on Cochlear Implantation. *Otol Neurotol*, *39*(1), e12–e19. https://doi.org/10.1097/MAO.0000000000001631

Chiossi, J. S. C., & Hyppolito, M. A. (2017). Effects of residual hearing on cochlear implant outcomes in children: A systematic-review. *Int J Pediatr OtorhiInt J Pediatr Otorhinolaryngol*, *100*, 119–127. https://doi.org/10.1016/j.ijporl.2017.06.036

Chundu, S., & Flynn, S. L. (2014). Audiogram and cochlear implant candidacy – UK perspective. *Cochlear Implants Int*, *15*(4), 241–244. https://doi.org/10.1179/1754762813Y.0000000052

Coyne I. (2008). Children's participation in consultations and decision-making at health service level: A review of the literature. *Int J Nurs Stud*, *45*(11):1682–9.

Coyne, I., Amory, A., Kiernan, G., & Gibson, F. (2014). Children's participation in shared decision-making: Children, adolescents, parents and healthcare professionals 'perspectives and experiences. *Eur J Oncol Nurs*, *18*(3), 273–280. https://doi.org/10.1016/j.ejon.2014.01.006

de Kleijn, J. L., van Kalmthout, L. W. M., van der Vossen, M. J. B., Vonck, B. M. D., Topsakal, V., & Bruijnzeel, H. (2018). Identification of pure-tone audiologic thresholds for pediatric cochlear implant candidacy: A systematic review. *JAMA Otolaryngol Head Neck Surg*, *144*(7), 630–638. https://doi.org/10.1001/jamaoto.2018.0652

Duncan, J. (2009). Parental readiness for cochlear implant decision-making. *Cochlear Implants Int*, *10*(1), 38–42. https://doi.org/10.1002/cii.384

Feenstra, B., Lawson, M. L., Harrison, D., Boland, L., & Stacey, D. (2015). Decision coaching using the Ottawa family decision guide with parents and their children: A field testing study. *BMC Med Inform Decis Mak*, *15*(1), 5. https://doi.org/10.1186/s12911-014-0126-2

Feenstra, B., Boland, L., Lawson, M. L., Harrison, D., Kryworuchko, J., Leblanc, M., & Stacey, D. (2014). Interventions to support children's engagement in health-related decisions: A systematic review. *BMC Pediatr*, *14*(1), 109. https://doi.org/10.1186/1471-2431-14-109

Fitzpatrick, E. M., Olds, J., Durieux-Smith, A., McCrae, R., Schramm, D., & Gaboury, I. (2009). Pediatric cochlear implantation: How much hearing is too much? *Int J Audiol*, *48*(2), 91–97. https://doi.org/10.1080/14992020802516541

Hawley, S. T., & Morris, A. M. (2017). Cultural challenges to engaging patients in shared decision making. *Patient Education and Counseling*. https://doi.org/10.1016/j.pec.2016.07.008

Hyde, M., Punch, R., & Komesaroff, L. (2010). Coming to a decision about cochlear implantation: Parents making choices for their deaf children. *J Deaf Stud Deaf Educ*, *15*(2), 162–178. https://doi.org/10.1093/deafed/enq004

Jull, J., Giles, A., Lodge, M., Boyer, Y., & Stacey, D. (2015). Cultural adaptation of a shared decision-making tool with Aboriginal women: A qualitative study. *BMC Medical Informatics and Decision Making*. https://doi.org/10.1186/s12911-015-0129-7

LeBlanc, A., Herrin, J., Williams, M. D., Inselman, J. W., Branda, M. E., Shah, N. D., Heim, E. M., Dick, S. R., Linzer, M., Boehm, D. H., Dall-Winther, K. M., Matthews, M. R., Yost, K. J., Shepel, K. K., & Montori, V. M. (2015). Shared decision making for antidepressants in

primary care a cluster randomized trial. *JAMA Internal Medicine*. https://doi.org/10.1001/jamainternmed.2015.5214

Légaré, F., Adekpedjou, R., Stacey, D., Turcotte, S., Kryworuchko, J., Graham, I. D., Lyddiatt, A., Politi, M. C., Thomson, R., Elwyn, G., & Donner-Banzhoff, N. (2018). Interventions for increasing the use of shared decision making by healthcare professionals. *Cochrane Database Syst Rev*. https://doi.org/10.1002/14651858.CD006732.pub4

Lipstein, E. A., Brinkman, W. B., Fiks, A. G., Hendrix, K. S., Kryworuchko, J., Miller, V. A., Prosser, L. A., Ungar, W. J., & Fox, D. (2015). An emerging field of research: Challenges in pediatric decision making. *Med Decis Making*, *35*(5), 403–408. https://doi.org/10.1177/0272989X14546901

Moore L & Kirk S. (2010). A literature review of children's and young people's participation in decisions relating to health care. *J Clin Nurs*, *19*(15–16):2215–25.

O'Connor, A. M., Tugwell, P., Wells, G. A., Elmslie, T., Jolly, E., Hollingworth, G., McPherson, R., Bunn, H., Graham, I., & Drake, E. (1998). A decision aid for women considering hormone therapy after menopause: Decision support framework and evaluation. *Patient Educ Couns*, *33*(3), 267–279. https://doi.org/10.1016/S0738-3991(98)00026-3

Stacey, D., Légaré, F., Lewis, K., Barry, M. J., Bennett, C. L., Eden, K. B., ... & Trevena, L. (2017). Decision aids for people facing health treatment or screening decisions. *Cochrane Database Syst Rev*, (4).

Teagle, H. F. B., Park, L. R., Brown, K. D., Zdanski, C., & Pillsbury, H. C. (2019). Pediatric cochlear implantation: A quarter century in review. *Cochlear Implants Int*, *20*(6), 288–298. https://doi.org/10.1080/14670100.2019.1655868

Appendix A: Data extraction form for the chart review

Date of Birth: ___ ___/___ ___ ___/___ ___ (ALL DATES ARE DD/MMM/YY) Gender: ☐ Male ☐ Female

Date of Last Visit: ___ ___/___ ___ ___/___ ___

ROUTE TO REFERRAL		
☐ Screened	☐ Not screened (Referred)	☐ Unknown

SCREENING
Date of Screening: ___ ___/___ ___ ___/___ ___ Location: _____
Outcome: ☐ Pass ☐ Refer ☐ No Result/Incomplete ☐ Did Not Test ☐ Not Screened

HEARING LOSS DIAGNOSIS		
Date of Referral:	___ ___/___ ___ ___/___ ___	
Date of First Assessment:	___ ___/___ ___ ___/___ ___	Location: _____
Hearing Loss:	☐ Yes ☐ No ☐ Inconclusive; monitored	
Date of Hearing Loss Confirmation:	___ ___/___ ___ ___/___ ___	Location: _____

Notes on HL Diagnosis

RISK STATUS At Risk Not At Risk Not at CHEO

ETIOLOGY

Known: YES NO

*Category	*Etiology	
1	familial	_____
2	defect ENT	_____
3	syndromic	_____
4	prenatal infection	_____
5	NICU graduate	
6	meningitis	
7	middle ear disease	
8	ototoxic meds/oncology	
9	unknown	

