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The Development and Exploration of the Influence of a Decision Aid for Sequential Pediatric Bilateral Cochlear Implantation

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The development and exploration of the influence of a decision aid for sequential paediatric bilateral cochlear implantation

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Abstract

Introduction:
Bilateral paediatric cochlear implantation is a new clinical option for children with bilateral severe to profound hearing loss. The benefits and risks associated with the procedure need to be understood by parents and considered in light of their personal values before making an acceptable decision.

Objectives:
- To explore the decision-making needs of parents regarding cochlear implants (CIs).
- To provide updated, comprehensive information on the risks and benefits associated with paediatric bilateral CIs.
- To develop and pilot a decision aid for sequential paediatric bilateral CIs.

Methods:
- Clinicians and parents were interviewed using a semi-structured approach, regarding CI decisions that are perceived to be difficult, barriers and facilitators to decision-making, and potential strategies for overcoming barriers.
- Published research on paediatric CI benefits and risks was systematically gathered and synthesized. A retrospective chart review of local CI surgeries was also undertaken to estimate risks.
- A decision aid was designed based on parents needs, reviews of benefits and risks, and international (IPDAS) standards.
- A pre-test post-test design was used to pilot the decision aid and measure knowledge and decisional conflict aid with parents. The acceptability of the decision aid was assessed by parents and clinicians post-decision aid.

Results:
The need for additional decisional support was identified for some parents, particularly for bilateral CI decisions. The benefits and risks of bilateral implantation were systematically identified, documented, and estimated. A decision tool was designed to provide information about the options, their risks and benefits, as well as tools for parents to clarify and communicate the value they attribute to the risks and benefits. The piloted decision aid was acceptable to parents and clinicians and significant improvements in parents' knowledge were noted.

Conclusion:
Parents considering the bilateral CI decision have needs that are not being met with current decision support. An intervention that addresses this clinical gap was designed with updated evidence of benefits and risks and shows promise in improving parental decision-making for their children with hearing loss.

Dedication

For Hannah and Colin

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List of Symbols, Abbreviations, and Nomenclature

α: alpha

ABR: auditory brainstem response

AdSpon: Adaptive Spondee Test

AVT: Auditory Verbal Therapy

BICI: bilateral cochlear implant

CAEP: cortical auditory evoked potentials

CI: cochlear implant

CIHA: cochlear implant and hearing aid

CRISP: Children's Realistic Index of Speech Perception

dBHL: deciBel at Hearing Level

ESPT: Early Speech Perception Test

FM: frequency modulation

HA: Hearing Aid

HINT: Hearing in Noise Test

IPDAS: International Patient Decision Aid Standards

LNT: Lexical Neighbourhood Test

MAA: Minimum Audible Angle

Mdn: median

Mn: Mean

MLNT: Multisyllabic Lexical Neighbourhood Test

N: number of participants or number of studies

NH: normal hearing

ODSF: Ottawa Decision Support Framework

p: probability

PedsQL: Pediatric Quality of Life Measure

R/L: right/left

SD: Standard Deviation

SRM: Spatial Release from Masking

SSQ: Speech, Spatial, and Qualities of Hearing Scale

T: t-test result

US: United States

USFDA: United States Food and Drug Administration

WNL: within normal levels

Z: Z-test results

CHAPTER 1

Introduction, Theoretical Frameworks, and Methodology

Statement of the problem

Cochlear implants (CI) have become the standard of care for children with severe to profound hearing loss (Berg et al., 2007). Research has shown that CIs improve speech, language, and communication outcomes for these children (Moog & Geers, 2003; Thoutenhoofd et al., 2005). Originally, cochlear implantation was performed only in one ear due to the cost of the device, surgical risks, and the potential for language acquisition with unilateral auditory stimulation. Recently, more centres are performing bilateral cochlear implantation in children with the assumption that the two devices will provide children with binaural hearing ability (Berg et al., 2007).

The bilateral implantation procedure involves the simultaneous or sequential surgical implantation of a medical device in each ear to directly stimulate the auditory nerve. The implantation in both ears provides children with severe to profound hearing losses with bilateral auditory stimulation which may offer benefits such as sound localization and improved speech recognition in noise (Brown & Balkany, 2007; Ching et al., 2007; Murphy & O'Donoghue, 2007; Schafer & Thibodeau, 2006). These are the primary benefits that are accrued through binaural hearing in normal hearing individuals as well as those with hearing aids who are aided binaurally (Colburn et al., 2006; Mencher & Davis, 2006).

Early research examining the effectiveness of bilateral CIs for children that focused on sound localization and speech recognition in noise tasks indicated improvements for some, but not all children (Litovsky, Johnstone, Parkinson, Peters, & Lake, 2004; Litovsky, Parkinson, Arcaroli, Peters, Lake, et al., 2004). This variability may be due to differences in the characteristics of the population studied, such as age at implantation, or variations in experimental methods used. More recent work has shown consistent benefits on these

measures for many children, particularly those who are implanted following a shorter period of deafness (Brown & Balkany, 2007; Ching et al., 2007; Murphy & O'Donoghue, 2007; Schafer & Thibodeau, 2006). The increase in the number of bilateral implantations needs to be based on scientific evidence that clearly demonstrates the advantages of this approach. In addition, it is unclear if the benefits associated with bilateral implantation are justified in light of the risks associated with the procedure.

Because bilateral implantation is relatively recent, as well as a variation from the standard of care that is mainly unilateral implantation, questions arise from clinicians, parents, and CI users regarding the effectiveness, the benefits and risks, of the new bilateral procedure (Brown & Balkany, 2007; Berg, et al., 2007). Because of this, there is a need to review the scientific evidence for these multiple stakeholders.

One of the gaps in the literature is an overall estimation of risks associated with cochlear implantation. While there are individual hospital-based studies, no studies estimate overall rates. Current estimates suggest that major complications occur in 0 (James & Papsin, 2004) to 7.66% of CI surgeries (Migrov, Muchnik, Kaplan-Neeman & Kronenberg, 2006). There is an increased post-surgical risk of meningitis among children who have received an implant compared to children without implants and it is considered a major risk of the device. Facial nerve paralysis, vestibular problems, and risks associated with the use of a general anaesthetic are some of the additional complications of CI surgery (Fayad, Wanna, Micheletto, & Parisier, 2003; Fina et al., 2003; Gysin, Papsin, Daya, & Nedzelski, 2000). These are some of the risks associated with CI that may influence parental decision-making. There is also the risk of additional surgery due to device failure or trauma. Additionally, once CI surgery is undertaken, there is a high risk of losing residual hearing in that ear (Bergeron,

2000; Boggess, Baker, & Balkany, 1989). Therefore, a parent's decision to use a CI is typically irreversible.

At diagnosis, parents are often unfamiliar with hearing loss and rehabilitation options. Ninety percent of children with hearing loss are born to parents who have never experienced hearing loss themselves (Northern & Downs, 1991). The lack of knowledge surrounding hearing loss, the options for communication development, and the technologies for rehabilitation can be overwhelming for parents. In addition, the diagnosis of a hearing loss for these parents can be distressing and parents often go through a period of mourning and loss (Moses & Vantteche-Wulatin, 1981). Parents must absorb significant technical and scientific information and make choices for their children during this period of loss. They are often required to decide about communication options and early amplification before the CI or BICI decision so they have some information on hearing loss prior to making this decision. Parents making the unilateral CI decision, need to gain knowledge on the risks and benefits of the device that are now well documented in the literature (Moog & Geers, 2003; Thoutenhoofd et al., 2005). Parents contemplating BICIs need to gain similar knowledge on the advantages and risks of BICI, but this information is not as readily available. At the moment, the majority of BICIs have been sequential. Parents making the sequential BICI decision will have benefited from their experience with one CI for their child, but again would need to have information on the benefits and risks of BICIs. This need for information would also apply to parents considering simultaneous BICIs. This information, together with the consideration of parent's values is essential in allowing parents to make an informed decision on bilateral implantation.

Currently, there are no data indicating how parents react to the bilateral cochlear implantation decision. The literature does indicate that parents may find it stressful to make

the decision about whether or not to undergo unilateral CI surgery for their children (Incesulu, Vural, & Erkam, 2003; Most & Zaidman-Zait, 2003; Steinberg, Brainsky, Bain, Montoya, Indenbaum, & Potsic, 2000). While the decision to undergo unilateral implantation appears to be stressful for parents, most parents are opting to go ahead with implementation for their children. Now, with the need to decide about bilateral cochlear implantation, there is an additional component that parents must consider. Adding to the complexity of the decision is the fact that there is little information available for parents and clinicians regarding the benefits and risks associated with bilateral implantation.

Evidence-based information is essential for parents to make informed decisions for their children that are consistent with their values. Recent audiology and otolaryngology literature has included papers on the need for evidence of bilateral CI effectiveness for parents, researchers, and clinicians (Gregoret, 2003; Berg et al., 2007). One approach to disseminating knowledge for health-care consumers, including parents, is through the use of decision aids (O'Connor & Edwards, 2001). Decision aids are interventions that have three goals: 1) To prepare a person for decision making and provide facts about a person's condition, the options and their features, 2) To help people to clarify their values (the features that matter most to them), 3) To help people to share their values with their health care practitioner and others (International Patient Decision Aid Standards [IPDAS], 2006). Decision aids have been shown to improve the quality of the decision and the decision making process (O'Connor et al., 2004). Currently, no decision aid exists for any paediatric audiology topics, including the bilateral CI decision (Cochrane Inventory of Patient Decision Aids, 2004).

Purpose and Objectives

The specific objectives for the project were:

1) To explore the decision-making needs of parents regarding their experiences with unilateral and BICIs, including their: (a) perceived options, advantages, and disadvantages; (b) parental perceptions of their knowledge and expectations, values, support, and resources available to them; (c) decisional conflict and contributing factors when making the decisions; and (d) the need for a decision aid to support parents and clinicians
2) To provide clinicians, families, and researchers with updated, comprehensive information on the available paediatric BICI literature
3) To estimate risks for cochlear implantation at a local CI center and in the published literature
4) To develop a decision aid for sequential bilateral paediatric cochlear implantation that is acceptable to clinicians and parents, using International Patient Decision Aid Standards (IPDAS), the Ottawa Decision Aid Template, and available data
5) To pilot a decision aid and describe changes in knowledge and decisional conflict after using a patient decision aid among parents.

Present State of Knowledge

Paediatric Hearing Loss

Approximately 1 to 4 of 1000 children will be identified with a severe or profound permanent congenital hearing loss at birth or shortly afterwards (Fortnum et al., 2001; Mehl & Thomson, 2002). Causes of congenital hearing loss include prenatal infections, genetic conditions, or trauma during birth. Another group of children have acquired losses. These losses can be due to infections such as meningitis, ototoxic medications, or trauma (Northern

& Downs, 1991). Children with a unilateral hearing loss (normal hearing in only one ear) acquire language in much the same way as a child with normal hearing. A child with a permanent bilateral hearing loss (loss in both ears) often requires specialized early interventions to acquire language (Samson-Fang, Simons-McCandless, & Shelton, 2000). The level of the hearing loss often correlates with the level of intervention required for language learning.

Benefits of Unilateral Cochlear Implantation in Children with Hearing Loss

Children with bilateral, severe to profound hearing losses are eligible for cochlear implantation. CIs consist of an internal receiver, an electrode array (that is surgically implanted into the cochlea of the inner ear), and an external speech processor that processes sounds and transmits them across the skin to the implanted device. Following surgical implantation of the device, the electrode array is activated and controlled by an external speech processor programmed according to electrophysiological results (neural response imaging or telemetry) and the child's perception of sounds in the environment. Programming of the processor requires monitoring and repeated modifications by an audiologist to ensure that the child is hearing all that she should. Technological advances have made multi-channel devices the norm in place of the single channel devices that were originally developed (Thoutenhoofd et al., 2005).

The criteria for CI use in children that were established in 1990 have been expanded since then (Candidacy Criteria, 2008; United States' Food and Drug Administration [USFDA], 1990). Originally used only in children with profound hearing loss, now children with lesser degrees of hearing loss are eligible for CIs. In addition, children may be implanted with CIs at one year of age or earlier as in some cases of meningitis (Candidacy

Criteria, 2008). The candidacy criteria have been expanded due to the positive clinical and research results on the accessibility of environmental sounds and speech to children and adults with a CI (Thoutenhoofd et al., 2005). Prior to the use of CIs, many children with profound deafness were unable to detect soft speech sounds, even with hearing aids. The successes in speech and sound detection among CI users with profound hearing losses has led to the expansion of the use of CI devices to children with lesser degrees of hearing loss, namely those with severe and severe to profound losses. The expanded criteria and positive outcomes associated with CIs have made it the standard of care for children with severe to profound hearing loss (Berg et al., 2007).

A variety of medical, speech and language, educational, social, and family outcomes of cochlear implantation for children with severe to profound hearing losses have been examined in the literature. All of these outcomes are of interest to clinicians and parents considering cochlear implantation. A recent systematic review examined studies published between 1994 and 2002 that looked at these outcomes in children implanted before age five (Thoutenhoofd et al., 2005). Thoutenhoofd et al. (2005) evaluated 145 articles from the CI literature and reported that a large number of studies show improved hearing decibel levels for children following cochlear implantation. The authors also reported that speech recognition improved for children with *profound* losses who are implanted compared to those who use conventional amplification. CIs can help make more of the vowels, consonants, and supra-segmental features of language available to children with profound hearing losses and as a result this may contribute to improved oral language development (Martin & Clark, 1996). For children with *severe* losses who are implanted, there is less robust evidence available to indicate if speech perception is improved with cochlear implantation compared to hearing aid use (Thoutenhoofd et al., 2005).

The evidence therefore is described as strong and the outcomes robust for the improved speech perception and production of children with profound hearing losses with CIs. Nonetheless, variations in speech perception exist for children with hearing losses with similar audiometric configurations. For example, some children with severe or profound hearing loss seem better able to perceive speech and develop oral language than others, even before CI surgery (Geers & Brenner, 2003; Gordon, Daya, Harrison, Papsin, 2000). Factors such as pre-implantation language and speech perception abilities (Gantz, Tyler, Woodworth, Tye-Murray, & Fryauf-Bertschy, 1994) and age at implantation (Harrison, Panesar, El-Hakim, Abdolell, Mount, & Papsin. 2001; Nikolopoulos, Archbold, Lutman, & O'Donoghue, 2000) have all been shown to influence post-implant speech perception and production. Furthermore, the rehabilitative choice of the parents, family characteristics, and a child's cognitive ability are all hypothesized to influence speech perception and production however little research exists to support these hypotheses.

The systematic review by Thoutenhoofd et al. (2005) reported that the research on the outcomes for language development was inconclusive or contradictory. While some studies indicated improved oral language development for many children using CIs, others showed that children do not improve their oral language development significantly. There are both pre-implant (duration of deafness and the age of child) and post-implant factors (poor speech processor programming and limited rehabilitation therapy) that seem to be predictors of poor language acquisition (Gordon, Daya, Harrison, Papsin, 2000; van Dijk, van Olphen, Langereis, Mens, Brokx, & Smoorenburg, 1999). One of the limitations of the studies included in the systematic review was that many of the included studies were conducted on convenience samples of small groups of children, using a wide variety of outcome measures

and testing administration approaches. Significant methodological heterogeneity exists in the early cochlear implant literature included in this systematic review.

A recently published, multi-site, study of 181 children that used standardized language measures showed greater improvements in language of children using CIs as compared to earlier evaluations of children using hearing aids (Moog & Geers, 2003). In fact, half of the 181 participating children had some language scores within the range of normally hearing peers (Geers et al., 2003). The researchers of this study examined language comprehension and production. This research clearly demonstrated the benefits of cochlear implantation for children on a variety of these standardized measures.

There are a number of research strengths associated with this important study. This study had an adequate sample size to control for child and family characteristics that previous, smaller sampled studies were unable to do. In addition, all included children had used cochlear implants for 4 to 7 years. As such, this study also provides evidence about the medium- to long-term outcomes of cochlear implantation rather than some of the earlier outcomes, such as speech perception and production that was examined in the early CI literature. The positive outcomes identified in this research may also be most relevant for parents currently making the CI decision. The children in the Geers and Moog (2003) study, as recipients of CIs in the late 1990's, rather than the early 1990's, may have benefited from standardized fitting and rehabilitation strategies developed following the early years of cochlear implantation and that remain in place. These results have contributed to the strength of support of CIs.

The research on educational placement and communication approach used by children with CIs is also inconclusive (Thoutenhoofd et al., 2005). Although children with CIs tend to be placed in mainstream schools more frequently, it is uncertain if this is a result

of the CI itself or other confounding variables (Thoutenhoofd et al., 2005). Studies have compared current rates of mainstreaming in children with permanent hearing loss with those of previous years, however educational trends in increased inclusion of students with disabilities over the past number of years potentially confound a retrospective comparison (Archbold, Nicolopoulos, Lutman, & O'Donoghue, 2002; Archbold, Nicolopoulos, Tait, O'Donoghue, Lutman, & Gregory, 2000). It is also uncertain if parents of children with implants request mainstream placement more frequently than parents of children without CIs.

Little evidence was reported to be available for outcome measures such as employment and educational outcome, social and psychological outcomes, and quality-of-life measures (Thoutenhoofd et al., 2005). This may be due to the fact that paediatric cochlear implantation is still relatively new and that participants of late school or employment age may not yet be available. A recent additional systematic review of the paediatric cochlear implantation quality of life research reported that there are methodological limitations in drawing conclusions regarding the improvement in quality of life of CI users due to the potential of bias in subject selection and heterogeneity of tools used (Lin & Niparko, 2007). The lack of studies of strong methodological rigor does not indicate that there is a lack of quality of life improvements for child users of CIs, rather, it indicates that there is a lack of good quality evidence documenting either the benefits or the lack thereof. Evidence also remains unclear regarding the relative benefits of CIs to hearing aids for children who are borderline audiologic candidates and those with comorbid disabilities due to a lack of research examining outcomes in these populations (Thoutenhoofd et al., 2005).

Strong systematic review and longitudinal cohort study research evidence exists to support the use of CIs to improve sound detection, speech perception, and language improvements for children with severe to profound hearing losses (Moog & Geers, 2003; Thoutenhoofd et al., 2005). These results have made the CI option the standard of care and the option of choice to most, although not all, parents and clinicians. There remains a need for additional longitudinal research to examine some of the additional outcomes of interest that may include educational outcomes and quality of life.

Risks of Cochlear Implantation in Children with Hearing Loss

While CIs can provide significant benefits for many children, there is also the potential for complications or adverse events with implant surgery and/or device use. There are both minor and major risks associated with CI surgery (Cohen et al., 1991). Major surgical complications are those that require additional surgery or medical management in a hospital setting (e.g. facial nerve paralysis, flap infection requiring hospitalization, blood loss). Minor complications are defined as those that are treated with medical management on an outpatient basis (e.g. flap infections requiring oral antibiotics, haematoma). Possible adverse effects due to implantation use can include tinnitus or imbalance. Increased post-surgical risk of bacterial meningitis among children who have received an implant has been highly publicized and is considered a major risk of the device (Health Canada, 2003; USFDA, 2006). Mastoiditis, otitis media presenting as postauricular edema and/or erythema, may also occur in some patients due to the removal of the mastoid during the CI surgery. These adverse events are not due to the implant itself but occur as a result of the surgical procedure used during implantation. This complication can be major, requiring intravenous antibiotics and hospitalization, or minor, requiring oral antibiotics on an outpatient basis.

While all child CI recipients with a normal life expectancy will experience a device

failure at some point during their lives, some will experience a device that will fail to function optimally prematurely. Device failure can occur due to an internal defect in the device or due to trauma to the device from impact to the head (Battmer et al., 2007). In both cases, the device malfunction requires additional surgery to explant the malfunctioning device and reimplant a new one. Additionally, once CI surgery is undertaken, there is a high risk of losing residual hearing in that ear (Bergeron, 2000; Boggess, Baker, & Balkany, 1989). Therefore, the parental decision to use a CI is typically irreversible.

Current estimates of complications of CI surgery available in the published literature are derived primarily from hospital-based retrospective studies (Arnoldner et al., 2005; Bhatia et al., 2004; Black et al. 2007; Cunningham et al., 2004; Gysin et al., 2000; James et al., 2004; Kandogan et al., 2005; Maurer et al., 2005; Migrov et al., 2006a; Migirov et al., 2006b; Parisier et al., 2001; Postelmans et al., 2006; Weise et al., 2005; Stratigouleas et al., 2006; Venail et al., 2007; Yu et al., 2001). There is considerable variation in the complication rates reported in these studies. For example, some studies report no minor complications in their results (James et al., 2004) while others report rates as high as 18.75% (Postelmans et al., 2006). Retrospective studies of medical complications associated with paediatric CI surgery suggest that the rate of major complications range from 0% of CI surgeries (James et al., 2004) to 7.66 (Migrov, Muchnik, Kaplan-Neeman & Kronenberg, 2006).

Ethical or Cultural Issues Arising from Cochlear Implantation

In addition to the surgical risks and benefits there are ethical or cultural issues that arise with the decision regarding cochlear implantation. Some individuals and organizations have had negative perceptions of the CI technology, particularly when used with pre-lingual

infants (Crouch, 1997). As hearing loss does not in itself present a medical problem but is a potential risk factor for the lack of development of spoken language (but not sign language) the risks of surgery are perceived by some as being unnecessary (Peters, 2000).

For the most part, there are three distinct types of communication development options for children with significant hearing loss. Auditory-oral communication uses spoken language, amplification devices, and the child's residual hearing (Reamy and Brackett, 1999). Manual methods use some form of American Sign Language (ASL) which is a distinct language used by many members of the Deaf community in North America. There is also a third bilingual/ bicultural approach that seeks to provide children with hearing loss an opportunity to learn both spoken (through amplification) and signed languages (through ASL instruction).

Prior to the use of CIs, children with severe and profound hearing losses had limited access to auditory stimulation even with hearing aids and often became users of ASL through their educational environment. These children became a unique cultural group that used its own language and had hearing loss as one of its defining features. With the increased use of CIs, some in the Deaf community have felt that the existence of ASL and Deaf culture itself has been threatened due to the increase in auditory-oral programs for CI users and the reduction in ASL and bilingual/bicultural programs available for CI users (Nevins & Chute, 1996). While many in the hearing community, particularly hearing parents of children with hearing loss, may feel that CIs provide children with the opportunity to become more active members of the *hearing* community and have actively sought out auditory-oral communication programs for their children. The cultural and ethical issues raised by the Deaf community may be relevant for some parents making the CI decision and should be considered by all parents making the CI decision (National Association of the Deaf, 2000).

The strength of the opposition to CIs has varied over the time since the introduction of the devices as well as by geographic location (Thoutenhoofd et al., 2005). Currently, both the Canadian Association of the Deaf (2007) and the National Association of the Deaf of the United States (2000) acknowledge that there is a role for CI use for both adults and children. These organizations emphasize that parents of children with hearing loss should be aware of all their options as well as be informed of the risks and benefits associated with these options prior to making a decision regarding cochlear implantation. Both the American and Canadian groups emphasize that CIs are assistive devices that may improve sound and speech detection but that do not preclude the use of, or need for sign language. The Canadian Association of the Deaf (2007) currently states in their position statement that children with CIs are important members of their community and that they hope that these children will be offered bilingual/bicultural opportunities.

Bilateral Cochlear Implantation

The improved speech production, perception, and language outcomes for children with unilateral CIs have made CIs the standard of care for children with bilateral severe to profound hearing losses (Berg et al., 2007; Moog & Geers, 2003). A recent trend is that more centres are performing bilateral cochlear implantation in children (Berg et al., 2007). Bilateral implantation involves the simultaneous or sequential surgical implantation of the device in both ears. At the moment, as most children have already been provided with a single CI, the majority of bilateral cochlear implant (BICI) surgeries have been sequential (Shafir, 2007). Bilateral implantation provides children with severe to profound hearing losses with bilateral auditory stimulation. As with any new medical procedure, questions arise from clinicians, parents, and CI users regarding the effectiveness, the benefits and risks,

of paediatric bilateral cochlear implantation (Brown & Balkany, 2007; Berg, et al., 2007). Bilateral implantation is not currently the standard of care for children with severe to profound hearing loss.

There are a number of reasons for pursuing bilateral cochlear implantation. For one, the psychoacoustic literature has demonstrated the benefits of binaural hearing for those with normal hearing and for many with bilateral amplification using measures of speech recognition and sound localization (Colburn et al., 2006; Mencher & Davis, 2006). Speech recognition can be measured in noise or in quiet. Speech recognition in noise is typically improved in binaural compared to monaural hearing conditions (Zurek, 1993). Sound localization refers to the ability to identify and locate the source of a sound in space (Brown & Balkany, 2006). Those with available binaural signals take advantage of the head shadow, binaural summation, and binaural squelch effects to improve their speech recognition in noise and sound localization performance (Brown & Balkany, 2007). As such, it is a logical area for bilateral cochlear implantation research to identify if the same benefits are accrued through the auditory stimulation provided by two CIs.

Recent literature reviews examined the literature on bilateral cochlear implantation have explored the effectiveness of BICIs on sound localization and speech recognition in noise measures (Brown & Balkany, 2007; Ching et al., 2007; Murphy & O'Donoghue, 2007; Schafer & Thibodeau, 2006). These reviews included studies examining the effectiveness of bilateral cochlear implants (BICIs) compared to unilateral CI use in adults and children. All four reviews (Brown & Balkany, 2007; Ching et al., 2007; Murphy & O'Donoghue, 2007; Schafer & Thibodeau, 2006) concluded that there are benefits for patients receiving bilateral stimulation compared to use of a single CI on measures of speech recognition in noise and sound localization. Children with increased residual hearing in their non-implanted ear may

receive benefit from a hearing aid in their second ear (Ching et al., 2007; Schafer & Thibodeau, 2006). For children who cannot use hearing aids in their second ear due to severity of hearing loss, this bilateral stimulation would only occur through the use of a second CI.

A second reason for implanting a device in the second ear is to try and reduce the effects of auditory deprivation on the child's developing auditory system. As a child develops from infancy to adulthood, a number of changes take place in the developing auditory pathways that affect their responses to sound (Werner & Grey, 1998). The development is hypothesized to be due to changes in the external ear, cochlear and neural pathways, and attentional factors. While some measures of audition, such as auditory sensitivity, undergo rapid changes and approach adult levels of hearing relatively early (Werner & Grey, 1998), complex auditory processing, such as temporal processing, does not approach adult levels until late childhood (Jensen & Neff, 1993). In order for the bilateral auditory system to develop during this critical period, children must be exposed to auditory stimuli and speech in both ears. Children with hearing loss can experience this stimulation through the use of bilateral hearing aids or CIs, or a CI and a hearing aid (Ching, van Wanrooy, & Dillon, 2007). For those children with insufficient residual hearing for amplification, the binaural auditory stimulation required for the development of this auditory system may only be possible through the use of two CIs.

As BICIs become increasingly available in paediatric CI centres (Berg et al., 2007), it is essential that up-to-date research information is available for parents and clinicians on a variety of outcomes of interest. To date, sound localization and speech recognition in noise measures are important and have been explored in the literature (Brown & Balkany, 2007; Ching et al., 2007; Murphy & O'Donoghue, 2007). Speech, language, education, and quality

of life outcomes are also important to those making decisions about bilateral implantation and have not been included in published reviews to date. Recent audiology and otolaryngology literature has included papers on the need for evidence for parents, researchers, and clinicians about the benefits of bilateral implantation (Gregoret, 2003; Berg et al., 2007). In addition, the risks of acquiring a second CI need to be ascertained as part of the necessary evidence for decision-making.

Parental Decision Making

The risks and benefits surrounding the decision to pursue either unilateral or bilateral cochlear implantation must be weighed and considered in light of a parent's or family's personal values. The risks and benefits may be valued differently depending upon what matters most to the individual making the decision. All individuals make decisions with different past experiences and cultural or personal values.

There is currently no available literature on parental experiences of the BICI decision-making process. However, having a better understanding of the unilateral CI decision-making process may provide a way to reduce parental stress during this process or to meet any specific information needs that are identified. Publications from countries around the world have described the challenges parents face in deciding whether or not to undergo unilateral cochlear implantation for their children (Incesulu, Vural, & Erkam, 2003; Li, Bain, & Steinberg, 2004; Most & Zaidman-Zait, 2003; Peters, 2000; Sach & Whynes, 2005; Sorkin & Zwolan, 2008; Steinberg, et al., 2000). Incesulu et al. (2003) report that 81% of parents responding to a survey indicated that making the CI decision was the most difficult aspect of the implantation process for them. Most and Zaidman-Zait (2003) also describe the high parental stress during the implantation decision-making process and the specific parental needs for information to aid in the process.

In contrast, Sach and Whynes (2005) of the UK report that most of the 216 interviewed families found the decision regarding implantation to be straightforward. They did, however, describe the overall stress for families undergoing cochlear implantation. A very recent survey of parents in the US indicates that those who chose the CI for their child felt that they lacked "comprehensive and bias-free" information when making the decision (Sorkin & Zwolan, 2008, p.1).

Differences in parental perspectives of the CI decision making process appear throughout the literature. These variations may be due to the decision making process for individual families and differences across regions or CI centres. Alternatively, the reported stress may be due to the uncertain knowledge of the rates of risks and benefits, uncertain parental values regarding their decision about communication approaches (e.g., oral or sign language) that may be linked to the CI decision, inadequate information about options, or pressure from clinicians or other family members. The need for additional information is also highlighted in the literature. Kluwin and Stewart (2000) interviewed 35 families who had undergone cochlear implantation with their children. While most of the families were satisfied with the information they received, eight families felt that they would have liked more information on the surgery and rehabilitation process. Neuss (2004) describes, using case studies of parents of children with hearing loss, the search for information that both mothers and fathers undertake before deciding to undergo cochlear implantation. These studies emphasize the importance of sharing information for some parents.

There is no available literature describing the bilateral cochlear implantation decision-making process from the perspective of parents. From the literature on unilateral cochlear implant decision making it appears that there can be increased stress and a lack of knowledge for at least some parents making the cochlear implant decision. In addition, there

appears to be a need for comprehensive information on the benefits and risks of CIs to help parents with decision-making. As the bilateral CI decision is an even newer procedure, with less available evidence to support decision-making, and not currently the standard of care, it is likely that parents also require additional information for this decision-making.

Knowledge Translation

Early definitions of knowledge translation (KT) separated the concept into two distinct components: 1) the dissemination of research into user-friendly, accessible, and tailored documents and information for various stakeholders, and 2) the uptake of this knowledge and research by various users (Lomas, 1997). More recent definitions have broken down the process more precisely, "Knowledge translation is a dynamic and iterative process that includes synthesis, dissemination, exchange and ethically sound application of knowledge to improve the health of Canadians, provide more effective health services and products and strengthen the health care system," (Canadian Institute of Health Research, 2008). This CIHR (2008) definition emphasises the multiple components of KT by including the synthesis, dissemination, exchange, and application of research in its definition. All elements are essential for getting research into action to improve the health of the population.

CIHR describes that KT can be accomplished by using two different types of activities: "end of grant KT" activities, and "integrated KT" activities. End of grant KT activities involve the dissemination of knowledge to relevant stakeholders following the completion of a grant or primary research. Typically this includes peer-reviewed publications, educational sessions, and media releases. In contrast, integrated KT activities involve the stakeholders at all stages of the project, including setting the research question and primary research objectives. The integrated KT projects may produce findings that are

more relevant to stakeholders, however it may be extremely time consuming in establishing consensus and ensuring that all stakeholders are included at all stages in the project's development.

Both end of grant and integrated KT projects need to consider all relevant stakeholders as the projects progress. Health care providers, policy-makers, researchers, and patients may all be relevant stakeholders depending on the nature of the project. Including patients in their care and health decisions is an essential component of KT and the primary focus of this thesis work (Holmes-Rover et al, 2001; Coulter, 1997). Another key element of knowledge translation is improving patient knowledge and developing appropriate tools for knowledge dissemination for these stakeholders (Logan & Graham, 1998).

Decision Aids for Evidence Based Decision Making

One approach to translating knowledge for health-care consumers is through the use of patient decision aids (O'Connor & Edwards, 2001). Decision aids are tools that, "prepare a person for decision making. They provide facts about a person's condition, the options, and their features. They help people to clarify their values (the features that matter most to them). They help people to share their values with their health care practitioner and others," (International Patient Decision Aid Standards (IPDAS, 2006).

Decision aids are particularly important in health situations where a choice between two or more treatments or options is available and no clear standard of care is available based on evidence (O'Connor & Edwards, 2001). Eddy (1992) makes a distinction between standards of care, guidelines, and options while Wennberg (2002) distinguishes between "effective" and "preference sensitive" care. When adequate scientific evidence documenting clear benefits and few harmful effects associated with one health choice is available (i.e. a

clear standard of care or "effective" care) there is often little conflict regarding the choice that is to be made. KT activities for "effective" care often try and increase the uptake of these health interventions. However, many health decisions are not based on clear evidence, or the evidence that is available indicates that the choices carry both harms and benefits that may be differentially viewed depending upon the personal preferences of the client (O'Connor, Legare, & Stacey, 2003; Wennberg, 2002). KT activities for these "preference sensitive" decisions focus on providing information about the benefits and harms of each available option. When individuals are faced with making preference-sensitive decisions, there can be increased decisional conflict. Decisional conflict is the state of uncertainty about the best course of action (O'Connor, 1995).