ONSET OF HEARING LOSS		
Congenital	☐	A PHL that is confirmed after a refer result on screening
Early onset	☐	A PHL that is recognized in the neonatal period, defined for this study as including children referred and diagnosed before 6 months of age.
Late onset	☐	A PHL that is diagnosed following a screening pass and/or a least one diagnostic assessment with thresholds within normal limits
Progressive	☐	A change in audiometric thresholds after diagnosis of ≥ 20 dB in pure tone average
Unknown	☐	A permanent hearing loss (PHL) diagnosed after infancy with no history of screening, no risk factors identified and unknown etiology

PHL: Permanent hearing loss

INTERVENTION: HABILITATION TYPE

HA Fitting	Date of first HA (R)	___ ___/___ ___ ___/___ ___ ___ DAY MONTH YEAR
HA Fitting	Date of first HA (L)	___ ___/___ ___ ___/___ ___ ___ DAY MONTH YEAR

HA: Hearing aid

Cochlear Implant(s) (CI)

Unilateral ☐ Sequential ☐ Simultaneous ☐

A) Ear implanted ☐ Left ☐ Right

Location of surgery: ☐ CHEO ☐ Civic ☐ Other:_____

Date of CI: ___/___ ___ ___/___ ___

Location of Speech Processor Fitting: ☐ CHEO ☐ Civic ☐ Other centre _____

Date of activation: ___ ___/___ ___ ___/___ ___Processor Type: _____

B) Ear implanted ☐ Left ☐ Right

Location of surgery: ☐ CHEO ☐ Civic ☐ Other:_____

Date of CI Implantation: ___ ___/___ ___ ___/___ ___

Location of Speech Processor Fitting: ☐ CHEO ☐ Civic ☐ Other centre _____

Date of activation: ___ ___/___ ___ ___/___ ___Processor Type: _____

Reason for CI Delay

Audiology Evaluations

Pre-CI														
Date	Source		Click Threshold	250 Hz	500 Hz	1000 Hz	2000 Hz	3000 Hz	4000 Hz	6000 Hz	8000 Hz	SDT	SRT	Type of HL
		R												
			PTA											
		L												
			PTA											
Rx and Amplification Use														

Post CI_1y														
Date	Source		Click Threshold	250 Hz	500 Hz	1000 Hz	2000 Hz	3000 Hz	4000 Hz	6000 Hz	8000 Hz	SDT	SRT	Type of HL
		R												
			PTA											
		L												
			PTA											
Rx and Amplification Use														

Post CI _ Most recent														
Date	Source		Click Threshold	250 Hz	500 Hz	1000 Hz	2000 Hz	3000 Hz	4000 Hz	6000 Hz	8000 Hz	SDT	SRT	Type of HL
		R												
			PTA											
		L												
			PTA											
Rx and Amplification Use														

Speech Perception

Pre-CI																			
Date		dB	device	PBK_W	PBK_Ph	HINT_Q	HINT_N	MLNT_W	MLNT_Ph	ESP_P	ESP_S	ESP_M	GASP_W	GASP_S	WIPI	LittlEARs	IT_MAIS	MAIS	MUSS
	R																		
	L																		

Post CI_1y																			
Date		dB	device	PBK_W	PBK_Ph	HINT_Q	HINT_N	MLNT_W	MLNT_Ph	ESP_P	ESP_S	ESP_M	GASP_W	GASP_S	WIPI	LittlEARs	IT_MAIS	MAIS	MUSS
	R																		
	L																		

Post CI_ Most recent																			
Date		dB	device	PBK_W	PBK_Ph	HINT_Q	HINT_N	MLNT_W	MLNT_Ph	ESP_P	ESP_S	ESP_M	GASP_W	GASP_S	WIPI	LittlEARs	IT_MAIS	MAIS	MUSS
	R																		
	L																		

Appendix B: Peer Review of Electronic Strategies (PRESS) form reviewed by a librarian

PRESS Guideline — Search Submission & Peer Review Assessment

SEARCH SUBMISSION: THIS SECTION TO BE FILLED IN BY THE SEARCHER

Searcher: Eunjung Na	
Date submitted: 09 Jan 2019	Date requested by: *[Maximum = 5 working days]*

Systematic Review Title:

A systematic review of the benefits and risks related to cochlear implantation for children with usable residual hearing

This search strategy is ...

*	My PRIMARY (core) database strategy — First time submitting a strategy for search question and database
	My PRIMARY (core) strategy — Follow-up review NOT the first time submitting a strategy for search question and database. If this is a response to peer review, itemize the changes made to the review suggestions
	SECONDARY search strategy— First time submitting a strategy for search question and database
	SECONDARY search strategy — NOT the first time submitting a strategy for search question and database. If this is a response to peer review, itemize the changes made to the review suggestions

Database

(i.e., MEDLINE,CINAHL...): *[mandatory]*

MEDLINE

Interface

(i.e., Ovid, EBSCO...): *[mandatory]*

Ovid

Research Question

(Describe the purpose of the search) *[mandatory]*

To review and summarize the evidence on the benefits and risks related to cochlear implant compare to hearing aids, for children with usable residual hearing

1

Eunjung Na

PICO Format

(Outline the PICOs for your question — i.e., Patient, Intervention, Comparison, Outcome, and Study Design — as applicable)

P	Targeting population up to 18 years who have usable residual hearing (pure-tone average ≤90 dBHL)
I	Unilateral or bilateral cochlear implants
C	(1) Use of hearing aids / (2) No comparison (pre and post study)
O	(1) primary : Auditory abilities, speech perception, receptive and expressive language abilities, vocabulary, communicative competence, surgery-related complications, medical-related complications, equipment-related complications (2) secondary: academic achievement, behavioral issues, social integration, negative consequences
S	RCTs, controlled clinical trials, quasi-experimental studies, retrospective and prospective cohort studies, pre-post comparison studies

Inclusion Criteria

(List criteria such as age groups, study designs, etc., to be included) *[optional]*

Exclusion Criteria

(List criteria such as study designs, date limits, etc., to be excluded) *[optional]*

Inclusion	Exclusion
Study designs	***Study designs***
Randomized controlled trial Controlled clinical trial Quasi Randomized controlled trial Cohort study Case-control study	Case-series/case report Expert opinions
Participants	***Participants***
Age group ≤ 18 years Children who have usable residual hearing (PTA ≤ 90dBHL)	Age group > 18 years Children who have profound hearing loss (PTA 90dBHL) Children who have other disabilities
Intervention	***Intervention***
Unilateral CI Bilateral CIs	Unilateral CI and HA same ear Hearing assistive devices other than CI
Comparison	***Comparison***
HA (unilateral and bilateral) Bone conduction HA	Unilateral CI and HA same ear Hearing assistive devices other than HAs
Outcomes	***Outcomes***
Primary outcomes Benefits: Auditory abilities Sound localization Speech perception Words perception Sentence perception Speech perception in noise	Hearing preservation outcomes after CI surgery

Eunjung Na

Receptive and expressive language
Vocabulary
Communicative competence
knowing how to use grammar
respond to language appropriately
interpret context
repair communication
Risks (CI):
Surgery-related complications
anesthetic complications
cholesteatoma
cerebrospinal fluid leakage
facial nerve injury
loss of residual hearing
Risks (CI and HA):
Medical-related complications
wound infections
meningitis
vertigo
headaches
tinnitus
skin irritation, soreness
Equipment-related complications
device failure (revision surgery)
improper sound level and quality
discomfort
Negative consequences
low self-esteem
reduced quality of life
Secondary outcomes
Academic achievement
Behavioral issues
Social integration
Quality of life

Was a search filter applied?

No

If YES, which one(s) (e.g., Cochrane RCT filter, PubMed Clinical Queries filter)? Provide the source if this is a published filter. *[mandatory if YES to previous question — textbox]*

Other notes or comments you feel would be useful for the peer reviewer? ***[optional]***

Eunjung Na

Please copy and paste your search strategy here, exactly as run, including the number of hits per line. ***[mandatory]***

09 Jan 2019

1. cochlear implants/	9117
2. (cochlear adj3 implant*).tw.	13122
3. (cochlear adj3 instrument*).tw.	22
4. (auditory adj3 prosthes#s).tw.	238
5. (cochlear adj3 prosthes#s).tw.	303
6. (auditory adj3 implant*).tw.	839
7. 1 or 2 or 3 or 4 or 5 or 6	14865
8. hearing aids/	8097
9. (hearing adj3 aid*).tw.	8769
10. (hearing adj3 device*).tw.	1135
11. (hearing adj3 technolog*).tw.	319
12. (hearing adj3 instrument*).tw.	249
13. (hearing adj3 prosthes#s).tw.	162
14. (ear adj3 mold*).tw.	91
15. (ear adj3 mould*).tw.	54
16. BAHA.tw.	592
17. amplification*.tw.	124026
18. (middle ear* adj3 implant*).tw.	596
19. (middle ear* adj3 prosthes#s*).tw.	151
20. (middle ear* adj3 technolog*).tw.	8
21. (middle ear* adj3 instrument*).tw.	13
22. (middle ear* adj3 device*).tw.	112
23. 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19 or 20 or 21 or 22	135568
24. exp Child/ or adolescent/ or exp infant/	3358803
25. (infan* or newborn* or new-born* or perinat* or neonat* or baby* or babies or toddler* or minor* or boy or boys or boyhood or girl* or kid or kids or child* or schoolchild* or schoolage*).tw.	2195743
26. (adolescen* or youngster* or juvenil* or youth* or teen* or under age* or pubescen*).tw.	381077
27. exp pediatrics/ or (pediatric* or paediatric* or peadiatric*).tw.	329339
28. (prematur* or preterm*).tw.	184892
29. 24 or 25 or 26 or 27 or 28	4342277
30. (residual adj3 hear*).tw.	1197
31. (partial adj3 hear*).tw.	638
32. (partial adj3 threshold*).tw.	98
33. ((off adj3 label*) or off-label*).tw.	6656
34. ((cut adj3 off*) or cutoff* or cut-off*).tw.	95664
35. (low adj3 (frequenc* or asymmetric*)).tw.	61017
36. (hearing adj3 loss).tw.	41922
37. 30 or 31 or 32 or 33 or 34 or 35 or 36	204040
38. 7 and 23 and 29 and 37	665

Eunjung Na

(Add more space, as necessary.)

PEER REVIEW ASSESSMENT: THIS SECTION TO BE FILLED IN BY THE REVIEWER

Reviewer: Marie-Cécile Domecq	Email: mdomecq@uottawa.ca	Date completed: Jan, 16th 2019

1. TRANSLATION

A ---No revisions	X
B --- Revision(s) suggested	☐
C --- Revision(s) required	☐

If "B" or "C," please provide an explanation or example:

2. BOOLEAN AND PROXIMITY OPERATORS

A ---No revisions	X
B --- Revision(s) suggested	☐
C --- Revision(s) required	☐

If "B" or "C," please provide an explanation or example:

3. SUBJECT HEADINGS

A ---No revisions	☐
B --- Revision(s) suggested	X
C --- Revision(s) required	☐

If "B" or "C," please provide an explanation or example:

MeSH missing for the 4th concept "residual hearing" (line 30-37)
Some MeSH terms could be use: Hearing loss/ or hearing loss, sensorineural/

4. TEXT WORD SEARCHING

A ---No revisions	☐

5

Eunjung Na

B --- Revision(s)suggested	X
C --- Revision(s) required	☐

If "B" or "C," please provide an explanation or example:

Line 16 : search for the acronym BAHA
Please add a new line to search the long form of it, at least.
Line 17 = bone-anchored hearing.tw.

Simplication of search syntax : use the OR/
Ex. Line 23
23. 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19 or 20 or 21 or 22
Will become:
23. OR/8-22
Same for lines 7, 29, 37

5. SPELLING, SYNTAX, AND LINE NUMBERS

A ---No revisions	☐
B --- Revision(s)suggested	X
C --- Revision(s) required	☐

If "B" or "C," please provide an explanation or example:

Eunjung Na

Simplication of search syntax :
Lines 4,5,13,19 : uses prosthes* in your syntax, for ex. (auditory adj3 prosthes*).tw
Line 34: off* change for off?

Simplication of search syntax :

6. LIMITS AND FILTERS

A ---No revisions	X
B --- Revision(s) suggested	
C --- Revision(s) required	

If "B" or "C," please provide an explanation or example:

OVERALL EVALUATION (Note: If one or more "revision required" is noted above, the response below must be "revisions required".)

A ---No revisions	
B --- Revision(s) suggested	X
C --- Revision(s) required	

Additional comments:

Bravo, Eunjung, very good search strategy !!

Appendix C: Search Strategy- Medline

1. cochlear implants/
2. (cochlear adj3 implant*).tw.
3. (cochlear adj3 instrument*).tw.
4. (auditory adj3 prosthes*).tw.
5. (cochlear adj3 prosthes*).tw.
6. (auditory adj3 implant*).tw.
7. or/1-6
8. hearing aids/
9. (hearing adj3 aid*).tw.
10. (hearing adj3 device*).tw.
11. (hearing adj3 technolog*).tw.
12. (hearing adj3 instrument*).tw.
13. (hearing adj3 prosthes*).tw.
14. (ear adj3 mold*).tw.
15. (ear adj3 mould*).tw.
16. BAHA.tw.
17. Bone anchored hearing aid*.tw.
18. amplification*.tw.
19. (middle ear* adj3 implant*).tw.
20. (middle ear* adj3 prosthes*).tw.
21. (middle ear* adj3 technolog*).tw.
22. (middle ear* adj3 instrument*).tw.
23. (middle ear* adj3 device*).tw.
24. or/8-23
25. exp Child/ or adolescent/ or exp infant/
26. (infan* or newborn* or new-born* or perinat* or neonat* or baby* or babies or toddler* or minor* or boy or boys or boyhood or girl* or kid or kids or child* or schoolchild* or schoolage*).tw.
27. (adolescen* or youngster* or juvenil* or youth* or teen* or under age* or pubescen*).tw.
28. exp pediatrics/ or (pediatric* or paediatric* or peadiatric*).tw.
29. (prematur* or preterm*).tw.
30. or/25-29
31. exp hearing loss/ or hearing loss, sensorineural/
32. (residual adj3 hear*).tw.
33. (partial adj3 hear*).tw.
34. (partial adj3 threshold*).tw.
35. ((off adj3 label*) or off-label*).tw.
36. ((cut adj3 off) or cutoff or cut-off).tw.
37. (low adj3 (frequenc* or asymmetric*)).tw.
38. or/31-37
39. 7 and 24 and 30 and 38