Decision aids are tools specifically designed to help patients become informed regarding preference sensitive options. They have been developed for over 252 health decisions (Cochrane Inventory of Patient Decision Aids, 2008). Both screening and treatment issues have been addressed using decision aids. A Cochrane systematic review of the effectiveness of decision aids shows that the decision-making quality and process is improved with their use (O'Connor et al., 2002). Improved knowledge and reduced decisional conflict were found among those who use decision aids compared to those who receive usual care. Decision aids were also shown to increase participation in decision-making without increasing anxiety as well as creating more realistic expectations of outcomes. The effectiveness of decision aids in improving the decision-making process and in appropriately translating research knowledge regarding "preference sensitive" decisions to consumers makes them applicable in many healthcare situations, including to the BICI decision.

Preference Sensitive Decisions in Audiology

In the field of Audiology, providing evidence based practice continues to be a challenge (Robinson, 1999). Despite calls to improve the knowledge base in the field, searches of systematic review and randomized controlled trial databases reveal limited results for audiology using keywords such as hearing and audiology (Cochrane Collaboration, 2004). Those systematic reviews that have been conducted often conclude that there is inadequate evidence to recommend an intervention option or particular practice due to the lack of available evidence (Schachter et al, 2002; Thompson et al, 2001). As such, many audiology clinical decisions may be deemed to be "preference sensitive" rather than "effective" (Wennberg, 2002). Examples include the decisions for adults with minimal hearing loss to use hearing aids or not, the style of hearing aid chosen, the type of communication development program chosen for children with hearing loss. Based on the available literature, the BICI decision, also appears to be a preference-sensitive decision.

In order for audiology clients to make decisions that are well-informed and consistent with their personal values, it is essential that the available relevant research knowledge is disseminated. Decision aids are the most relevant tool for disseminating the current state of knowledge, explaining the choices, and exploring the personal preferences of patients (O'Connor & Edwards, 2001). No decision aids have been developed for any audiology decisions. There appears to be a need for comprehensive information on the benefits and risks of bilateral CIs to help parents with this new decision-making and a decision aid is the most appropriate way to provide this decisional support.

Population Health Approach

There are a number of key elements of the Population Health Approach that are reflected in this thesis. The key elements of this approach include: addressing the determinants of health, focusing on the health of the population, basing decisions on evidence, increasing upstream investments, applying multiple strategies, collaborating across sectors and levels, employing mechanisms for public involvement, and demonstrating accountability for health (Health Canada, 2001). This thesis work will use an evidence-based approach, address the determinants of health of child development and personal health practices, engage multiple stakeholders in the development of the intervention, apply multiple strategies to improve decision-making, and intervene upstream in the decision-making process.

One of the key determinants of health in Population Health is "healthy child development". Parents are key contributors to the health of their children. Parent/child interactions are essential for language, cognitive, and social development. The decisions that parents make for their children with regards to cochlear implantation may have a long-lasting influence on the relationship that they will have with each other. Providing a tool that helps parents make the best choice for their family and their child is an important component of their relationship and for the development of their child.

A second key determinant of health addressed by this project is, "personal health practices and coping skills of the population" (Health Canada, 2001). In order to positively influence the personal health practices of the population it is important that evidence of positive health practices be made available to health care consumers. Patients or health care consumers, require research dissemination for making optimal health decisions and

improving their personal health practices. This thesis work will address the determinant of health that is personal coping skills and health practices.

The evidence-based nature of the Population Health approach is highly relevant to this thesis. This thesis work increases the available evidence for parents and clinicians and provides a tool for sharing research knowledge with parents. The bilateral cochlear implant decision aid acts as a model for the development of decision aids in the field of Audiology and for the use of an evidence-based approach in general.

The stakeholders included in the research are audiologists, auditory verbal therapists, ear-nose-and-throat surgeon, psychologist, parents, and researchers from Nursing, Economics, and Audiology. By including these stakeholders in the research process the final decision-making tool will be more relevant and inclusive of a variety of perspectives.

Summary

The decision to pursue cochlear implantation is made based on clinical information regarding a child's hearing loss, age, family support, and other health factors, as well as the best available scientific evidence. However it is also dependent upon family preferences, values, and involvement. The paediatric cochlear implantation decisions, both unilateral and bilateral, are ones in which a great deal of information must be shared with the family and must be adequately understood in order for these decisions to be evidence-based. The bilateral cochlear implant decision is relatively new decision for parents and clinicians. As the benefits are different for the bilateral cochlear implant decision, parents' values of these benefits may also differ. These factors make the development of a decision aid potentially important. Decision aids are an effective tool for ensuring the risks and benefits of the surgery are understood accurately and that expectations are realistic (O'Connor et al., 2004).

There is a role for decision aids in ensuring that parental knowledge is high and that the decision to undergo bilateral cochlear implantation is consistent with parental values regarding the benefits and risks.

Conceptual frameworks

In developing and evaluating a decision aid there have typically been three types of frameworks used; prescriptive, descriptive, and transactional. Prescriptive frameworks assume that human beings are unable to adequately consider all of the factors required in decision-making. As such, decision analysis and computer programming are often included to help in decision-making that will maximize a person's expected utility (Sox, 1999). Decision-making is viewed as a rational process. While descriptive frameworks may use some probability calculations, they do not undertake formal utility modelling like prescriptive frameworks. Prescriptive models are often perceived as being too impractical and free of the context and values which are attached to decision-making. Descriptive models are based upon patient education and value clarification as well as the presentation of the probability of outcomes (O'Connor et al, 1998). Unlike prescriptive frameworks, a decision is not prescribed to the patient at the conclusion of the use of decision aid using a descriptive framework. Transactional frameworks focus on the roles of the patient and clinician (Charles, Whelan, & Gafni, 1999). The decisional support provided is unique and dependent upon the specific needs of the patient. There is a strong emphasis placed on the participatory role of the patient during the decision-making process and decision aids are used during the clinical encounter.

While each of these frameworks has a role depending upon the decision to be made, a descriptive framework is used to guide the development of the decision aid for parents

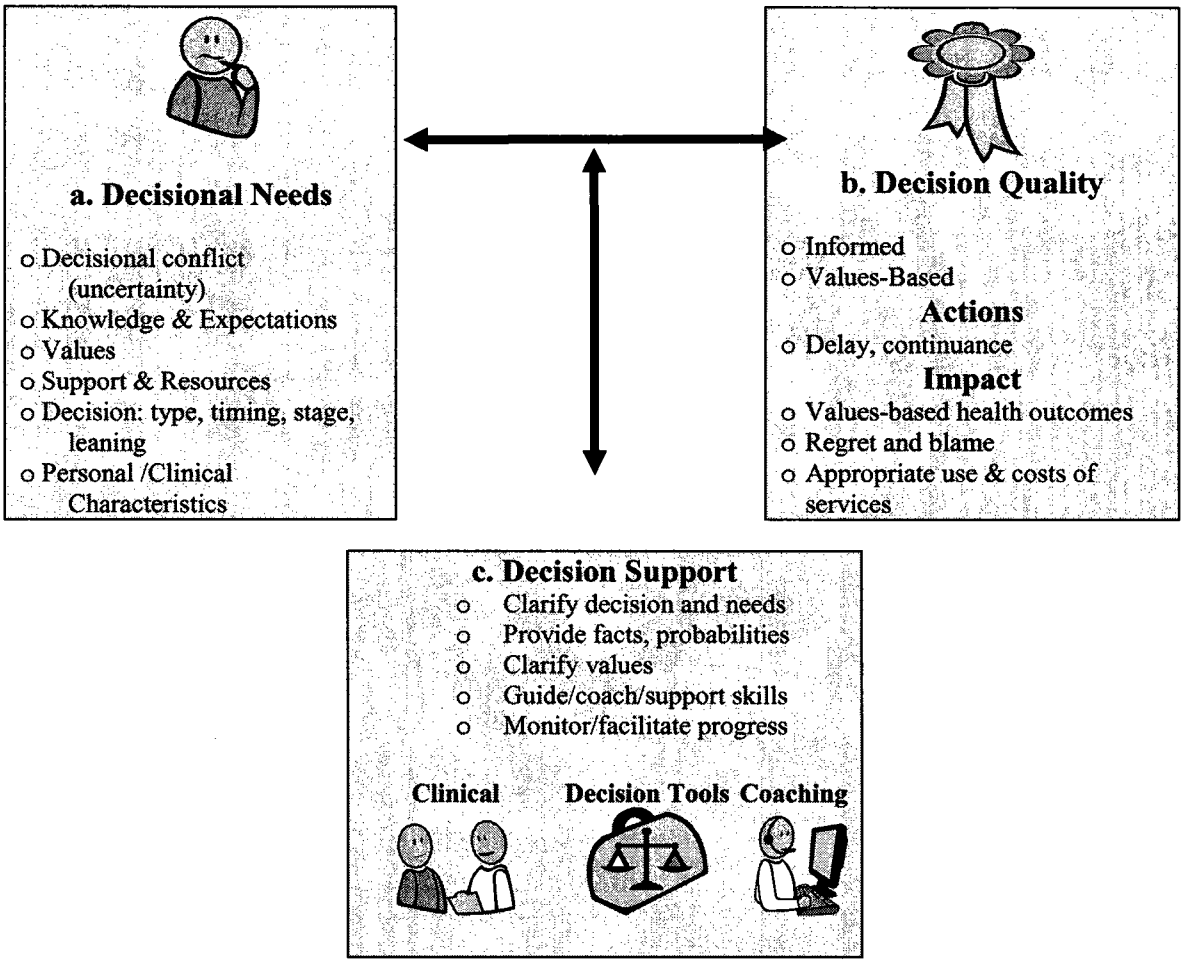
choosing to undergo bilateral cochlear implantation for their children. The Ottawa Decision Support Framework (ODSF) (O'Connor et al., 1998) is chosen as the framework for guiding this needs assessment and is presented in Figure 1.

This framework is appropriate for decisions that: "(1) are stimulated by a new circumstance, diagnosis, or developmental condition; (2) require careful deliberation because of the uncertain and / or value sensitive nature of the benefits and risks; and (3) need relatively more effort in the deliberation stage than the implementation stage," (O'Connor et al., 1998, p.268).

The paediatric cochlear implementation decision meets each of these criteria. This theoretical framework depicts how a family's decisional needs and decisional quality influence each other. Decisional needs are deficits in: (1) elements of the decision (timing, stage, leaning); (2) decisional conflict; (3) knowledge and expectations; and (4) values clarity. Decision support is used to address decisional needs to improve the quality of decisions. Each of these factors is explored for the paediatric BICI decision in the first project of the thesis- the needs assessment. For example, parents' may express decisional needs regarding:

- Inadequate knowledge of options, unrealistic expectations of outcomes, unclear values, and personal uncertainty regarding the course of action (decisional conflict), lack of receptivity to contemplate options based on their stage of decision making and strong predispositions toward the options;

Figure 1: Ottawa Decision Support Framework



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AM O'Connor, Ottawa Decision Support Framework to Address Decisional Conflict © 2006

- Unclear or biased perceptions of what important others think they ought to do, inadequate support for decision making, pressures they feel to choose a certain option, and mismatches between their preferred and actual roles in decision making;
- Inadequate personal and external resources to make a decision;
- Special needs based on education, culture, and health status of their child.

Decisional support as provided by the ODSF is accomplished by 1) providing access to information on the health situation, options, and outcomes, 2) re-aligning expectations by presenting probabilities of outcomes, 3) clarifying personal values for outcomes, 4) providing guidance in decision making. Each of these supports for improving decisional support is applied when the needs assessment identifies a sub-optimal determinant of decision. Applying this framework, a decision aid can be developed that provides these decisional supports for the CI decision. This decision aid will be used as an adjunct to counselling which clarifies and re-enforces risk communication, values clarification, and decision coaching.

The third element of the framework is decision quality. In 2005, the International Patient Decision Aid Standards Collaboration (IPDAS) developed a set of criteria for determining the quality of decision aids (See Appendix A). The IPDA standards require that a decision aid is effective in improving both the decision quality and process. Decision quality is high if it is informed and value-based. The decision process must also be informed and value based. The decision-making process for parents making the CI decision for their children will be examined using measures of decisional conflict, knowledge, values, and satisfaction with decision-making as delineated by the ODSF and required by IPDAS.

In addition to the use of the ODSF, the Population Health Promotion Model (Health Canada, 2005) served as an underlying theoretical framework for the thesis research (See Figure 2). This is a three dimensional model of health that includes the Ecological model, the Ottawa Charter on Health Promotion, and the ten determinants of health, each on one of the three dimensions. This model represents how the health of a population is determined by the determinants of health and is influenced through the use of a variety of health promotion strategies. The health promotion strategies include strengthening community actions,

building healthy public policy, creating supportive environments, developing personal skills, and reorienting personal skills. These health interventions are focused on the individual, the family, a community, a sector or system, or society. The entire population health promotion model rests on an evidence base. No population health intervention acts upon all of these elements within one initiative. Rather, the model represents all of the different types of population health interventions that are possible.

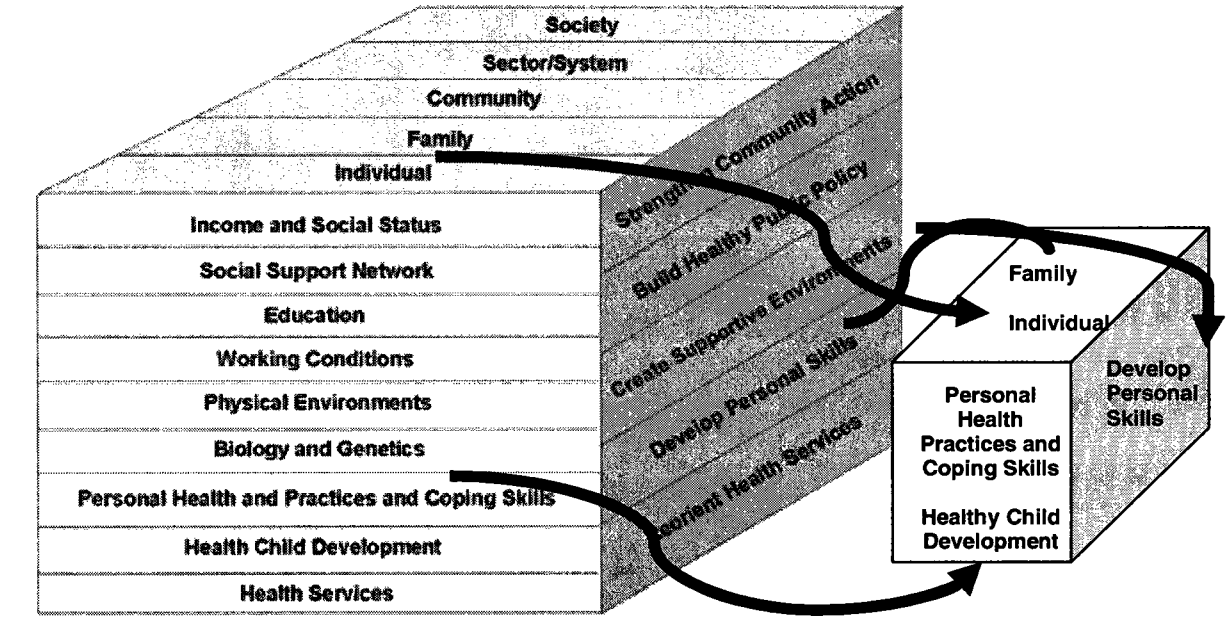
The decision aid intervention developed in this thesis research fits into the population health model as a health promotion strategy that develops personal skills for the family or individual to address their personal health practices, coping skills, and knowledge and healthy child development. Figure 2 presents the population health model and a representation of this thesis research as one element from the larger model. The evidence base of this population health promotion model was an essential component of this thesis research.

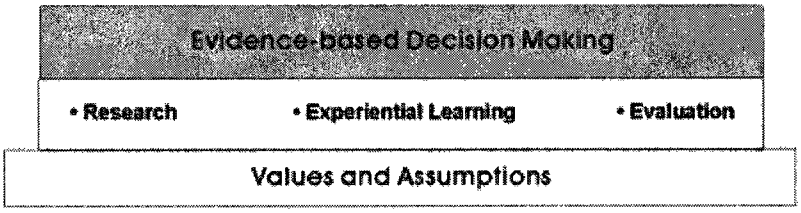
Methods

This thesis project is an example of integrated KT. Each component of the project includes stakeholders to ensure relevance of the project for parents and clinicians. There are three related studies that will accomplish the thesis objectives: the needs assessment, the development of the decision aid, and the examination of parent and practitioner response to the decision aid. These three projects, their specific objectives, design, and methodology are summarized in Table 1 and described in detail in the method sections. Together, these three projects, and the four papers that emanate from them, will form an original contribution to the fields of Population Health, Audiology, and Decision Making. This work will also meet

the thesis requirements of the Faculty of Graduate and Postdoctoral Studies and the Population Health PhD Program.