Appendix D: Data extraction form for the systematic review

DATA EXTRACTION FORM

Study Characteristics

1. Reference number
2. Year of publication
3. First author
4. Title
5. Journal
6. Country in which the study was conducted
7. Funding source type

Study data

General

1. Study design
2. Sample size
3. Quality assessment
4. Study objective(s)
5. Definition of residual hearing

Population Characteristics

1. Clinical setting
2. Gender
3. Age at study
4. Age at CI
5. Age of identification
6. Severity of hearing loss before CI (unaided PTA)

Intervention

1. Types of interventions
2. Number of children with CI

Comparators

1. Types of comparators (HAs or Pre CI)
2. Audiological information

Outcomes

1. Speech perception (or speech recognition)
2. Auditory comprehension measures
3. Receptive language
4. Expressive language
5. Speech outcomes

6. Social communication skills
7. Phonological related skills
8. Academic achievement
9. Behavioral issues,
10. Social integration
11. Surgery-related complications
12. Medical-related complications
13. Equipment-related complications
14. Psychological related complications

Appendix E: Research Ethics Board certificate

University of Ottawa

Université d'Ottawa University of Ottawa

Bureau d'éthique et d'intégrité de la recherche Office of Research Ethics and Integrity

June 26, 2017

Elizabeth Fitzpatrick
Professor
Faculty of Health Sciences
University of Ottawa
elizabeth.fitzpatrick@uottawa.ca

Co-Investigators: Eunjung Na, CHEO
JoAnne Whittingham, CHEO (Coordinator)

Re: U of O Ethics file no. A06-17-05 – "Rationale for developing a decision support tool for children outside typical cochlear implant candidacy criteria"

Dear Professor Fitzpatrick and colleagues,

Thank you for the protocol documents and Certificates of Approval from the CHEO REB (# 17/100X) for your project named above.

This is to confirm that, in accordance with the agreement between the University of Ottawa and CHEO REB, the University of Ottawa has authorized this board to act as Board of Record for the review and oversight of research involving human subjects conducted at or through the hospital.

We remind you of your obligation to:

- Follow all procedures of the CHEO REB including reporting and renewal procedures;
- Submit to the authority of the CHEO REB and that you are subject to CHEO REB requirements, including, without limitation, the requirement to modify or stop the research on demand of the CHEO REB.

If you have any questions, please contact our ethics office at 562-5387.

Sincerely yours,

Office of Research Ethics and Integrity

550, rue Cumberland 550 Cumberland Street
Ottawa (Ontario) K1N 6N5 Canada Ottawa, Ontario K1N 6N5 Canada

(613) 562-5387 • Téléc./Fax (613) 562-5338
http://www.recherche.uottawa.ca/deontologie/
http://www.research.uottawa.ca/ethics/

Children's Hospital of Eastern Ontario (CHEO)

Whittingham, JoAnne

From:
Sent:
To:
Cc:
Subject:

CHEO

Research Ethics Board
Annual Renewal Approval Letter

Principal Investigator: Dr. Elizabeth Fitzpatrick

REB Protocol No: 17/100X
Romeo File No: 20170297
Project Title: 17/100X - Rationale for developing a decision support tool for children outside typical cochlear implant candidacy criteria
Primary Affiliation: Clinical Research\Audiology
Protocol Status: Active
Approval Date: May 2, 2019
Approval Expiry Date: May 15, 2020

This is to notify you that the CHEO REB has granted approval to the renewal for the above named research study for a period of one year. The renewal was reviewed in the delegated stream and approved by the Chair or a delegate of the Chair. Decisions made by the Chair under delegated review are ratified by the full Board at its subsequent meeting.

Approval is granted with the understanding that the investigator agrees to comply with the following requirements:

1. The investigator must conduct the study in compliance with the protocol and any additional conditions set out by the Board.
2. The investigator is responsible for complying with all applicable guidelines and regulations regarding the ethical conduct of research with humans, as applicable to the research project.
3. Investigators must obtain annual renewal approval prior to the expiry date stated above.
4. The investigator must not implement any deviation from, or changes to, the protocol without the approval of the REB except where necessary to eliminate an immediate hazard to the research subject, or when the change involves only logistical or administrative aspects of the study (e.g., change of telephone number or research staff). As soon as possible, however, the implemented deviation or change, the reasons for it and, if appropriate, the proposed protocol amendment(s) should be submitted to the Board for review and approval.
5. The investigator must, prior to use, obtain approval from the Board for changes to the study documentation, e.g., changes to the informed consent letters, recruitment materials.

6. Investigators must obtain approval from the Board of French version(s) of the consent/assent form(s), unless a waiver has been granted. An interpreter should be offered to participants as required or at the request of the participant throughout the course of research.
7. For clinical drug or device trials, investigators must promptly report to the REB all adverse events that are both serious and unexpected (SAEs) or unexpected and untoward occurrences (including the loss or theft of study data and other such privacy breaches.
8. For SAE reports on clinical drug trials, the investigator must also comply with the hospital-wide Policy regarding, Procedures for Considering Medical Error in the Differential Diagnosis of Severe Adverse Events (SAE) Associated with the Drugs Administered in a Clinical Trial.
9. Investigators must promptly report to the REB any new information regarding the safety of research subjects (e.g., changes to the product monograph or investigator's brochure of drug trials). Where available, any reports produced by the Data Safety Monitoring Board should also be promptly submitted to the REB for acknowledgement.
10. Investigators must notify the REB of any study closures (closed to accrual, temporary, premature or permanent).
11. Investigators must submit a study closure event form at the conclusion of the study.

If you have any questions, pertaining to this letter, please contact the Research Ethics Board Office at

Regards,

Chair, Research Ethics Board
Président, Comité d'éthique de la recherche

CHEO Research Ethics Board Approval - Delegated Review

Principal Investigator: Dr. Elizabeth Fitzpatrick
REB Protocol No: 18/129X
Romeo File No: 20180498
Project Title: CEHOREB# 18/129X - Cochlear implants for children with residual hearing: Decisional support needs of parents and children
Primary Affiliation: Clinical Research\Audiology
Protocol Status: Active
Approval Date*: November 20, 2018
Approval Expiry Date:** November 15, 2019

Documents Reviewed & Approved:

Document Name	Comments	Version Date
Recruitment Materials	Clinician information letter	11/14/2018
Consent Form	Educational providers consent form clean copy	11/14/2018
Consent Form	Clinicians consent form clean copy	11/14/2018
Consent Form	Parents consent form clean copy	11/14/2018
Recruitment Materials	Educational providers recruitment poster	11/14/2018
Recruitment Materials	Parents and children recruitment poster	11/14/2018
Protocol	Protocol clean copy	11/19/2018
Consent Form	Participant Consent	10/29/2018
Assent Form	child assent form	10/12/2018
Recruitment Materials	Information Letter for Educational Providers	10/29/2018
Investigator Response	Letter responding to itemized REB feedback, signed by the local principal investigator	10/16/2018
Other Document	Interview guide	8/29/2018
Other Document	Home visit safety plan	10/9/2018

This is to notify you that the Children's Hospital of Eastern Ontario Research Ethics Board has granted approval to the above named research study on the date noted above. Your project was reviewed within the delegated stream, which is reserved for projects that involve no more than minimal risk to human participants.

Final approval is granted for the above noted study, with the understanding that the investigator agrees to comply with the following requirements:

1. The investigator must conduct the study in compliance with the protocol and any additional

conditions set out by the Board.

2. The investigator is responsible for complying with all applicable guidelines and regulations regarding the ethical conduct of research with humans, as applicable to the research project.
3. Approval for studies that include an investigational device(s) is contingent upon the investigator securing an Investigational Testing Authorization notice from Health Canada.
4. Investigators must obtain annual renewal approval prior to the expiration date stated above.
5. The investigator must not implement any deviation from, or changes to, the protocol, consents or assents without the approval of the REB except where necessary to eliminate hazard to the research subject, or when the change involves only logistical or administrative aspects of the study (e.g., change of telephone number or research staff). As soon as possible, however, the implemented deviation or change, the reasons for it, and, if appropriate, the proposed protocol amendment(s) should be submitted to the Board for review and approval.
6. The investigator must, prior to use, obtain approval from the Board for changes to the study documentation, e.g., changes to the informed consent letters, recruitment materials.
7. Investigators must obtain approval from the Board of French version(s) of the consent/assent form(s), unless a waiver has been granted. An interpreter should be offered to participants as required or at the request of the participant throughout the course of research.
8. The investigator must promptly report to the REB all unexpected and untoward occurrences (including the loss or theft of study data and other such privacy breaches).
9. Investigators must notify the REB of any study closures (closed to accrual, temporary, premature or permanent).
10. Investigators must submit a study closure event form at the conclusion of the study.

Should you have any questions or concerns, please do not hesitate to contact the Research Ethics Board Office.

Chair, Research Ethics Board
Président, Comité d'éthique de la recherche

* The final approval date for initial delegated study applications approved with or without modifications will be the date the REB has determined that the conditions of approval have been satisfied.

** The expiry date of REB approval for initial study applications will be as follows:

- If the date of approval was **on or before** the 15th of the month, the expiry date will be the 15th of the month prior to the date of review and approval by the Chair and/or delegate *in the following year*;
- If the date of review and approval was **after** the 15th of the month, the expiry date will be the 15th of the month in which the date of review and approval by the REB *in the following year*.

Ottawa-Carleton Research and Evaluation Advisory Committee (OCREAC)

Ottawa-Carleton Research and Evaluation Advisory Committee (OCREAC)

November 21, 2018

Eunjung Na
Roger Guindon Hall
University of Ottawa
451 Smyth Rd.
Ottawa, Ontario
K1H 8M5

Re: Cochlear implants for children with residual hearing: Decisional support needs of parents and children

Dear Ms. Na,

The Ottawa-Carleton Research and Evaluation Advisory Committee has reviewed your application for an extension to your study and is granting you **approval** to conduct your study in **the 2018-2019 academic year**. This approval is for the Ottawa Catholic School Board and the Ottawa-Carleton District School Board.

To facilitate contact with itinerant teachers serving students who are deaf or hard of hearing, please contact me for the Ottawa Catholic School Board and please contact Katherine Magner for the Ottawa-Carleton DSB.

We thank you for approaching the Ottawa-Carleton area school boards as a venue for your study and we look forward to receiving a copy of your results.

Sincerely,

On behalf of the Ottawa-Carleton Research and Evaluation Advisory Committee

Ottawa Catholic School Board
570 Hunt Club Road West ▪ Nepean ▪ Ontario ▪ K2G 3R4

Ottawa-Carleton District School Board
133 Greenbank Road ▪ Nepean ▪ Ontario ▪ K2H 6L3

Appendix F: Interview guide

Interview guide questions for parents

Interview guide questions for Parents/Caregivers

1. Tell me about how you made the decision for a cochlear implant(s) for your child. What factors were important for you to consider when making the decision to get a cochlear implant for your child rather than continuing with hearing aid use (*e.g.* speech-language development, hearing at school, less difficulty in social situations)?

2. How did you feel when you were making the decision about continuing with hearing aids or going forward with a cochlear implant (s); were there things that you were worried about?

    a. What worried you most?

3. Tell me about the process of discussing the options with the practitioners, for example with the audiologist, the listening and spoken language specialist (AVT therapist), and the cochlear implant surgeon, with the itinerant teacher?

4. What kind of information did you have when making the decision about a cochlear implant(s)? Where did you find the information? (e.g. internet, books/articles, practitioners, other parents). Did you have enough information?

    a. Was it clear? Did it fit with your child?

5. Were there parts of the decision-making that were difficult for you? For example, were you missing information about options, or about benefits and risks of cochlear implantation?

6. What other kinds of information would be helpful in guiding you to make a decision about a cochlear implant(s)? What kind of tools should be developed to help you facilitate this discussion?

7. Tell me about your expectations for the CI.

8. Tell me about your child's (and your) experience with your CI. Does it help?

    a. Give me some examples
    b. Are their things you did not / do not like about the CI?

9. If you have the opportunity to make the decision again in the future, how would you like to be involved in the process? Are there things you would like to see changed? What other kinds of information would be helpful in making a decision about a cochlear implant (s)?

10. What recommendations do you have for other families going through this process?

11. What recommendations do you have for the providers and/or teachers for parents going through this process?

Interview guide questions for practitioners

Interview Guide for Clinicians & Itinerant Teachers

1. In your experience, what factors were important for families to consider when making the decision to get a cochlear implant for their child with borderline hearing rather than continuing with hearing aid use?

    a. For example, speech-language development? Better hearing function at school? Or less difficulty in social situations?

2. What issues are of most concern to you when considering the need to go forward with a cochlear implant?

    a. For example, losing residual hearing? Surgical complications? Or device failure?

3. Can you describe your role in helping families make this decision? Have you recommended that a child with residual hearing receive (or not receive) a cochlear implant?

    a. At what point did you bring it up?
    b. Had the parent ever mentioned it?

4. What prompted you to discuss a cochlear implant(s) with families? Do you think that you had enough information to help you guide parents?

5. Tell me about the process of discussing the options with the parents and child?

6. Did you discuss the options with other practitioners? Tell me about the process of discussing the options with other practitioners (e.g. cochlear implant team, (other) itinerant teachers, classroom teachers),

7. Were any aspects difficult for you when discussing this decision with families? Which ones? How difficult was it?

    a. Were you concerned about the parents being upset/worried?