Figure 2: Population health promotion model and a representation of the thesis research within this model.





Population Health Promotion Model

The representation of the BICI decision aid intervention as one element of the Population Health Promotion Model.

Table 1: Description of projects, objectives, and methods used in thesis

Major Projects and Objectives	Research Design	Method	Products
Needs Assessment • To explore the decision-making needs of parents regarding unilateral and bilateral CIs, including their: (a) perceived options, advantages, and disadvantages; (b) parental perceptions of their knowledge and expectations, values, support, and resources; (c) decisional conflict (d) the need for a decision aid.	• Semi-structured interviews with parents currently making the decision and with parents post-decision using the ODSF • Semi-structured interviews with clinicians	**Participants:** 3 families awaiting their CI surgery, 5 families post-CI, 9 clinicians **Measures:** Semi-structured interview guide based on standard Ottawa template **Analysis:** Content analysis for interview data using a deductive coding strategy based on ODSF, descriptive statistics for structured interview responses	**Chapter 2:** An assessment of parents' decision-making regarding paediatric CIs
Development resources for the decision aid • To provide clinicians, families, and researchers with updated, comprehensive information on the available paediatric BICI literature • To estimate risks for cochlear implantation at a local CI center and in the published literature	• Systematic Review • Secondary analysis of existing local database of patient cohort for estimating probabilities of risk • Systematic standardized methods for development of decision aid with expert panel	**Procedure:** • Critical review of benefits to ensure evidence-based development of decision aid • Estimating probabilities of harms using local databases and available literature • Development of a decision aid using the Ottawa decision aid Template and applies International Patient Decision Aid Standards (IPDAS) standards	**Chapter 3:** Bilateral paediatric CIs: A critical review **and** **Chapter 4:** Estimation of risks of paediatric cochlear implantation
Description of parents' and practitioners' responses to the decision aid • To develop a decision aid for sequential bilateral paediatric cochlear implantation that is acceptable to clinicians and parents, using IPDAS standards, the Ottawa Decision Aid Template, and available data • To pilot a decision aid and describe changes in knowledge and decisional conflict after using a patient decision aid among parents.	• Pre-test, post-test design to test parents' knowledge and decisional conflict • Acceptability questionnaire and semi-structured interviews with clinicians who have used the decision aid	**Participants:** 8 Parents undergoing BICI decision-making, 5 clinicians **Measures:** Standardized measures of knowledge and decisional conflict taken pre-, and post- decision aid. Acceptability questionnaire completed post-decision aid **Analysis:** Descriptive statistics, paired t-tests, content analysis of interviews using an inductive coding strategy	**Chapter 5:** The development and piloting of a decision aid for parents considering sequential bilateral cochlear implantation for their child with hearing loss

References

Archbold, S.M., Nikolopoulos, T.P., Tait, M., O'Donoghue, G.M., Lutman, M.E., & Gregory, S. (2000). Approach to communication, speech perception and intelligibility after paediatric cochlear implantation. *British Journal of Audiology, 34*, 257-264.

Archbold, S.M., Nikolopoulos, T., Lutman, M.E., & O'Donoghue, G.M. (2002). The educational settings of profoundly deaf children with cochlear implants compared with age-matched peers with hearing aids: implications for management. *International Journal of Audiology, 41*, 157-161.

Arnoldner, C., Baumgartner, W.D., Gstoettner, W., & Hamzavi, J. (2005). Surgical considerations in cochlear implantation in children and adults: a review of 342 cases in Vienna. *Acta Oto-Laryngologica, 125*, 228-234.

Battmer, R.D., O'Donoghue, G.M., & Lenarz, T. (2007). A multicenter study of device failure in European cochlear implant centers. *Ear and Hearing, 28,* 95S-99S.

Berg, A.L., Ip, S.C., Hurst, M., & Herb, A. (2007). Cochlear implants in young children: informed consent as a process and current practices. *American Journal of Audiology, 16*, 13-28.

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. *Otology and Neurotology, 25,* 730–739.

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 International, 8*, 53–67.

Berg, A. L., Ip, S. C., Hurst, M. & Herb, A. (2007). Cochlear implants in young children: Informed consent as a process and current practices. *American Journal of Audiology, 16,* 13-28.

Bergeron, F. (2000). Residual hearing following cochlear implantation: Effect of time and device. *Advances in Otorhinolaryngology, 57*, 389-92.

Boggess, W. J., Baker, J. E., & Balkany, T. J. (1989). Loss of residual hearing after cochlear implantation. *Laryngoscope, 99,* 1002-1005.

Brown, K.D. & Balkany, T.J. (2007). Benefits of bilateral cochlear implantation: a review, *Current Opinions in Otolaryngology, Head and Neck Surgery, 15*, 315-8.

Canadian Association of the Deaf (2007). CAD's position on cochlear implants. Retrieved from http://www.cad.ca/en/issues/cochlear_implants.asp on January 25, 2009

Canadian Institute of Health Research (2008). Knowledge Translation Overview. Retrieved from http://www.cihr.gc.ca/e/29418.html on August 20, 2008

Candidacy Criteria. (2008). Retrieved February 9, 2008 from http://www.cochlear.com/Support/631.asp

Charles, C, Whelan, T., & Gafni, A. (1999). What do we mean by partnership in making decisions about treatment? *British Medical Journal, 319*, 780-782.

Ching, T.Y., van Wanrooy, E. & Dillon, H. (2007). Binaural-bimodal fitting or bilateral implantation for managing severe to profound deafness: a review, *Trends in Amplification, 11*, 161-92.

Cochrane Collaboration, The, (2004). Search of reviews and website conducted on September 29, 2004 at http://www.cochrane.org.

Cochrane Inventory of Patient Decision Aids. (2008). Retrieved June 10, 2008 from http://decisionaid.ohri.ca/cochinvent.php

Cohen, N. L., Hoffman, R. A., & Stroschein, M. (1988). Medical or surgical complications related to the Nucleus multichannel cochlear implant. *Annals of Otology, Rhinology, and Laryngology Supplemental, 135*, 8-13.

Colburn, H.S., Shinn-Cunningham, B., Kidd, G. & Durlach, N. (2006). The perceptual consequences of binaural hearing, *International Journal of Audiology, 45*, S34-S44.

Coulter, A. (1997). Partnerships with patients: The pros and cons of shared decision making. *Journal of Health Services Research Policy, 2,* 112-121.

Crouch, R. (1997). Let the deaf be deaf: reconsidering the use of cochlear implants in prelingually deaf children. *The Hastings Center Report, 27*, 14-21.

Cunningham, C.D., Slattery, W.H., & Luxford, W.M. (2004). Postoperative infection in cochlear implant patients. Otolaryngology - Head & Neck Surgery 131: 109-114.

Eddy, D. (1992). *A manual for assessing health practices and designing practice policies: the explicit approach*. Philadelphia, American College of Physicians.

Fayad, J. N., Wanna, G. B., Micheletto, J. N., & Parisier, S. C. (2003). Facial nerve paralysis following cochlear implant surgery. *Laryngoscope, 113*, 1344-1346.

Fina, M., Skinner, M., Goebel, J. A., Piccirillo, J. F., Neely, J. G., & Black, O. (2003). Vestibular dysfunction after cochlear implantation. *Otology and Neurotology, 24,* 234-242.

Fortnum, H. M., Summerfield, A. Q., Marshall, D. H., Davis, A. C., & Bamford, J. M. (2001). Prevalence of permanent childhood hearing impairment in the United Kingdom and implications for universal neonatal hearing screeing: Questionnaire based ascertainment study. *British Medical Journal, 8*, 536-540.

Gantz, B.J., Tyler, R.S., Woodworth, G.G., Tye-Murray, N., & Fryauf-Bertschy, H. (1994). Results of multichannel cochlear implants in congenital and acquired prelingual deafness in children: five-year follow-up. *American Journal of Otology, 15S*, 1-7.

Geers, A. & Brenner, C. (2003). Background and educational characteristics of prelingually deaf children implanted by five years of age. *Ear and Hearing, 24*, 2S-14S.

Gordon, K.A., Daya, H., Harrison, R.V., Papsin, B.C. (2000). Factors contributing to limited open-set speech perception in children who use a cochlear implant. *International Journal of Pediatric Otorhinolaryngology, 56*, 101-111.

Gregoret, L. (2003). Bilateral cochlear implants: are they in your child's future?. *Volta Voices, 10*, 10-15, 34.

Gysin, C., Papsin, B. C., Daya, H., & Nedzelski, J. (2000). Surgical outcomes after paediatric cochlear implantation: diminution of complications with the evolution of new surgical techniques. *Journal of Otolaryngology, 29*, 285-289.

Harrison, R.V., Panesar, J., El-Hakim, H., Abdolell, M., Mount, R.J., & Papsin, B. (2001). The effects of age of cochlear implantation on speech perception outcomes in prelingually deaf children. *Scandinavian Audiology, 30*, 73-78.

Health Canada (2001). *The Population Health Template: Key Elements and Actions That Define A Population Health Approach*. Retrieved February 2, 2009 from http://www.phac-aspc.gc.ca/ph-sp/pdf/discussion-eng.pdf

Health Canada (2005). What is the population health approach? Retrieved September 7, 2005 from http://www.phac-aspc.gc.ca/ph-sp/phdd/approach/approach.html#health

Holmes-Rovner, M., Llewellyn-Thomas, H., Entwistle, V., Coulter, A., O'Connor, A., & Rovner, D.R. (2001). Patient choice modules for summaries of clinical effectiveness: a proposal. *British Medical Journal, 322,* 664-667.

Incesulu, A., Vural, M., & Erkam, U. (2003). Children with cochlear implants: Parental perspective. *Otology & Neurotology, 24,* 605-611.

International Patient Decision Aid Standards. (2008). What are decision aids? Retrieved June 10, 2008 from http://ipdas.ohri.ca/what.html

James, A.L., & Papsin, B.C. (2004). Cochlear implant surgery at 12 months of age or younger. *Laryngoscope, 114*, 2191-2195.

Jensen, J.K. & Neff, D.L. (1993). Development of basic auditory discrimination in preschool children. *Psychology Science, 4,* 104-107.

Kandogan, T., Levent, O., & Gurol, G. (2005). Complications of paediatric cochlear implantation: experience in Izmir. *Journal of Laryngology & Otology, 119*, 606-610.

Kluwin, T. N., & Stewart, D. A. (2000). Cochlear implants for younger children: A preliminary description of the parental decision process and outcomes. *American Annals of the Deaf, 145,* 26-32.

Li, Y., Bain, L., & Steinberg, A. G. (2004). Parental decision-making in considering cochlear implant technology for a deaf child. *International Journal of Pediatric Otolaryngology, 68,* 1027-1038.

Lin, F.R. & Niparko J.K. (2006). Measuring health-related quality of life after pediatric cochlear implantation: a systematic review. *International Journal of Pediatric Otorhinolaryngology, 70*,1695-1706.

Litovsky, R.Y., Johnstone, P.M., Parkinson, A., Peters, R. & Lake, J. (2004). Bilateral cochlear implants in children. *International Congress Series*, 1273, 451-454.

Litovsky, R.Y., Parkinson, A., Arcaroli, J., Peters, R., Lake, J. et al. (2004). Bilateral cochlear implants in adults and children, *Archives of Otolaryngology, Head and Neck Surgery, 130*, 648-55.

Logan, J. & Graham, I.D. (1998). Toward a comprehensive interdisciplinary model of health care research use. *Science Communication, 20*, 227-246.

Lomas, J. (1997). Improving research dissemination and uptake in the health sector: beyond the sound of one hand clapping, McMaster University Centre for Health Economics and Policy Analysis. Policy Commentary C97-1.

Martin, F.N., & Clark, J.G. (1996). *Hearing Care for Children.* Needham Heights, MA: Allyn & Bacon.

Maurer, J., Marangos, N., & Ziegler, E. (2005). Reliability of cochlear implants. Otolaryngology, *Head and Neck Surgery, 132*, 746–750.

Mehl, A. L. & Thompson, V. (2002). The Colorado newborn hearing screening project, 1992-99: On the threshold of effective population-based newborn hearing screening. *Pediatrics, 109.* E7.

Mencher, G.T. & Davis, A. (2006). Bilateral or unilateral amplification: is there a difference? A brief tutorial, *International Journal of Audiology, 45,* S3-S11.

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 International, 7,* 194-201.

Moog, J. S., & Geers, A. E. (2003). Epilogue: Major findings, conclusions and implications for deaf education. *Ear and Hearing, 24*, 121S-125S.

Moses, K. & Van Hecke-Wulatin, M. (1981). The socio-emotional impact of infant deafness: A counseling model, in *Early Management of Hearing Loss*, G. Mencher and S. Gerber ed. Grune and Stratton, 243-278.

Most, T., & Zaidman-Zait, A. (2003). The needs of parents of children with cochlear implants. *The Volta Review, 103*, 99-113.

Murphy, J. & O'Donoghue, G. (2007). Bilateral cochlear implantation: an evidence-based medicine evaluation, *Laryngoscope*, *117*, 1412-18.

National Association of the Deaf (2000). Position statement on cochlear implants. Retrieved online at http://www.nad.org/site/pp.asp?c=foINKQMBF&b=138140 on September 29, 2004.

Neuss, D. (2006). The ecological transition to auditory-verbal therapy: Experiences of parents whose children use cochlear implants. *Volta Review, 106,* 195-222.

Nevins, M. & Chute, P. (1996). *Children with cochlear implants in educational settings*. San Diego, CA: Singular Publishing.

Nikolopoulos, T., Dyar, D., Gibbin, K. P. (2004). Assessing candidate children for cochlear implantation with the Nottingham Children's Implant Profile (NChIP): the first 200 children. *International Journal of Pediatric Otorhinolaryngology, 68,* 127-135.

Northern, J., & Downs, M. P. (1991). *Hearing in children* (4th ed.). Baltimore, MD: Williams & Wilkings.

O'Connor, A. M. (1995). Validation of a decisional conflict scale. *Medical Decision Making, 15,* 25-30.

O'Connor, A., & Edwards A. (2001). The role of decision aids in promoting evidence-based patient choice. In A. Edwards, G. Elwyn (Eds.), *Evidence-based patient choice* (pp. 220-42). Oxford, UK: Oxford University Press.

O'Connor, A. M., Legare, F., & Stacey, D. (2003). Risk communication in practice: the contribution of decision aids. *British Medical Journal, 327*, 736-740.

O'Connor, A.M., Stacey, D., Entwistle, V., Llewellyn-Thomas, H., Rovner, D., Holmes-Rovner, et al. (2002). Decision aids for people facing health treatment or screening

decisions (Cochrane Review). Retrieved on July 18, 2008 from http://www.cochrane.org/reviews/en/ab001431.html

O'Connor, A. M., Tugwell, P., Wells, G. A., Elmslie, T., Jolly, E., Hollingworth, G., et al. (1998). A decision aid for women considering hormone therapy after menopause: Decision support framework and evaluation. *Patient Education and Counseling, 33*, 267-279.

Parisier, S.C., Chute, P.M., Popp, A.L., & Suh, G.D. (2001). Outcome analysis of cochlear implant reimplantation in children. *Laryngoscope, 111*, 26-32.

Peters, E. (2000). Our decision on a cochlear implant. *American Annals of the Deaf, 145*, 263-267.

Postelmans, J.T.F., Cleffken, B., & Stokroos, R.J. (2007). Post-operative complications of cochlear implantation in adults and children: Five years' experience in Maastricht. *The Journal of Laryngology and Otology, 121*, 318-323.

Reamy, C.E. & Brackett, D. (1999). Communication Methodologies. *Otolaryngologic Clinics of North America, 32*, 1103-1116.

Robinson, K. (1999). Evidence-based medicine and its implications for audiological science. *British Journal of Audiology, 33*, 9-16.

Sach, T. H., & Whynes, D. K. (2005). Paediatric cochlear implantation: The views of parents. *International Journal of Audiology, 44,* 400-407.

Samson-Fang, L., Simons-McCandless, M., & Shelton, C. (2000). Controversies in the field of hearing impairment: early intervention, educational methods, and cochlear implants. *Infants and Young Children, 12*, 77-88.

Schachter, H. M., Clifford, T.J., Fitzpatrick, E., Eatmon, S., MacKay, M., Johnston, C. J., Sampson, M., & Moher, D. (2002). *Systematic review of interventions for hearing loss in children.* (Health Canada, Contract # 030SS.HT091-01001). Ottawa, Ontario.

Schafer, E.C. & Thibodeau, L.M. (2006). Speech Recognition in noise in children with cochlear implants while listening in bilateral, bimodal, and FM-system arrangements, *American Journal of Audiology, 15*, 114-26.

Shafir, D.N. (2007). Study researches bilateral cochlear implants, *ASHA Leader, 5*, 19.

Sorkin, D. L., & Zwolan, T. A. (2008). Parental perspectives regarding early intervention and its role in cochlear implantation in children. *Otology and Neurootology, 29*, 137-141.

Sox, H. (1999). What is a good decision?, *Effective Clinical Practice, 2*, 196-197

Steinberg, A., Brainsky, A., Bain, L., Montoya, L., Indenbaum, M., & Potsic, W. (2000).

Parental values in the decision about cochlear implantation. *International Journal of Pediatric Otorhinolaryngology, 55,* 99-107.

Stratigouleas, E.D., Perry, B.P., King, S.M., & Syms, C.A. (2006). Complication rate of minimally invasive cochlear implantation. *Otolaryngology - Head & Neck Surgery, 135*: 383-386.

Thompson, D.C., McPhillips, H., Davis, R.L., Lieu, T.A., Homer, C.J., & Helfand, M. (2001). Universal newborn hearing screening: Summary of evidence. *Journal of the American Medical Association, 286*, 2000-2010.

Thoutenhoofd, E., Archbold, A., Gregory, S., Lutman, M., Nikolopoulos, T., & Sach, T.H. (2005). *Paediatric Cochlear implantation: Evaluating outcomes*. London: Whurr.

United States Food and Drug Administration (1990). Devices at FDA: Nuleus 22 Channel Cochlear Implant System in Children. http://www.accessdata.fda.gov/scripts/cdrh/devicesatfda/index.cfm?db=PMA&id=7987 [August 6, 2008].

United States Food and Drug Administration (2006). FDA Public Health Notification: Continued Risk of Bacterial Meningitis in Children with Cochlear Implants with a Positioner Beyond Twenty-Four Months Post-Implantation. http://www.fda.gov/cdrh/safety/020606-cochlear.html [January 19, 2009].

van Dijk, J.E., van Olphen, A.F., Langereis, M.C., Mens, L.H., Brokx, J.P., & Smoorenburg, G.F. (1999). Predictors of cochlear implant performance, *Audiology, 38*, 109–116.

Venail, F., Sicard, M., Piron, J.P., Artiered, F., Uziel, A., & Mondain, M. (2007). Cochlear implantation safety and reliability in paediatric cases. *Audiological Medicine, 5*, 210-217.

Weise, J.B., Muller-Deile, J., Brademann, G., Meyer, J.E., Ambrosch, P., & Maune, S. (2005). Impact to the head increases cochlear implant reimplantation rate in children. Auris *Nasus Larynx, 32,* 339-343.

Yu, K.C.Y., Hegarty, J.L., Gantz, B.J., & Lalwani, A.K. (2001). Conservative management of infections in cochlear implant recipients. *Otolaryngology-Head and Neck Surgery, 125,* 66-70.

Wennberg, J. E. (2002). Unwarranted variations in healthcare delivery: Implications for academic medical centres. *British Medical Journal, 325*, 961-964.

Werner, L.A. & Gray, L. (1998). Behavioral studies of hearing development. In E.W. Rubel, A.N. Popper, & R.R. Fay. (Eds.) *Development of the Auditory System* (pp.12-72.). New York: Springer-Verlag.

Zaidman-Zait, A., & Jamison, J. (2004). Searching for cochlear implant information on the

internet maze: Implications for parents and professionals. *Journal of Deaf Studies and Deaf Education, 9,* 413-426.

Zurek, P.M. (1993). Binaural advantages and directional effects in speech intelligibility. In: Studebaker GA, Hochberh I, eds. *Acoustical Factors Affecting Hearing Aid Performance*. 2nd ed. Boston, MA: Allyn & Bacon; 1993:255-276.

CHAPTER 2

An Assessment of Parents' Decision-Making Regarding Paediatric Cochlear Implants

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An Assessment of Parents' Decision-Making Regarding Paediatric Cochlear Implants

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Abstract

Parents of children with severe to profound hearing loss have to make a number of fundamental decisions for their children. These decisions include communication and amplification options. In particular, the parents must decide whether and when their child will receive cochlear implants, and whether these will be implanted unilaterally or bilaterally. The objective of this study was to describe the decision-making needs of parents making the cochlear implant decision for their children. Semi-structured interviews were conducted with eight parents and eight cochlear implant team members at a Canadian cochlear implant centre to document parental and clinician recollections and opinions of the decision-making process related to a unilateral or bilateral cochlear implantation. The results demonstrated that the decision to go ahead with a cochlear implant was consistently based on the parents' preferences for spoken communication for their children. Parents reported satisfaction with the cochlear implant decision-making process. Two of eight parents felt that additional information on unilateral cochlear implantation risks and benefits should have been provided. Four of eight parents described how more information on the experiences of other families would have been helpful for their decision. Parental and clinical perceptions of the bilateral implantation decision were highly variable. All parents stated that additional information on bilateral cochlear implantation was needed. Based on the results of the interviews, it is concluded that there are needs for information on and resources, especially for bilateral cochlear implantation decision-making.

Key Words: Hearing Loss, Cochlear Implants, Needs Assessment, Decision Making, Audiology

Almost immediately after the diagnosis, the parents of children with bilateral, permanent, sensorineural hearing loss are required to make a number of fundamental decisions regarding the communicative rehabilitation of their child. These decisions involve the use of the choice of amplification or cochlear implantation, and the communication approach for their child. Most of these parents have never experienced hearing loss, which may make the decisions more difficult and daunting (Northern & Downs, 1991). Their lack of knowledge regarding hearing loss, options for communication, and technologies for rehabilitation can be overwhelming for parents. They must absorb significant amounts of technical and scientific information during a period of grief about their child's hearing loss (Anagnostou, Graham, & Crocker, 2007; Kurtzer-White & Luterman, 2003). While the treatment team can provide parents with necessary information, the actual decision-making process is usually invisible to the professionals. A better understanding of the parents' process for decision-making may allow the cochlear implant team to reduce some of the parental stress and anxiety during this delicate and emotional time.

Family decisions about treatment vary depending on the severity and characteristics of the child's hearing loss. A child with any significant degree of bilateral hearing loss usually requires specialized early interventions in order to develop oral language (Samson-Fang, Simons-McCandless, & Shelton, 2000). A child with a severe to profound hearing loss may require considerable intervention in the form of amplification and aural rehabilitation in order to develop functional spoken communication. For these children, cochlear implants (CIs) are one of the available options. The criteria for CI use in children with significant hearing loss have expanded considerably since the initial approval of the device by the American Food and Drug Administration in 1990 (Candidacy Criteria, 2008). Originally

used in older children with profound hearing loss, now children one year of age and even younger with severe to profound losses are routinely eligible for CIs (Thoutenhoofd et al., 2005). Because binaural hearing is important for sound localization and speech intelligibility in noise, bilateral implantations have become common in some paediatric centres (Berg, Ip, Hurst, & Herb, 2007).

When parents are considering cochlear implantation for their child, they are interested in the medical, speech and language, educational, and social outcomes of other users of the device. A systematic review of the effectiveness of unilateral paediatric cochlear implants reports that there are consistent benefits for children who use CIs rather than hearing aids in terms of hearing sensitivity levels and speech perception (Thoutenhoofd, et al., 2005). A recently published, multi-site study shows greater improvements in the language of children using CIs as compared to earlier evaluations of children using hearing aids (Moog & Geers, 2003). The evidence is less clear-cut regarding the relative benefits of CIs over hearing aids for children with residual hearing and children with comorbidities or congenital syndromes (Thoutenhoofd et al., 2005). Practices for the latter populations vary across CI centres.

There are risks associated with the CI surgery that may influence parental decision-making. Early studies estimated that 18% of cochlear implant surgeries were accompanied by some type of minor or major complication (Cohen, Hoffman, & Stroschein, 1988). The current estimates suggest that major complications range from 3 to 4% of CI surgeries (Tambyraja, Gutman, & Megerian, 2005). One major risk is the post-surgical complication of meningitis among children who have received an implant. Recent work has attributed the increased risk of meningitis, in part, to a particular positioner device that has since been withdrawn from the market (U.S. Food and Drug Administration, 2007; Biernath, et al.,

2006). Vaccinations to prevent meningitis continue to be recommended for the entire CI recipient population. Facial nerve paralysis, vestibular problems, and risks associated with the use of a general anaesthetic are some of the other complications of CI surgery (Fayad, Wanna, Micheletto, & Parisier, 2003; Fina et al., 2003; Gysin, Papsin, Daya, & Nedzelski, 2000). Finally, once the CI surgery is undertaken, there is a high risk of losing the residual hearing in that ear (Bergeron, 2000; Boggess, Baker, & Balkany, 1989). Therefore, the parental decision to use a CI is typically irreversible.

Bilateral implants have recently become available in many paediatric cochlear implant centres, although not yet universally in Canada. The research indicates that there are benefits for patients receiving bilateral stimulation compared to the use of a single CI, demonstrated on measures of speech recognition in noise and sound localization (Brown & Balkany, 2007; Ching, van Wanrooy,. & Dillon, 2007; Murphy & O'Donoghue, 2007; Schafer & Thibodeau, 2006). New guidelines for patient selection and other position papers have also recently been published (William House Cochlear Implant Study Group, 2008; Perreau, Tyler, Witt, & Dunn, 2007). The addition of the bilateral implantation option further complicates the parental and clinical decision-making process. Recent audiology and otolaryngology literature has discussed the need for additional evidence of bilateral CI effectiveness above and beyond the improved speech recognition in noise and sound localization (Berg et al., 2007; Gregoret, 2003).

Parental Cochlear Implant Decision-Making

Publications from around the world have described the challenges parents face in deciding on cochlear implantation for their children (Sorkin & Zwolan, 2008; Sach & Whynes, 2005; Li, Bain, & Steinberg, 2004; Incesulu, Vural, & Erkam, 2003; Most &

Zaidman-Zait, 2003; Peters, 2000; Steinberg, et al., 2000). Incesulu et al. (2003) report that 81% of parents responding to a survey indicated that the CI decision was the most difficult aspect of the implantation process for them. Most and Zaidman-Zait (2003) also describe the high parental stress during the implantation decision-making process and the specific parental needs for information to aid in the process.

In contrast, Sach and Whynes (2005) report that most of the 216 families interviewed in the UK found the decision regarding implantation to be straightforward. They did, however, describe the overall stress for families undergoing cochlear implantation. A very recent survey of parents in the US indicates that those who chose the CI for their child felt that they lacked "comprehensive and bias-free" information when making the decision (Sorkin & Zwolan, 2008).

The medical decision-making literature makes a distinction between preference-sensitive and effective decisions (Wennberg, 2002). In medical decision-making, a decision is considered preference-sensitive when the available evidence indicates that there are several available choices that carry both harms and benefits. In such a scenario, the personal beliefs and preferences of the patient may affect his or her perception of the relative weight of the harms and benefits of an intervention. The patient's care must therefore acknowledge these preferences (O'Connor, Legare, & Stacey, 2003; Wennberg, 2002). This is in contrast to effective care. In an effective care scenario, the benefits of a treatment clearly outweigh possible harmful treatment effects. Based on findings in the pertinent literature, the CI decision appears to be preference-sensitive.

When individuals are faced with making preference-sensitive decisions, they can experience increased decisional conflict. Decisional conflict is the state of uncertainty about the best course of action (O'Connor, 1995). Previous CI studies have not referred to, nor

measured, the decisional conflict in parents making the CI decision. They also have not contextualized the CI decision within the broader medical decision-making literature.

The literature indicates that there is variability in the decision-making process across geographical regions, cultural backgrounds, and CI centres (Sorkin & Zwolan, 2008; Sach & Whynes, 2005; Li, Bain, & Steinberg, 2004; Incesulu, Vural, & Erkam, 2003; Most & Zaidman-Zait, 2003; Peters, 2000; Steinberg, et al., 2000). However, even within an individual CI centre, families may experience very different forms and levels of decisional conflict and emotional stress. The reported stress may be due to uncertainty about possible risks and benefits. The parents may be uncertain or conflicted in their values regarding communication approaches (e.g., oral or sign language) that may be linked to the CI decision. They may feel that they have inadequate information about their options, or feel under pressure from clinicians or other family members. Having a better understanding of the CI decision-making process may identify a way to reduce parental stress during this process or to meet any specific information needs that are identified. There is currently no available literature on parental perceptions of the bilateral CI decision.

Purpose

The present study was undertaken to investigate the decision-making process and the needs of parents regarding unilateral and bilateral CIs. The research objectives were to explore:

(a) The parental and clinician perceptions of the unilateral and bilateral decisions: How did parents and clinicians perceive different options with regards to their respective advantages and disadvantages?

(b) The parents' and clinicians' perceptions of their knowledge, values and

expectations, as well as the support and resources available to them during CI decision-making.

(c) The parents' recollections of the manifestations of decisional conflict and their contributing factors during the decision-making process.

(d) The need for a formal decision aid to support parents and clinicians in the cochlear implant decision-making process.

Framework

The Ottawa Decision Support Framework (ODSF; O'Connor et al., 1998) was chosen as the framework to guide the needs assessment in the present study. A schematic overview of the ODSF is presented in Figure 1. This framework is appropriate for decisions that "(1) are stimulated by a new circumstance, diagnosis, or developmental condition, (2) require careful deliberation because of the uncertain and/ or value-sensitive nature of the benefits and risks, and (3) need relatively more effort in the deliberation stage than the implementation stage" (O'Connor et al., 1998, p.268). The paediatric cochlear implementation decision meets each of these criteria. The ODSF depicts how a family's decisional needs and decisional quality influence each other. Decisional needs include (a) elements of the decision, such as timing, stage, and leaning, (b) decisional conflict, (c) knowledge and expectations, and (d) values. Decision support can be used to address decisional needs to improve the quality of decisions.

Method

Participants and Recruitment

A sample of parents at various stages of decision-making were recruited. Eligible participants included parents whose children were (a) were currently assessed for CI

candidacy; (b) were awaiting surgery; or c) had undergone surgery within the last 2 years, and had used their implants for at least 6 months. All families had to speak and understand English because the interviews were conducted in English.

The hospital CI clinicians were also invited to participate. The potential participants included audiologists, rehabilitation therapists, a psychologist, a social worker, and a CI surgeon. Consent for participation was obtained from each participant prior to study commencement. Ethical approval for the study was received from the Children's Hospital of Eastern Ontario and the University of Ottawa, Research Ethics Boards.

Interview Procedure

A semi-structured interview guide was developed based on the standard needs assessment questions of Jacobson and O'Connor (2006). The open-ended questions were guided by the ODSF. Interview questions for parents and professionals focused on (a) reactions and decisions surrounding the identification of their child's hearing loss, (b) the options available to them, (c) the perceived benefits and risks associated with their options, (d) manifestations of decisional conflict (uncertainties), (e) knowledge and expectations, (f) values, (g) support and resources including usual roles in decision making, (h) patient characteristics such as age of identification and aetiology, (i) barriers and facilitators in receiving decision support, and (j) potential strategies for overcoming barriers. A copy of the interview guide used with parents can be found in Appendix B. While there was only a single open-ended question on bilateral implantation, this sparked considerable discussion and additional follow-up questions were asked depending on parent responses.

The parent interviews lasted between 30 and 60 minutes and were conducted at a mutually agreeable location, either in the parents' home or at the clinic. The clinician

interviews lasted about 30 minutes and were held at the clinic. The interviews were audio recorded and transcribed. The clinical characteristics of the children undergoing the CI implantation were obtained during the interview.

Analysis and Interpretation

A mixed methods approach was used in analysing the data. This approach seeks to use both qualitative and quantitative research methods to answer research questions (Tashakkori & Teddlie, 2003). The interview data from the parents and clinicians were analyzed together. Frequencies and counts were used where appropriate to describe structured, quantifiable responses that corresponded to the answer templates in the interview guide. A deductive coding strategy based on the ODSF was used to analyze the content of the open-ended responses. Similar items were grouped together based upon the elements in the ODSF. Nuances in the responses were qualitatively explored based on the clinical characteristics, such as the child's age at the diagnosis of the hearing loss, the aetiology, the presence of co-existing health issues, and the current status in the implantation process (pre- or post-implant). Due to the exploratory nature of the research and the small sample size, no statistical analysis could be undertaken to formally quantify the effect of these factors on parental responses.