8. If you have the opportunity to guide decision-making with other families in the future, how would you like to be involved in the process? Are there things you would like to see changed?

    a. For example, would you like to be involved in the meetings at the hospital?

9. What other kinds of information would be helpful in guiding families to make a decision about a cochlear implant(s)? What kind of tools should be developed to help you facilitate this discussion?

10. What recommendations do you have for other practitioners (*e.g.* other healthcare providers, other teachers) going through this process?

Appendix G: Informed consent

Informed consent - parents

CHEO

uOttawa

Cochlear implants for children with residual hearing: Decisional support needs of parents and children

INFORMATION LETTER / CONSENT FORM (Parents)

We are inviting you to participate in this project because your child has received a cochlear implant(s) at CHEO. The purpose of this project is to learn about how families make the decision to get a cochlear implant for children with usable residual hearing. We will gather the perspectives of children and youth, parents and practitioners. Researchers at the University of Ottawa and the Children's Hospital of Eastern Ontario Research Institute (CHEO) are conducting this project.

Information

Hearing loss can affect speech and language development, academic achievement, and psycho-social well-being. We know it is important for children with hearing loss to receive appropriate interventions as early as possible. Children with severe to profound bilateral sensorineural hearing loss may receive little benefit from hearing aids. In these cases, cochlear implants are recommended.

Cochlear implant decision-making for children with residual hearing, who have the ability to hear some sounds and already benefit from hearing aid use, can be more difficult than for children with profound hearing loss. Families may be unclear about the potential benefits and the risks following cochlear implantation. In addition, there is no clear cut-off hearing level for cochlear implant surgery. For these reasons, parents and practitioners may not be sure cochlear implantation is the best option for children with residual hearing.

There is a lack of good information on what supports are the most helpful to families when they are making this decision. It is important to explore the views of parents, children, youth and practitioners about what supports would be helpful when making a decision about cochlear implantation.

The results of this study will help us find out if parents of children with usable residual hearing have difficulty when making a decision about cochlear implantation. This will help us to agree on the kind of supports that may help children and their families when they are facing this decision.

At CHEO we expect to interview 10-12 parents. As well, we will interview 10-12 children who are old enough to participate in an interview. We will also interview audiologists, auditory-verbal therapists and doctors at CHEO. Also we will interview 5-6 itinerant teachers. The study is expected to be recruiting from October 2018 to September 2019.

Procedures

If you agree to participant, we will ask you to take part in an interview. The interview done with you and/or your child, will take about 40 to 60 minutes. The interviews will be audio-recorded and transcribed. The interview will typically be scheduled during the day and can be done by phone or video conference. If you prefer to interview in person, then we can schedule a time and place that is convenient for you.

The questions for the interview will ask about your experience in how you decided on cochlear implant surgery for your child, the motivation for cochlear implantation, what your expectations were, and what you think about the results. We will also ask you information about your family and about your child's hearing loss.

In addition, we will access information from your child's medical record including a description of your child's audiogram, information on the age of diagnosis and any relevant medical information.

Benefits, Risks and Inconvenience

Benefits

If you decide to participate, you may or may not benefit from participating in this study; however, we hope that the data gathered as part of this study will help us provide better support to parents who are considering a cochlear implant for their child.

Risks/Inconvenience

We know of no harm or risk that taking part in this study could cause you. A potential discomfort may include you feeling uncomfortable with some of the questions being asked if they are sensitive or evocative. If you feel uncomfortable, you may choose not to answer a question.

Will I be paid to participate?

You will not be paid to take part in this study.

Can I withdraw?

You can withdraw from the study at any time without any impact to your family's future care at CHEO. Please discuss with your investigator if you would like to withdraw. If you withdraw your consent, the investigator will no longer collect and disclose your information for the purpose of this study. Information that was already collected, as part of the interviews, will still be used by the research team to summarize how parents made the decision for their child to get a cochlear implant.

Will I be told about new information?

We will inform you of any new information that might change your decision to continue to participate in this research project. We will ask you again if you still want to be in the study. You can receive a copy of the study results at the end of the study. Please let the study team know if you like to receive a copy.

Confidentiality

The information from the interview will be used to describe how families with children with usable residual hearing made the decision to get a cochlear implant(s) and what information was the most helpful to them. In addition, we will describe what other supports families would find useful. All information will be kept strictly confidential. Your identity will not be disclosed to any person, except as required or permitted by law. Your name will not be identified in any publication, report or presentation. Comments from the interviews may be quoted but you will not be identified.

For this study we will be collecting some personal identifiers for the research purposes described in this consent form. Your/your child's name and CHEO medical record number, as well as your contact information (parent name, telephone number, and email address) will be kept in a document that links this information with a study ID, called a master list. The study ID will be used in all of the research documents to protect your privacy. The master list will be stored separately from the research data. It will be stored with password protection on a restricted computer securely stored in the Child Hearing Lab at the CHEO Research Institute with access restricted to the researcher, and the research team.

Representatives from the CHEO research Ethics Board and a member of the CHEO RI Quality Assurance and Risk Program may review your child's medical records, and the research data under the supervision of the Investigator and staff, to ensure that all research standards, guidelines and regulations are met.

Your child's full date of birth will be included in the research data along with the study ID so that we can calculate age at diagnosis of hearing loss and age at the time of the cochlear implant surgery. The research data and audio recordings produced from this study will be stored with password protection on a restricted computer in the Child Hearing Lab at the Children's Hospital of Eastern Ontario Research Institute. Only members of the research team and the individuals described above will have access to the data. Following

completion of the study the research data, audio recordings and master list will be kept for 7 years after the last publication of this study. They will then be destroyed. You will not be identified in any publication or presentation of this study.

Is the research team benefiting from the study?

The research team members are not benefiting personally, financially or in any other way from this study.

Alternatives to participation

There is no requirement to participate in this study, and your decision to participate, or not, will in no way affect the care received by your child at CHEO. If you choose to participate, you may withdraw from this study at any time for any reason. At any time during the study, you may decline to stop an interview.

What if I have questions?

If you have any questions concerning participation in this study, contact: Eunjung Na or JoAnne Whittingham,

This study has been reviewed and approved by the CHEO Research Ethics Board (REB). The CHEO REB is a committee of the hospital that includes individuals from different professional backgrounds. The Board reviews all human research that takes place at the hospital. Its goal is to ensure the safety of people taking part in research. The Board's work is not meant to replace a parent or child's decisions and choices that are best for them. You may contact the REB, for information regarding a patient's rights in research studies at although the REB cannot provide any health-related information about the study.

Consent Form Signatures

By signing this consent form I agree that:

- I am voluntarily agreeing to participate in this research study;
- I understand the information within this consent form;
- All of the risks and benefits of participation have been explained to me;
- All of my questions have been answered;
- I allow access to my child's medical records and/or personal information as described in this consent form; and
- I do not give up my legal rights by signing this form.

A copy of the signed Information Sheet and/or Consent Form will be provided to me.