Results

Characteristics of Participants

Seven families participated in the interviews. From these seven families, eight parents or guardians of eight children took part in the study. Four children had already received a unilateral CI, one had received bilateral implants, and three were awaiting their surgery for a unilateral implant. At the time of the interview, the children were between 1 and 5 years of

age. Two of the eight children had co-existing health issues at the time of diagnosis. One was recovering from meningitis and the other had a congenital health concern. Half of children were candidates for CIs upon diagnosis and the remaining four children had hearing losses that progressed to make them CI candidates. Two of the children had auditory neuropathies. Four of the children were only children. Two of the children had a sibling with hearing loss in a family of two children. Two of the children were the only child with a hearing loss in a family of two children.

One of the children was identified with a hearing loss after 18 months of age following medical referral, one child had meningitis as an infant, and six were identified through newborn hearing screening programs. All children used auditory-verbal therapy (AVT) as their primary communication approach. An effort was made to seek out families who had declined CI surgery. However, the families who were identified declined participation in the interview. Eight of the ten CI team members participated in individual interviews. The CI team members came from a range of disciplines with a wide range of experience in CI.

Identification of Hearing Loss and Early Decision Making

The responses of parents and guardians to the identification of hearing loss varied depending on the co-occurrence of other health issues at the time. Parents of children ($N = 6$) with no co-occurring health issues described the uncertainty and shock associated with the diagnosis:

> We were shocked. It was very painful. We just couldn't believe it. My wife was crying. It was a horrible experience. [parent of 3-year-old]

> At the beginning, when we found out about our child it was really hard for us. We didn't know what to do and where to go and how things were going to work for him in the future. We didn't know anything about if he's going to go for sign language or going to go for only hearing aids, or that. We didn't know anything. We didn't know what the hell's going on. [parent of 1-year-old child]

Parents who had children that suffered from meningitis and postnatal health problems (N = 2) described less shock at the identification of hearing loss than parents of children without co-occurring health concerns:

> Because he had other health issues at birth, I guess we kind of took it as a grain of salt. We were just really grateful that he made it through because he wasn't expected to live, and I figure if he had to have some sort of incapacity, I'd prefer the hearing to the eyesight. So I don't think we were ever in shock about it. I don't remember being in shock, anyway. [parent of 4-year-old child]

Parents and clinicians were asked to describe some of the decisions that had to be made following the identification of their child's hearing loss. Both groups identified the communication approach as the first decision that parents have to make. The parents made a distinction between using an aural/ auditory-verbal approach or sign language with their child. They did not describe struggling with the communication approach decision and all chose an auditory-verbal approach for their children. Other decisions named by both parents and clinicians included whether to use a hearing aid, the decision to undergo cochlear

implantation, the type of implant or manufacturer to choose, when to proceed with the implantation, and the decision for a parent to return to work or stay at home to teach their child.

Introduction of cochlear implants

When asked when and how the topic of CIs was first introduced to them, the parents provided varied answers. The parents of the four children who were initially diagnosed with profound hearing loss stated that the topic had been introduced at the time the diagnosis was shared. The parents with the four children with progressive hearing loss stated that the topic had been introduced a year or later after the initial diagnosis. Some parents had felt shocked when the clinicians had suggested a CI while others had been relieved:

> It actually came as a bit of a surprise to us because [my child] now wears a hearing aid and a cochlear implant, and so he had both ears equipped with hearing aids, and he was making progress, and we were getting language… So it was a little of a set-back emotionally.[parent of 3-year-old child]

> I kind of just heard from other parents in the department, like seeing them in the waiting room, and chatting about it. Their kids may have had hearing aids but now they had a cochlear implant, and now it's much better. So, we had a positive image right away from that because the parents were like, "Oh, yeah. No more feedback, no more… you know… they can hear so much better." Well, I think as his hearing started getting worse, we felt frustrated, so when it was first brought up with us, I think we kind of felt happy because, in a way, it was not… we weren't happy that it

was getting low like that, but we were happy that we were going to have another option because we were getting frustrated. [parent of 2-year-old child]

The parents and clinicians reported that the professions most likely to be involved in first discussing the CI with families were the audiologists and auditory-verbal therapists working with their children. Five out of the eight parents received information from other families in the clinic waiting room and the internet before discussing the CI with their clinicians.

Options available

When asked about their options regarding the CI decision, half of the parents perceived their decision as a choice between a CI and hearing aids. Of these parents, all had children with hearing losses that had progressed from severe to profound over time. The other half of parents perceived their decision as a choice between a CI and sign language. These parents had children with profound losses as a result of genetic losses, auditory neuropathies, and meningitis. The clinicians varied little in their perceptions of the options.

Five of the eight clinicians perceived parental decisions as a choice among three options: CI, hearing aids, or sign language. One clinician perceived the two options: CI or hearing aids. The remaining two clinicians perceived two different options: CI or sign language.

Perceived advantages and disadvantages of options

After identifying the available options, the parents and clinicians were asked to list some of the perceived advantages and disadvantages of each. Table 1 provides a summary of

the advantages and disadvantages that were generated by the parents and clinicians. All parents reported that the CI option was most consistent with their family's communication culture and linguistic backgrounds.

The clinicians' perceptions of parents' choices were consistent with parent's views. This consistency between the CI choice and the families' communication culture was the perceived benefit of the CI option:

> Most of the hearing impaired children are born into hearing families where spoken language is the language of the home whether or not there are other children. So, in terms of ease of natural language simulation in most families it would come through speaking. So, I mean, there would be an understanding in sign language that the parents are going to be learning a new language system. And then, also hopefully, to have other friends and extended family getting involved with communication systems as well. If sign language is the language at home for a particular family, I would think that that would be the natural option for those families. [clinician]

Two of the eight parents commented on their qualms making a decision for their child in light of the uncertainty that their child might later disagree with their decision. However, they felt that they were making the best decision for their family at the present time:

> We had concerns in having to make a decision for a child who might have made a different decision later... It is always possible later on to have the devices removed if she chose that later, and we wouldn't have had the opportunity, necessarily, to have

> the same opportunity to get [speech and language] results. [parent of 2-year old child]

In this study, most families found it difficult to perceive any benefits of sign language. All families had chosen AVT as their primary communication approach and were enrolled in a program. Seven parents reported that sign was not a fit for their family. However, one parent wished that their family had the option to communicate to their child in sign language. However, the parent realized that this would have been inconsistent with the AVT philosophy. The benefits regarding sign language that are listed in Table 1 were all derived from clinicians' interview data.

Manifestations of decisional conflict

The responses to the structured question on feelings during the CI decision-making process are summarized in Figure 2. The parents were also given an opportunity to expand and comment further.

The parents reported feeling most concerned about what could go wrong: "They make a hole in the bone, so there's no protection here only more... only that piece of equipment there." Another concern was that their child would not benefit sufficiently from the cochlear implant: "I was worried that it wouldn't work. I was really worried but at the same time that wasn't something that would stop us from trying the cochlear implant."

Parents did not report delaying the decision, wavering between choices, feeling uncertain about what was important, or dwelling excessively on the decision. All parents emphasized that they were confident that the CI decision was the appropriate decision for their family:

> Even though I didn't grow up with anyone with a hearing loss I knew it wasn't something I wanted my son to do. I want to hear the words, "I love you mom," I want to be able to just communicate and be able to tell him when his back is to me, "Can you go get your shoes?" [parent of three-year-old]

Factors contributing to decisional conflict

When asked which factors had contributed to decisional conflict, neither parents nor clinicians felt that parents were unclear about what was important to them or that they lacked the skills to make the CI decision. Figure 3 presents the responses to the structured question. The parents and clinicians responded similarly on most items. However, four of the eight parents felt that they had lacked information on the choices that other families had made regarding CIs. Only one clinician of eight perceived this as a possible gap in the information provided to families. In general, the clinicians reported that they consistently linked families with each other to provide mutual support and share information. One of the interviewed parents commented as follows:

> We wanted to know how it worked for other people, and we weren't that well connected to a lot of families. I had requested all along to be connected with families, and that never happened. I did it on my own. And I think that's one thing that every family should have that ability to connect very quickly, and it took us a while. [parent of 2-year old]

Clinicians and parents also varied slightly on their perception of pressure in decision-making. Parents did not report feeling pressure to choose the CI option. Four of the eight

clinicians reported that parents might feel pressure to make the decision to go ahead with cochlear implantation:

> We never pushed the parents into getting the implant, but it depends on how that's worded. You know, and I'm not there to see the [other team members], how they actually provide the info, but there might be a bit of solid pressure. [clinician]

Perception of Others' Opinions, Practices, Support, and Pressures

Parent participants reported that the individuals most likely to be involved in the CI decision were the audiologists, auditory-verbal therapists, and the CI surgeon. The parents valued the team approach to the CI process. Individuals outside of the CI team were not named as stakeholders in the decision-making. When asked about the influence that extended family members might have on the process, most parents said that there was little such influence.

When clinicians and parents were asked to describe the decision-making dynamic that they had or were experiencing, both parents and clinicians reported equally that it was either a shared decision or that the clinic team members provided support for them to make the decision themselves. No parent reported that the decision was made for them by clinicians.

Resources in the Decision-Making Process

Families and clinicians were asked about the type of information they accessed to make a decision and which additional resources they perceived as potentially beneficial. Most families felt that the clinicians provided them with adequate information on the

treatment options and the associated risks and benefits. The parents also commented on their need to seek additional information on their own to supplement the information from the clinic. This was often accomplished through the internet or by meeting families who had already experienced the CI process. Five parents reported meeting with other families before they made their decision or before they had the surgery. They all spoke highly of this contact with other families: "If I hadn't talked to those families I did talk to, I would have felt at a real loss for not knowing things. And having that ability to contact them was huge." The other three parents did not have the opportunity to meet other families. They indicated that they would have liked to have the interactions with others who have made similar decisions for their children.

Meeting with the surgeon to hear about the risks and benefits of the CI surgery was also mentioned as an important source of information for parents:

> The meeting with the physician before the surgery, that was obviously key. That was a really big one for us. To actually talk to the guy who was going to do this, and to find out whatever we can about success, failure, problems, all that kind of stuff. [parent of 2-year-old]

In terms of the appropriate format for sharing information, all parents and clinicians agreed that counselling from a health professional, information pamphlets, books, videos, and the internet were useful ways of helping with their decision-making. All participants were uncertain of the value of support or discussion groups for families making the same decision. Most parents suggested that it might be useful for some families but that they would be unlikely to use a support group.

When asked about who should disseminate the information, there were some variations in the responses. There was general agreement that the government, health societies and non-profit groups had only a small role to play in the development of resources for families. All clinicians and parents agreed that information materials should be prepared by medical staff and researchers. One clinician summarized the current challenge in providing unbiased research information:

> Preferably, I'd like nice unbiased research. Although, in reality, I mean, that's kind of difficult to still find, and a lot of the information that's available… and a lot of the research has been sponsored by one of the companies or the other, and so giving decent advice information sometimes is a little difficult. [clinician]

Four of the clinicians felt that CI manufacturers should create the information pamphlets, but the other four clinicians noted that this could result in biased information for families. While some parents (N=3) felt that the information should not come from CI manufacturers because of potential bias, other families felt that the manufacturers had a role to play in providing information.

Half of the parents felt that parents of children with CIs should help prepare information materials. This echoes the request for additional resources regarding the experiences of other families who have chosen CIs for their children. Clinicians did not feel strongly about parents' participation in preparing information.

Bilateral Cochlear Implantation

There was uncertainty and variability when participants were asked about their

perceptions of the bilateral CI option for their child. The parents and some clinicians discussed (a) their perceptions of the bilateral decision, benefits, and risks, (b) their pre-disposition to the bilateral cochlear implantation decision, and (c) some of the barriers to decision-making.

Perceptions of the bilateral cochlear implant decision. There were differences in the parent and clinician perceptions of the benefits and risks associated with bilateral implantation. In contrast to the benefits from unilateral implantation that all parents were unanimous about, only one parent brought up the additional benefits of a bilateral CI, such as an improvement in sound localization and hearing in noise. The other parents brought up their concerns regarding the perceived risks of a second CI. One parent reported concern about a second surgery:

> They suggested that we should do two because her right ear is not good, but still, I don't want to do that now, because this is her first one… because it's her brain. It's head surgery. It scares me. [parent of a 4-year-old child]

Parents did not report that the increased risks of meningitis and mastoiditis associated with the second surgery influenced their decision. However, a clinician commented that she was uncertain that parents fully understood the risks associated with the second surgery:

> Then we start talking about binaural implantation. I have a feeling that somehow, people are hiding their heads, putting their head in the sand. They're not really paying attention to those potential risks. If I had a child who was deaf, I don't know

> if I'd go for a binaural implant. I'd go for the first one, and I'd accept the risks. And you can't judge what people decide to do, but there is this feeling that they want the success, and they don't necessarily grasp the risks. The parents who have been through the case of mastoiditis, and meningitis have actually been quite brave about it, and have freely accepted those events, but I think we've been lucky. [clinician]

Pre-disposition to the bilateral cochlear implantation decision. Half of the interviewed parents expressed a great interest in receiving a second implant as soon as possible for their child:

> Now that I see that [the first cochlear implant] does work and I've been talking to different people about getting a second one it is something that we definitely want for our children. I feel like they should have that opportunity to have the direction finding [sound localization] [parent of a 3-year-old].

The four parents who were still at the decision-making stage responded that they were uncertain about what they would choose for their child:

> Everybody has a different reaction to doing it. I'm kind of, I'm cautious by nature, so I'm kind of, "Well, let's see if this is a good thing to do," as opposed to, "Yeah, I want to have him have that bilateral." [parent of a 3-year old]

One clinician also expressed her perception of parental uncertainty about the bilateral CI decision:

> Not all parents will want two implants for their kids, and that's fine. We've got... I think the decision-making is, it's going to be more variable. But we'll have to respect that. I know some parents have told me, "Well, we're going to get one, and we'll wait until something better comes up for the second one," or, "Nope. We'll go for two because I've read that two is better than one, and we've got two hearing aids, we want two implants." It depends on the parent. [clinician]

Barriers to bilateral cochlear implantation decision making. Many of the parental comments revealed barriers to bilateral cochlear implantation decision-making. In particular, they focused on their perceived lack of knowledge about the bilateral procedure. One parent who had been actively seeking bilateral implants for her children expressed her interest in having more research available to support her family's decision:

> Even with the bilateral, we believe that it's best for them, I do wish that there was more research stating exactly, "These are the advantages," or, "Hearing will improve in these ways." The research aspect, I wish there was a lot more out there. [parent of a 2- year-old]

Another parent and a clinician expressed similar requests for additional information and support for the bilateral decision:

> I know we don't have enough experience with two implants now to have a lot of data on it, so I don't feel comfortable enough with the counselling and all because it's not

there. [clinician]

People are going ahead and doing this, they've got to line up [the bilateral information] just the same way they should line up the cochlear implant information. So that parents who are even thinking about it know that there's a resource they can go to and start looking at that. [parent of 3-year-old]

Another parent voiced concerns regarding the difficulty in making decisions for her young son without his involvement:

We now see that bilateral implantation is possibly another decision we have to make in his lifetime. We actually hope that it will be in his lifetime as opposed to ours, but I also feel that I would like him to be able to make the decision. He's had it done once where we made the decision for him as the parent, but I like would like him, with whatever life experience he's had at that point to be able to decide whether or not he wants to do it. I would feel better about it. [parent of 3-year-old]

In making the decision for unilateral implantation, parents consistently reiterated their comfort with the decision for the CI. However, there was more uncertainty among parents with regards to the bilateral CI.

Discussion

Identification of Hearing Loss and Early Decision-Making

The parents' description of the initial shock and grief about the diagnosis of hearing

loss was consistent with other literature on the topic (Anagnostou et al., 2007; Kurtzer-White & Luterman, 2003). The current study identified differences in the magnitude of the parental reaction to the diagnosis depending on the child's co-existing health issues. There is no specific literature on the parental reactions to a diagnosis of hearing in parents of children with complex co-morbidities. This may be an area for further inquiry.

The families did not describe struggling with the decision about the communication approach for their child. Li et al., (2003) reported similar results in their survey regarding the attitudes, beliefs, and values of 83 parents of children with various levels of hearing loss. They reported that the second most influential factor in deciding about the communication modality, after the degree of a child's hearing loss, was the parents' desire to use spoken communication with their child. As all the children in the current study were CI candidates or recipients with severe to profound losses, the degree of hearing loss did not differentiate between the parents in this study.

Perception of the Cochlear Implant Decision

All parents emphasized that the perceived risks associated with the CI implantation were acceptable in relation to the value that they attributed to oral communication with their child. These findings were consistent with a study that examined the influence of parental values on the CI decision-making (Li et al., 2004). That study examined families from a variety of deaf communication programs: oral, sign, and total communication programs. In the 50 families that participated, 33 children proceeded with the CI surgery while the other 17 children did not. Among the 17 families who decided against the CI, the authors found that their attitude toward communication could be used as a statistical predictor for their final decision. The authors emphasized that a CI is often emotionally loaded for some families.