Signatures

Printed Participant's Name	Participant's Signature	Date
Printed Parent's Name (If consenting on the participant/child's behalf)	Parental Signature	Date
Printed Name of Person Who Conducted Consent Discussion	Signature of Person Who Conducted Consent Discussion	Date

Research Team:

Eunjung Na, M.Sc. / Ph.D. Candidate
Faculty of Health Sciences, University of Ottawa

Elizabeth Fitzpatrick, PhD, AUD(C) / Thesis Supervisor
Faculty of Health Sciences, University of Ottawa,
Investigator, CHEO Research Institute

Karine Toupin-April, PhD / Thesis Co-Supervisor
Faculty of Medicine, University of Ottawa,
Faculty of Health Sciences, University of Ottawa,
CHEO Research Institute

JoAnne Whittingham, M.Sc. / Research Coordinator
CHEO Research Institute

Informed consent - Clinicians

CHEO

uOttawa

Cochlear implants for children with residual hearing: Decisional support needs of parents and children

INFORMATION LETTER / CONSENT FORM (Clinicians)

Principal Investigator: Elizabeth Fitzpatrick, Ph.D., Thesis Supervisor
Co-investigators: Karine Toupin-April, Ph.D., Co-supervisor,
Eunjung Na, M.Sc., Ph.D. Candidate
Research Coordinator: JoAnne Whittingham, M.Sc.

Dr. Elizabeth Fitzpatrick
Children's Hospital of Eastern Ontario Research Institute – R1133
401 Smyth Road
Ottawa, Ontario
K1H 8L1

We are inviting you to participate in this research study because you work with children who have cochlear implants. The purpose of this project is to learn about how families make the decision to get a cochlear implant for children with usable residual hearing rather than continuing with hearing aids. We will gather the perspectives of children and youth, parents and CHEO practitioners and educators. Researchers at the University of Ottawa and the Children's Hospital of Eastern Ontario Research Institute (CHEO) are conducting this research project.

Before agreeing to take part in this study, it is important that you read and understand this document.

Why is this study being done?

Cochlear implant decision-making for children with residual hearing, who have ability to hear some sounds and benefit from hearing aid use, can be more difficult than for children with profound hearing loss. Families may be unclear about the potential benefits and the risks following cochlear implantation. In addition, there is no clear cut-off hearing level for cochlear implant surgery. For these reasons, parents and practitioners may not be sure cochlear implantation is the best option for children with residual hearing.

There is a lack of good information on what supports are the most helpful to families when they are making this decision. It is important, therefore, to explore the views of parents, children, youth and practitioners about what supports would be helpful when making a decision about cochlear implantation.

At CHEO, we expect to have 8-10 clinic staff (audiologists, CI surgeons, and therapists). We will also interview 10 to 12 parents as well as 10-12 children. We also hope to have 5-6 educational providers (itinerant teachers), through the parents and the school board, participate in individual or group interviews. Recruitment for the study will be done from October 2018 to September 2019.

Procedures

If you agree to participate, we will ask you to take part in an interview. The interview will take about 40 to 60 minutes. The interview will be scheduled during the day and will be done in person. We can schedule a time that works for you. The questions for the interview will ask about your experiences supporting families who are considering cochlear implant surgery for their child, from the point of the decision-making process.

Benefits, Risks and Inconvenience

Benefits

If you decide to participate, you may or may not benefit from participating in this study; however, we hope the data gathered will lead to better evidence-based support to families in the future.

Risks/Inconvenience

We know of no harm or risk that taking part in this study could cause you. A potential discomfort may include you feeling uncomfortable with some of the questions being asked if they are sensitive or evocative. If you feel uncomfortable, you may choose not to answer a question.

Will I be paid to participate?

You will not be paid to take part in this study.

Can I withdraw?

You can withdraw from the study at any time. Please discuss with the investigator if you would like to withdraw. If you withdraw your consent, the investigator will no longer collect your data. Information that was already collected will still be used by the investigator and research team.

Confidentiality

The information from the interview will be used to describe how families with children with usable residual hearing made the decision to get a cochlear implant(s) and what information was the most helpful to them. In addition, we will describe what other supports families would find useful. All information will be kept strictly confidential. Your identity will not be disclosed to any person, except as required or permitted by law. Your name will not be identified in any publication, report or presentation. Comments from the interviews may be quoted but you will not be identified.

For this study we will be collecting personal identifiers (full name, professional role, telephone number, and email address) for the research purposes described in this consent form. Your collected personal identifiers will be kept in a document that links this information with a study ID, called a master list. The study ID will be used in all of the research documents to protect your privacy. The master list will be stored separately from the research data. It will be stored with password protection on a restricted computer securely stored in the Child Hearing Lab at the CHEO Research Institute with access restricted to the researcher, and the research team.

Your professional role will be included in the research data along with the study ID. The research data and audio recordings produced from this study will be stored with password protection on a restricted computer in the Child Hearing Lab at the Children's Hospital of Eastern Ontario Research Institute. Members of the research team and the individuals described above will have access to the data. Following completion of the study the research data, audio recordings and master list will be kept for 7 years after the last publication of this study. They will then be destroyed. You will not be identified in any publication or presentation of this study.

Is the research team benefiting from the study?

The research team members are not benefiting personally, financially or in some other way from this study.

Alternatives to participation

Taking part in this study is voluntary. Your decision to participate or not in this study will not affect your employment at CHEO. You are free to withdraw from the study at any time and there will be no penalty to you. At any time during the interview, you may decline to answer a question or discontinue the interview.

What if I have questions?

If you have any questions concerning participation in this study, contact Eunjung Na or JoAnne Whittingham,

This study has been reviewed and approved by the CHEO Research Ethics Board (REB). The CHEO REB is a committee of the hospital that includes individuals from different professional backgrounds. The Board reviews all human research that takes place at the hospital. Its goal is to ensure the safety of people taking part in research. The Board's work is not meant to replace a parent or child's decisions and choices that are best for them.

You may contact the REB, for information regarding a patient's rights in research studies at although the REB cannot provide any health-related information about the study.

Consent form Signatures

By signing this consent form I agree that:

- I am voluntarily agreeing to participate in this research study;
- I understand the information within this consent form;
- All of the risks and benefits of participation have been explained to me;
- All of my questions have been answered;
- I do not give up my legal rights by signing this form.

A copy of the signed Information Sheet and/or Consent Form will be provided to me.