Identifying the value that parents place on oral rather than manual communication may be important in identifying those parents who will have difficulty with the CI decision-making process.

Options Available

In this study, the parents of children who had begun AVT and were already progressing in their oral language development before the CI decision arose did not perceive sign language as a viable option. In contrast, the families that had to make the CI decision immediately after their child's diagnosis perceived the decision as between CIs and sign language. The different perception of treatment options suggests that the parents of children with an early diagnosis chose the communication approach together with the CI.

Many of the advantages and disadvantages of the options listed by participants were consistent with previous findings in the literature (Sach & Whynes, 2005; Incesulu et al., 2003; Kluwin & Stewart, 2000). The primary perceived advantage of the CI option was the increased opportunity for speech and language exposure. This was followed by hopes for improved communication skills and becoming a member of the larger hearing community. It is a specific oddity of the Canadian context that a CI is fully covered by the public health care system while hearing aids are only partly funded. This was noted by one of the clinicians as an apparent benefit to parents. The influence of cost on the CI decision was not explored in the present study but may be of interest for future research.

The parents' qualms about making the CI decision for a child who might later resent that decision had been noted as a stress factor by Sach and Whynes (2005). In the survey by Incesulu et al. (2003), 6 of the 25 participating parents reported concern about later blame from their children. Parents in the current study explained that this concern was alleviated by

the fact that their children could still have the CI removed if they so chose.

Manifestations of decisional conflict

Like other studies, results of this study indicated that the parents were confident about the CI choice. Nevertheless, preparing to undergo surgery and the entire CI process was a stressful event for the families (Incesulu et al., 2003; Most & Zaidman-Zait, 2003). Identifying ways to adequately address and reduce this stress should be both a research and clinical priority.

Perception of Others Opinions, Practices, Support, and Pressures

Information about other families' decisions was reported to be the single most helpful piece of support for the CI decision. This is in agreement with the results of previous studies (Incesulu et al., 2003, Most & Zaidman-Zait, 2003). The discrepancy between parents' and clinicians' perceptions of what constituted adequate information on the decisions of other families should be noted. Notwithstanding the small sample size in the present study, it might be worthwhile to explore additional ways to put parents in touch with other families to share information and emotional support (Most & Zaidman-Zait, 2003).

Apart from the contact to other families, the contact with audiologists, auditory-verbal therapists, and surgeons was perceived as important during the decision-making process. The multi-disciplinary team provides an important support mechanism for parents of children with hearing loss (Fitzpatrick, Angus, Durieux-Smith-Graham, & Coyle, 2008; Most & Zaidman-Zait, 2003).

Resources to Make Decisions

The need for additional information is often closely associated with the particular

clinical profile of a child. In this study, the parents of children with auditory neuropathy and children with progressive loss requested additional information and resources for decision-making. Kluwin and Stewart (2000) interviewed 35 families who had undergone cochlear implantation with their children. They identified that most families were satisfied with the information they received. However, eight families felt that they would have liked more information on the surgery and rehabilitation process. While the majority of parents appear content overall with the available resources, there may be a need for more information for some families. Based on a series of case studies, Neuss (2006) described families' search for information before deciding on the CI. The results were similar to the current study. Most parents stated that they supplemented the information from clinicians with additional research on the internet or with discussions with other parents.

The parents' preferences for information in brochure format as well as on the internet was consistent with a national survey on the decision-making needs of Canadians (O'Connor, Drake et al., 2003). In addition, like the respondents in the national survey, the parents and clinicians interviewed in this study preferred that the materials be created and disseminated by medical and health-care specialists. In an examination of the internet resources available to parents making decisions about unilateral cochlear implantation for their children, Zaidman-Zait and Jamieson (2004) found that the majority of articles available for parents were from medical departments, consumer organizations, CI manufacturers, and health care providers. The researchers qualitatively evaluated the information provided on these websites and concluded that the available evidence for parents was neither peer-reviewed nor evidence-based, and that the creators of the sampled websites rarely referred parents to research that is available in the public domain. A recent survey of parents by Sorkin and Zwolan (2008) found a perceived lack of bias-free information on CIs.

Bilateral Cochlear Implantation

The parental responses to the question of bilateral CIs indicated that the decision was more difficult than for the unilateral CI. The parents were uncertain about the potential benefits and about the value of these benefits to their child or their family. At the time of this study, bilateral CIs were relatively new to this clinical setting and did not constitute the standard of care. In contrast to the unilateral CI, the value that the parents placed on the second CI does not appear consistent. The bilateral procedure may have been perceived as elective because a second device provides improvements in sound localization and speech intelligibility, compared to the boost of speech and language development associated with a unilateral CI.

As the bilateral clinical treatment option was relatively new, parents and clinicians perceived a lack of information and resources. These findings indicate a need to develop more information in user-friendly formats to support families in their deliberations of the bilateral CI option.

Planning for Decision-Support

Including patients in decisions about their health by providing research information is an important component of knowledge translation (Holmes-Rover et al., 2001; Coulter, 1997). Based on the results of this study, a knowledge translation tool to help families increase their knowledge about cochlear implantation prior to and during their decision-making would appear beneficial. The need for such a tool was pronounced for the bilateral CI decision. One approach to translating knowledge for health-care consumers is through the use of patient decision aids (O'Connor & Edwards, 2001). Decision aids are "tools designed to help people participate in decision making about health care options. They provide

information on the options and help patients clarify and communicate the personal value they associate with different features of the options" (International Patient Decision Aid Standards, 2008).

Decision aids can be particularly helpful in situations where a choice between two or more treatments options is available and no clear standard of care is available based on evidence (O'Connor & Edwards, 2001). They have been shown to improve the decision-making quality and process, to decrease anxiety, and to create more realistic expectations of outcomes (O'Connor et al., 2002). Currently, no decision aid exists for the decision to undergo paediatric unilateral or bilateral cochlear implantation (Cochrane Inventory of Patient Decision Aids, 2008).

Limitations

By only interviewing parents after their CI decision there would have been potential for the parental perceptions to have been influenced by recall bias, decisional regret and parents' need to appear content with their decision. This was addressed by including interviews of parents involved in prospective decision-making. We attempted to purposefully sample families who had chosen not to undergo cochlear implantation but these families elected not to participate.

The sample size for this study was relatively small. As only 20-24 children are implanted each year in the study CI centre, only 30-36 children were eligible to participate based on the inclusion criteria. The participant pool was further reduced because approximately 20% of the population in this clinic was French speaking and the interviews were only conducted in English. During the interviews, similar themes emerged from the parents indicating that sufficient data saturation was achieved even with the small sample.

Some demographic information was not collected from the participants. This included socioeconomic status, family support, and immigration status, These factors may also have had an impact on the decision-making of families. The clinical characteristics that were included and explored in the study (i.e., child's age at identification, co-existing health issues, and aetiology of hearing loss) could not be generalized to the entire population of families due to the small sample size in this study. Future research should explore the influence of all of these factors in a larger sample of families.

Only a single CI site was included in this study. This centre has a strong emphasis on AVT as the dominant treatment option for children and families. Families and clinicians from other centres may have different perceptions of the CI decision-making process.

Conclusions

The interviewed parents reported that their decision to undergo cochlear implantation for their children with severe to profound hearing loss was related to the value that their family placed on oral communication. Comprehensive information on the risks and benefits associated with cochlear implantation should be offered to all families. Parents also benefit from their interactions with, and the support offered by, families who have already made their choice. While the choice for a single CI appeared to be a value-based and presented little decisional conflict, the situation was more complex for parents contemplating a bilateral CI. Bilateral CI decision-making should be addressed systematically in further research to further understand and support parents of children with bilateral severe to profound hearing losses.

References

Anagnostou, F., Graham, J., & Crocker, S. (2007). A preliminary study looking at parental emotions following cochlear implantation. *Cochlear Implants International, 8*, 68-86.

Berg, A. L., Ip, S. C., Hurst, M., & Herb, A. (2007). Cochlear implants in young children: Informed consent as a process and current practices. *American Journal of Audiology, 16,* 13-28.

Bergeron, F. (2000). Residual hearing following cochlear implantation: Effect of time and device. *Advances in Otorhinolaryngology, 57*, 389-92.

Biernath, K. A., Reefhuis, J., Whitney, C. G., Mann, E. A., Costa, P., Eichwald, J., et al. (2006). Bacterial meningitis among children with cochlear implants beyond 24 months after implantation. *Pediatrics, 117*, 284-289.

Boggess, W. J., Baker, J. E., & Balkany, T. J. (1989). Loss of residual hearing after cochlear implantation. *Laryngoscope, 99,* 1002-1005.

Brown, K.D., & Balkany, T.J. (2007). Benefits of bilateral cochlear implantation: a review, *Current Opinions in Otolaryngology & Head and Neck Surgery, 15*, 315-8.

Candidacy Criteria. (2004). Retrieved February 9, 2008 from http://www.cochlear.com/Support/631.asp

Ching, T.Y., van Wanrooy, E. & Dillon, H. (2007). Binaural-bimodal fitting or bilateral implantation for managing severe to profound deafness: a review, *Trends in Amplification*, *11*, 161-92.

Cochrane Inventory of Patient Decision Aids. (2008). Retrieved June 10, 2008 from http://decisionaid.ohri.ca/cochinvent.php

Cohen, N. L., Hoffman, R. A., & Stroschein, M. (1988). Medical or surgical complications

related to the Nucleus multichannel cochlear implant. *Annals of Otology, Rhinology, and Laryngology Supplemental, 135*, 8-13.

Coulter, A. (1997). Partnerships with patients: The pros and cons of shared decision making. *Journal of Health Services Research Policy, 2,* 112-121.

Fayad, J. N., Wanna, G. B., Micheletto, J. N., & Parisier, S. C. (2003). Facial nerve paralysis following cochlear implant surgery. *Laryngoscope, 113*, 1344-1346.

Fina, M., Skinner, M., Goebel, J. A., Piccirillo, J. F., Neely, J. G., & Black, O. (2003). Vestibular dysfunction after cochlear implantation. *Otology and Neurotology, 24,* 234-242.

Fitzpatrick, E., Angus, D., Durieux-Smith, A., Graham, I. D., & Coyle, D. (2008). Parents' needs following identification of childhood hearing loss. *American Journal of Audiology, 17*, 38-49.

Gregoret, L. (2003). Bilateral cochlear implants: are they in your child's future? *Volta Voices, 10*, 10-15, 34.

Gysin, C., Papsin, B. C., Daya, H., & Nedzelski, J. (2000). Surgical outcomes after paediatric cochlear implantation: diminution of complications with the evolution of new surgical techniques. *Journal of Otolaryngology, 29*, 285-289.

Holmes-Rovner, M., Llewellyn-Thomas, H., Entwistle, V., Coulter, A., O'Connor, A., & Rovner, D.R. (2001). Patient choice modules for summaries of clinical effectiveness: A proposal. *British Medical Journal, 322,* 664-667.

Incesulu, A., Vural, M., & Erkam, U. (2003). Children with cochlear implants: Parental perspective. *Otology & Neurotology, 24,* 605-611.

International Patient Decision Aid Standards. (2008). What are decision aids? Retrieved June 10, 2008 from http://ipdas.ohri.ca/what.html

Jacobson, M. J., & O'Connor, A. M. (2006). Population needs assessment: A workbook for assessing patients' and practioners' decision making needs. Retrieved July 18, 2008 from http://decisionaid.ohri.ca/docs/implement/Population_Needs.pdf

Kluwin, T. N., & Stewart, D. A. (2000). Cochlear implants for younger children: A preliminary description of the parental decision process and outcomes. *American Annals of the Deaf, 145,* 26-32.

Kurtzer-White, E., & Lutterman, D. (2003). Families and children with hearing loss: Grief and coping. *Mental Retardation and Developmental Disabilities Research Reviews, 9,* 232-235.

Li, Y., Bain, L., & Steinberg, A. G. (2003). Parental decision-making and the choice of communication modality for the child who is deaf. *Archives of Pediatric Adolescent Medicine, 157,* 162-168.

Li, Y., Bain, L., & Steinberg, A. G. (2004). Parental decision-making in considering cochlear implant technology for a deaf child. *International Journal of Pediatric Otolaryngology, 68,* 1027-1038.

Moog, J. S., & Geers, A. E. (2003). Epilogue: Major findings, conclusions and implications for deaf education. *Ear and Hearing, 24,* 121S-125S.

Most, T., & Zaidman-Zait, A. (2003). The needs of parents of children with cochlear implants. *The Volta Review, 103,* 99-113.

Murphy, J., & O'Donoghue, G. (2007). Bilateral cochlear implantation: an evidence-based medicine evaluation, *Laryngoscope, 117,* 1412-18.

Neuss, D. (2006). The ecological transition to auditory-verbal therapy: Experiences of parents whose children use cochlear implants. *Volta Review, 106,* 195-222.

Northern, J., & Downs, M. P. (1991). *Hearing in children* (4th ed.). Baltimore, MD: Williams & Wilkings.

O'Connor, A. M. (1995). Validation of a decisional conflict scale. *Medical Decision Making, 15,* 25-30.

O'Connor, A. M., Drake, E., Wells, G. A., Tugwell, P., Laupacis, A., & Elmslie, T. (2003). A survey of the decision-making needs of Canadians faced with complex health decisions. *Health Expectations, 6,* 97-109.

O'Connor, A., & Edwards A. (2001). The role of decision aids in promoting evidence-based patient choice. In A. Edwards, G. Elwyn (Eds.), *Evidence-based patient choice* (pp. 220-42). Oxford, UK: Oxford University Press.

O'Connor, A. M., Legare, F., & Stacey, D. (2003). Risk communication in practice: the contribution of decision aids. *British Medical Journal, 327*, 736-740.

O'Connor, A.M., Stacey, D., Entwistle, V., Llewellyn-Thomas, H., Rovner, D., Holmes-Rovner, M., et al. (2002). Decision aids for people facing health treatment or screening decisions (Cochrane Review). Retrieved on July 18, 2008 from http://www.cochrane.org/reviews/en/ab001431.html

O'Connor, A. M., Tugwell, P., Wells, G. A., Elmslie, T., Jolly, E., Hollingworth, G., et al. (1998). A decision aid for women considering hormone therapy after menopause: Decision support framework and evaluation. *Patient Education and Counseling, 33*, 267-279.

Perreau, A.E., Tyler, R. S., Witt, S., & Dunn, C. (2007). Selection strategies for binaural and monaural cochlear implantation. *American Journal of Audiology*, *16*, 85-93.

Peters, E. (2000). Our decision on a cochlear implant. *American Annals of the Deaf, 145*,

263-267.

Sach, T. H., & Whynes, D. K. (2005). Paediatric cochlear implantation: The views of parents. *International Journal of Audiology, 44,* 400-407.

Samson-Fang, L., Simons-McCandless, M., & Shelton, C. (2000). Controversies in the field of hearing impairment: early intervention, educational methods, and cochlear implants. *Infants and Young Children, 12*, 77-88.

Schafer, E.C., & Thibodeau, L.M. (2006). Speech recognition in noise in children with cochlear implants while listening in bilateral, bimodal, and FM-system arrangements, *American Journal of Audiology*, *15*, 114-26.

Sorkin, D. L., & Zwolan, T. A. (2008). Parental perspectives regarding early intervention and its role in cochlear implantation in children. *Otology and Neurootology, 29*, 137-141.

Steinberg, A., Brainsky, A., Bain, L., Montoya, L., Indenbaum, M., & Potsic, W. (2000). Parental values in the decision about cochlear implantation. *International Journal of Pediatric Otorhinolaryngology, 55,* 99-107.

Tambyraja, R. R., Gutman, M. A., & Megerian, C. A. (2005). Cochlear implant complications; Utility of federal database in systematic analysis. *Archives of Otolaryngology and Head and Neck Surgery, 131,* 245-250.

Tashakkori, A., & Teddlie, C. (Eds.). (2003). *Handbook of mixed methods in social and behavioral research.* Thousand Oaks, CA: Sage.

Thoutenhoofd, E., Archbold, A., Gregory, S., Lutman, M., Nikolopoulos, T., & Sach, T.H. (2005). *Paediatric cochlear implantation: Evaluating outcomes*. London: Whurr.

United States Food and Drug Administration. (2007). Advice for patients with cochlear implants: New information on meningitis risk. Retrieved May 18, 2008 from http://69.20.19.211/cdrh/medicaldevicesafety/atp/101007-cochlear.html

Wennberg, J. E. (2002). Unwarranted variations in healthcare delivery: Implications for academic medical centres. *British Medical Journal, 325*, 961-964.

William House Cochlear Implant Study Group. (2008). Position statement on bilateral cochlear implantation. *Otology and Neurootology*, *29*, 107-8.