Signatures

Printed Participant's Name	Participant's Signature	Date
Printed Name of Person Who Conducted Consent Discussion	Signature of Person Who Conducted Consent Discussion	Date

Research Team:

Eunjung Na, M.Sc. / Ph.D. Candidate
Faculty of Health Sciences, University of Ottawa

Elizabeth Fitzpatrick, PhD, AUD(C) / Thesis Supervisor
Faculty of Health Sciences, University of Ottawa,
Investigator, CHEO Research Institute

Karine Toupin-April, PhD / Thesis Co-Supervisor
Faculty of Medicine, University of Ottawa,
Faculty of Health Sciences, University of Ottawa,
CHEO Research Institute

JoAnne Whittingham, M.Sc. / Research Coordinator
CHEO Research Institute

Informed consent - Teachers

CHEO

uOttawa

Cochlear implants for children with residual hearing: Decisional support needs of parents and children

INFORMATION LETTER / CONSENT FORM (Educational Providers)

Eunjung Na, M.Sc., Ph.D. Candidate
University of Ottawa, Roger Guindon Hall, 451 Smyth Road, Ottawa, ON K1H8M5
Institution: University of Ottawa / Children's Hospital of Eastern Ontario Research Institute

Elizabeth Fitzpatrick, Ph.D. / Thesis Supervisor
Senior Scientist / Full Professor
Children's Hospital of Eastern Ontario Research Institute / School of Rehabilitation Sciences, Faculty of Health Sciences

Karine Toupin-April, Ph.D. / Thesis Co-supervisor
Assistant Professor / Adjunct Professor / Associate Scientist
Department of Pediatrics, Faculty of Medicine / School of Rehabilitation Sciences, Faculty of Health Sciences / Children's Hospital of Eastern Ontario Research Institute

Research Coordinator: JoAnne Whittingham, M.Sc.

We are inviting you to participate in this research study because you work with children who have cochlear implants. The purpose of this project is to learn about how families make the decision to get a cochlear implant for children with usable residual hearing rather than continuing with hearing aids. We will gather the perspectives of children and youth, parents and CHEO practitioners and educators. Researchers at the University of Ottawa and the Children's Hospital of Eastern Ontario Research Institute (CHEO) are conducting this research project.

Before agreeing to take part in this study, it is important that you read and understand this document.

Why is this study being done?

Cochlear implant decision-making for children with residual hearing, who have ability to hear some sounds and benefit from hearing aid use, can be more difficult than for children with profound hearing loss. Families may be unclear about the potential benefits and the risks following cochlear implantation. In addition, there is no clear cut-off hearing level for cochlear implant surgery. For these reasons, parents and practitioners may not be sure cochlear implantation is the best option for children with residual hearing.

There is a lack of good information on what supports are the most helpful to families when they are

making this decision. It is important, therefore to explore the views of parents, children, youth and practitioners about what supports would be helpful when making a decision about cochlear implantation.

At CHEO we expect to interview 10-12 parents. As well, we will interview 10-12 children who are old enough to participate in an interview. We will also interview audiologists, auditory-verbal therapists and doctors at CHEO. Also we will interview 5-6 itinerant teachers. The study is expected to be recruiting from October 2018 to September 2019.

Procedures

If you agree to participate, we will ask you to take part in an interview. The interview will take about 40 to 60 minutes. The interview will be scheduled during the day and will be done in person. We can schedule a time that works for you. The questions for the interview will ask about your experiences supporting families who are considering cochlear implant surgery for their child, from the point of the decision-making process.

Benefits, Risks and Inconvenience

Benefits

If you decide to participate, you may or may not benefit from participating in this study; however, we hope that the data gathered will lead to better evidence based support to families in the future.

Risks/Inconvenience

We know of no harm or risk that taking part in this study could cause you. A potential discomfort may include you feeling uncomfortable with some of the questions being asked if they are sensitive or evocative. If you feel uncomfortable, you may choose not to answer a question.

Will I be paid to participate?

You will not be paid to take part in this study.

Can I withdraw?

You can withdraw from the study at any time. Please discuss with the investigator if you would like to withdraw. If you withdraw your consent, the investigator will no longer collect your data. Information that was already collected will still be used by the investigator and research team.

Confidentiality

The information from the interview will be used to describe how families with children with usable residual hearing made the decision to get a cochlear implant(s) and what information was the most helpful to them. In addition, we will describe what other supports families would find useful. All information will be kept strictly confidential. Your identity will not be disclosed to any person, except as required or permitted by law. Your name will not be identified in any publication, report or presentation. Comments from the interviews may be quoted but you will not be identified.

For this study we will be collecting personal identifiers (full name, telephone number, email address) for the research purposes described in this consent form. Your collected personal identifiers will be kept in a document that links this information with a study ID, called a master list. The study ID will be used in all of the research documents to protect your privacy. The master list will be stored separately from the research data. It will be stored with password protection on a restricted computer securely stored in the Child Hearing Lab at the CHEO Research Institute with access restricted to the researcher and the research team.

Your professional role but not none of the personal identifiers describe above, will be included in the research data along with the study ID. The research data and audio recordings produced from this study will be stored with password protection on a restricted computer in the Child Hearing Lab at the Children's Hospital of Eastern Ontario Research Institute. Members of the research team and the individuals described above will have access to the data. Following completion of the study the research data, audio recordings and master list will be kept for 7 years after the last publication of this study. They will then be destroyed. You will not be identified in any publication or presentation of this study.

Is the research team benefiting from the study?

The research team members are not benefiting personally, financially or in some other way from this study.

Alternatives to participation

Taking part in this study is voluntary. Your decision to participate or not in this study, will in no way affect the care received by any member of your family at CHEO and will not affect your employment at the school board. You are free to withdraw from the study at any time and there will be no penalty to you. At any time during the study, you may decline to answer a question or stop an interview.

What if I have questions?

If you have any questions concerning participation in this study, contact: Eunjung Na or JoAnne Whittingham,

This study has been reviewed and approved by the CHEO Research Ethics Board (REB). The CHEO REB is a committee of the hospital that includes individuals from different professional backgrounds. The Board reviews all human research that takes place at the hospital. Its goal is to ensure the safety of people taking part in research. The Board's work is not meant to replace a parent or child's decisions and choices that are best for them. You may contact the REB, for information regarding a patient's rights in research studies at although the REB cannot provide any health-related information about the study.

Consent form signatures

Thank you for agreeing to participate in the interview. Below you will find the consent document for the study. Please sign this document.

The information collected for this project is confidential and protected under the Municipal Freedom of Information and Protection of Privacy Act, 1989.

I have read and understood the request to participate in the study "Cochlear implants for children with residual hearing: Decisional support needs of parents and children".

Consent form Signatures

By signing this consent form I agree that:

- I am voluntarily agreeing to participate in this research study;
- I understand the information within this consent form;
- All of the risks and benefits of participation have been explained to me;
- All of my questions have been answered;
- I do not give up my legal rights by signing this form.

A copy of the signed Information Sheet and/or Consent Form will be provided to me.

Signatures

Printed Participant's Name	Participant's Signature	Date

Printed Name of Person Who Conducted Consent Discussion	Signature of Person Who Conducted Consent Discussion	Date

Research Team:

Eunjung Na, M.Sc. / Ph.D. Candidate
Faculty of Health Sciences, University of Ottawa

Elizabeth Fitzpatrick, PhD, AUD(C) / Thesis Supervisor
Faculty of Health Sciences, University of Ottawa,
Investigator, CHEO Research Institute

Karine Toupin-April, PhD / Thesis Co-Supervisor
Faculty of Medicine, University of Ottawa,
Faculty of Health Sciences, University of Ottawa,
CHEO Research Institute

JoAnne Whittingham, M.Sc. / Research Coordinator
CHEO Research Institute