Zaidman-Zait, A., & Jamison, J. (2004). Searching for cochlear implant information on the internet maze: Implications for parents and professionals. *Journal of Deaf Studies and Deaf Education, 9,* 413-426.

Table 1

Perceived Advantages and Disadvantages of Available Options as Described by Parents and Clinicians

Cochlear Implant Option		Hearing Aid Option		Sign Language Option	
Advantages	Disadvantages	Advantages	Disadvantages	Advantages	Disadvantages
- Greater exposure to speech and language at early age - Consistent with hearing family's culture - Potential for the child to use spoken communication - Potential for the child to communicate with larger community - Possible removal later if child chooses sign language - Cost of device is covered compared to HA	- Surgical risk - Increased risk of Meningitis - Making a decision for a child that might have made a different decision - Reduced possibility of using newer technology - Challenges of repairs, device failure, programming - Cosmetic issues of external and internal device - Travel and time for fitting and programming	- No surgical risk - Consistent with hearing family's culture - Ability to hear the child speak - Possible removal later if child chooses sign language - No loss of residual hearing	- Less speech and sound exposure from greater distances - Slower speech and language development than with CI - Challenges of feedback, HA repairs	- No surgical risk - Consistent with signing family's culture - A small, warm cultural community available to child	- Child enters a culture unfamiliar to hearing family and friends - Entire hearing family needs to acquire a new language - Living in a minority culture - Fewer employment /educational options available - Limited opportunity to chose oral communicatio n after childhood

Figure Captions

Figure 1. Ottawa Decision Support Framework

Figure 2. Parent responses to structured questions regarding reported behavioral manifestations of decisional conflict about the cochlear implant decision-making process.

Figure 3. Clinicians' and Parents' responses regarding what makes the cochlear implant decision difficult.

Figure 1

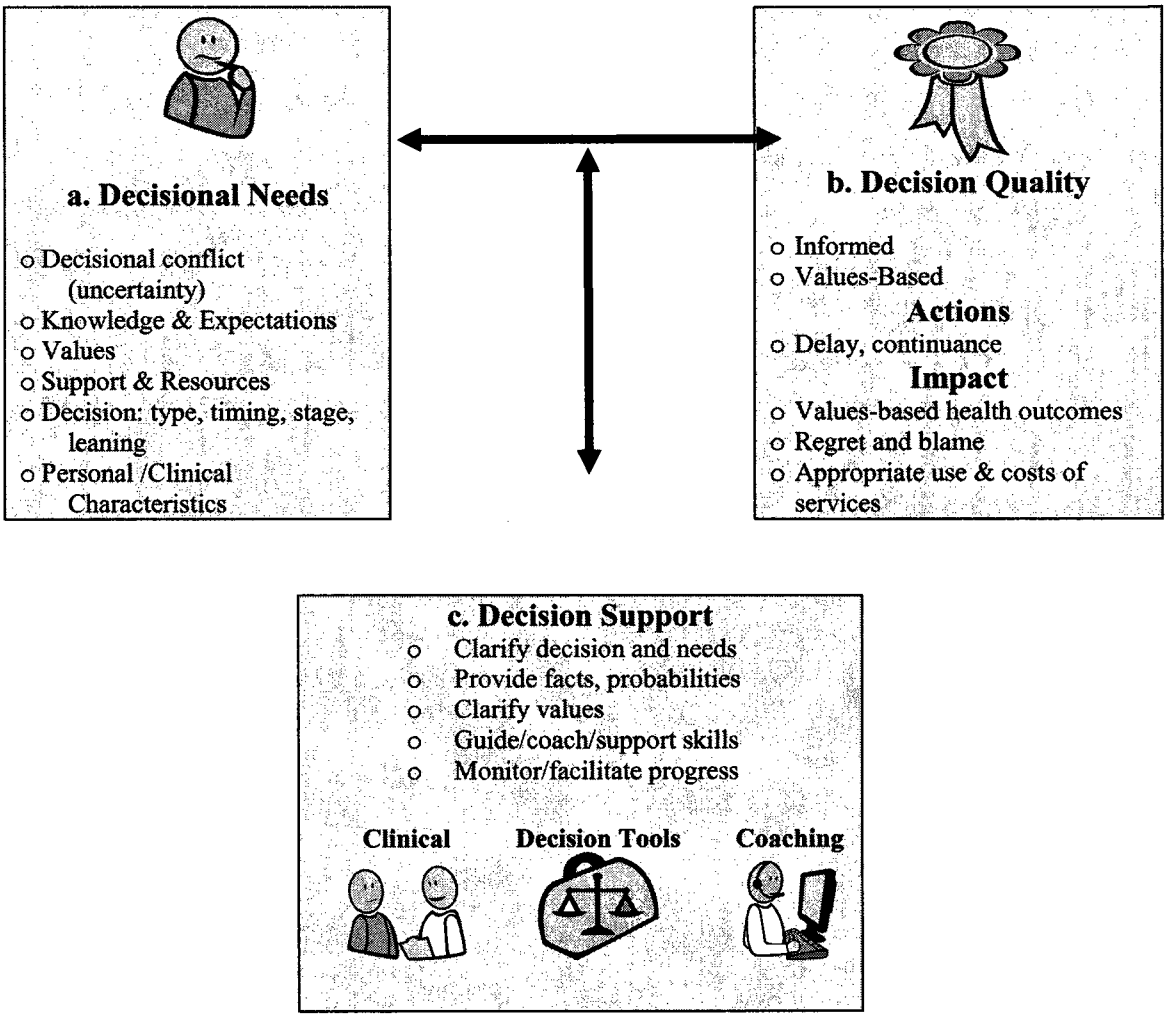


Figure 2

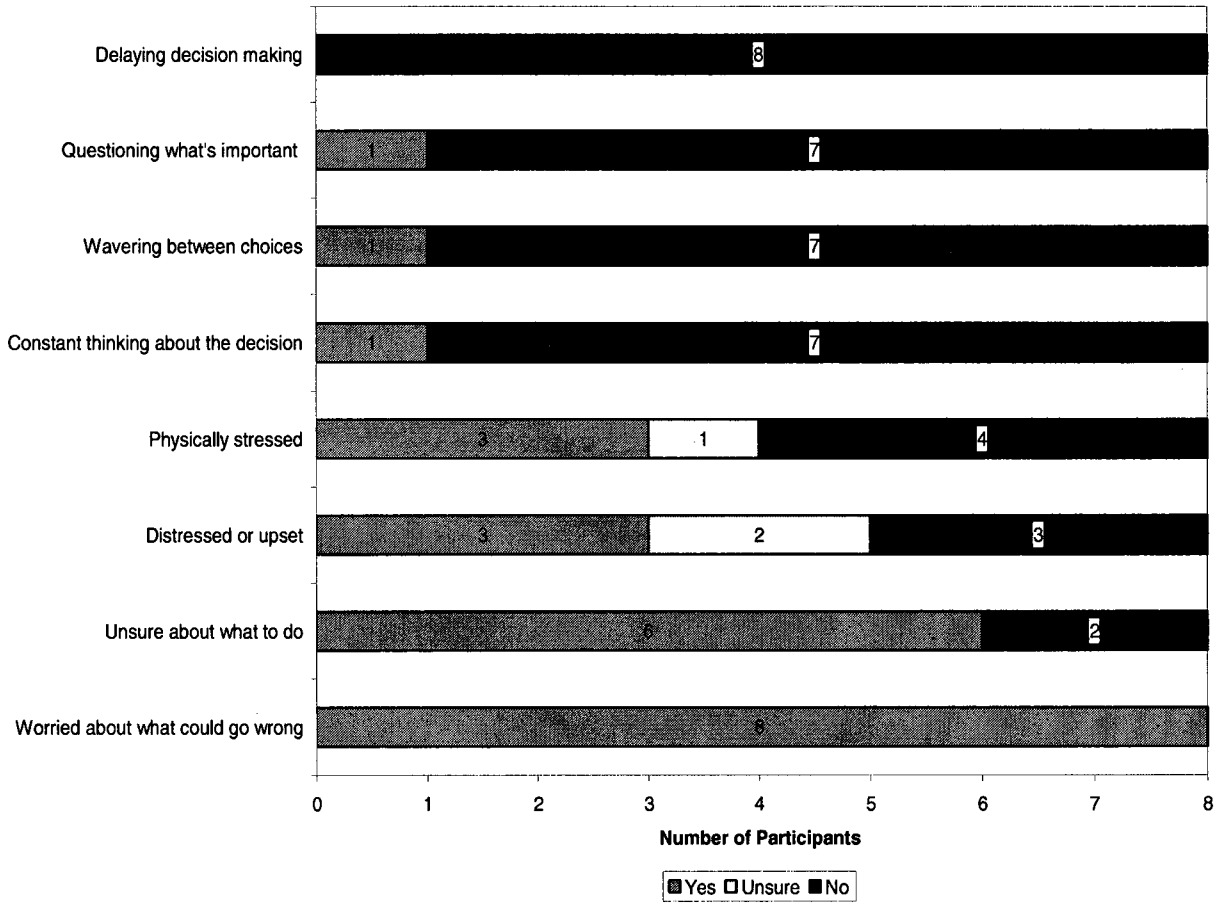
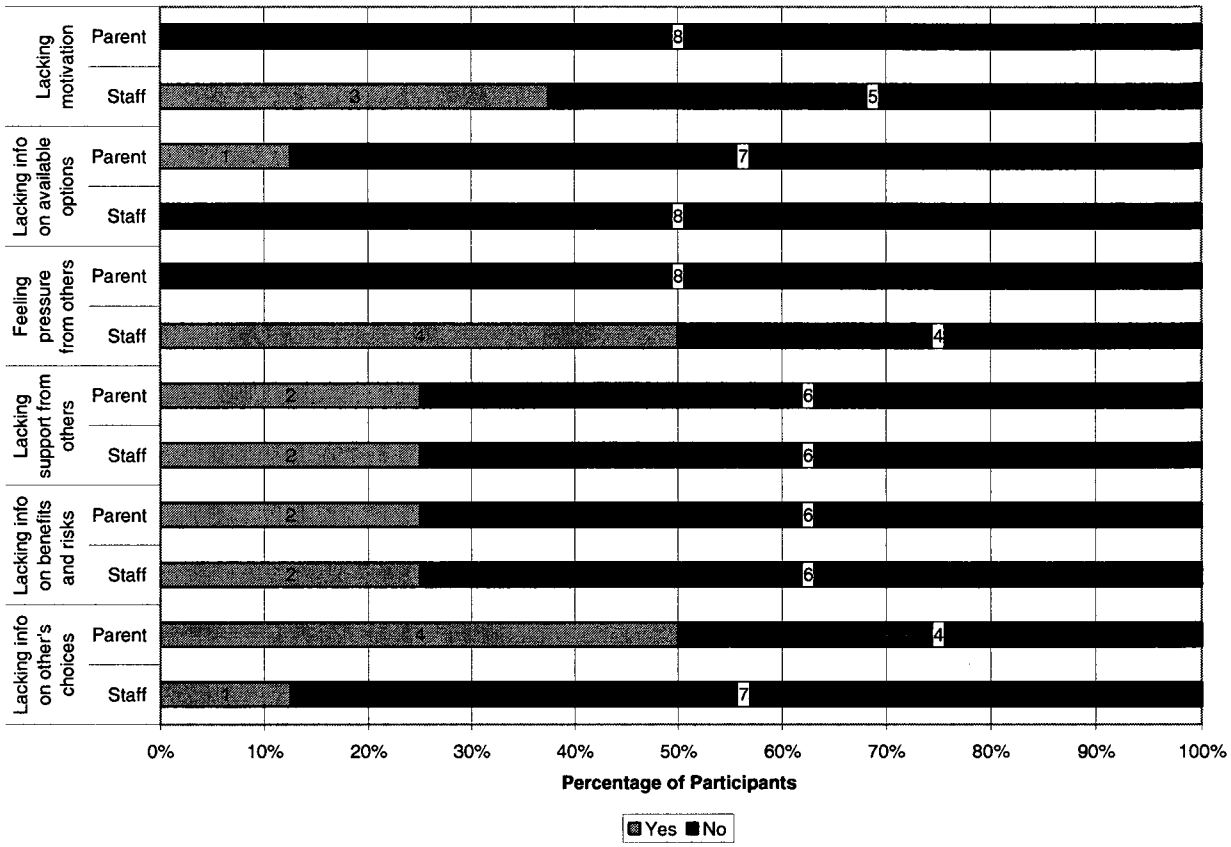

Delaying decision making
Questioning what's important
Wavering between choices
Constant thinking about the decision
Physically stressed
Distressed or upset
Unsure about what to do
Worried about what could go wrong
8
1
7
1
7
1
7
3
1
4
3
2
3
6
2
8
0
1
2
3
4
5
6
7
8
Number of Participants
Yes
Unsure
No

Figure 3


Lacking motivation
Parent
Staff
Lacking info on available options
Parent
Staff
Feeling pressure from others
Parent
Staff
Lacking support from others
Parent
Staff
Lacking info on benefits and risks
Parent
Staff
Lacking info on other's choices
Parent
Staff
0%
10%
20%
30%
40%
50%
60%
70%
80%
90%
100%
Percentage of Participants
Yes
No

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CHAPTER 3

Bilateral paediatric cochlear implants:

A critical review

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Bilateral paediatric cochlear implants:

A critical review

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Keywords: Bilateral, Binaural, Review, Cochlear implant, Children, Hearing

Abbreviations: ABR: auditory brainstem response, AdSpon: Adaptive Spondee Test, BICI: bilateral cochlear implant, CAEP: cortical auditory evoked potentials, CI: cochlear implant, CIHA: cochlear implant and hearing aid, CRISP: Children's Realistic Index of Speech Perception, ESPT: Early Speech Perception Test, FM: frequency modulation, HA: Hearing Aid, HINT: Hearing in Noise Test, LNT: Lexical Neighbourhood Test, MAA: Minimum Audible Angle, MLNT: Multisyllabic Lexical Neighbourhood Test, N: number of participants, NH: normal hearing, PedsQL: Pediatric Quality of Life Measure, R/L: right/left, SRM: Spatial Release from Masking, SSQ: Speech, Spatial, and Qualities of Hearing Scale, WNL: within normal levels.

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Abstract

A recent trend has been the implantation of bilateral cochlear implants (CIs) for children with severe to profound hearing loss. A review of available research on bilateral CIs was conducted to determine the support for this trend. A replicable review was undertaken to evaluate published research studies that examined the effectiveness of bilateral paediatric cochlear implantation. Databases, reference lists, and journals were searched for relevant documents using a pre-determined search protocol. Twenty-nine articles met the review's inclusion criteria and were retrieved and reviewed. This review adds to the previously published reviews on the topic by identifying additional paediatric studies. Sound localization and speech recognition in noise appear to be improved with bilateral compared to unilateral cochlear implants. Similarly, evoked potential measures suggest improved morphology when the second CI is implanted early. Well designed and controlled studies that explore a variety of outcomes including cost-effectiveness, quality of life, speech, language and psycho-educational measures should be further explored in order to provide additional support for parents and clinicians confronted with the bilateral cochlear implant decision.

Since the acceptance of cochlear implants (CIs) by the United States' Food and Drug Administration in 1990 (FDA, 2008), CIs have become the standard of care for children with severe to profound hearing loss. Over the years, the criteria for CI use in children have been expanded significantly. Originally used only in older children with profound hearing loss, now children with lesser degrees of hearing loss are eligible for CIs (Nucleus ® 24 Candidacy Criteria, 2008). In addition, children may be implanted with CIs earlier than one year of age (Thoutenhoofd et al., 2005).

A systematic review of the effectiveness of unilateral paediatric cochlear implantation published in 2005 highlighted that strong evidence exists for unilateral CIs improving hearing sensitivity levels and speech perception (Thoutenhoofd et al., 2005). The report identified that there was less definitive evidence regarding improved expressive language, communication, and quality of life outcomes. However, a multi-site study, that was not included in the review, reported language and literacy improvements in children using CIs compared to previously published reports of children using hearing aids (Moog & Geers, 2003). In fact, half of the 181 participating children had language scores within the normal range (Geers et al., 2003).

The improved speech, language, and communication outcomes for children with unilateral CIs have made CIs the standard of care for children with bilateral severe to profound hearing losses (Berg et al., 2007; Moog & Geers, 2003). A recent trend is that more centres are performing bilateral cochlear implantation in children (Berg et al., 2007). Bilateral implantation involves the simultaneous or sequential surgical implantation of the device in both ears. At the moment, as most children have already been provided with a single CI, the

majority of bilateral surgeries have been sequential (Shafir, 2007). Bilateral implantation provides children with severe to profound hearing losses with bilateral auditory stimulation. As with any new medical procedure, questions arise from clinicians, parents, and cochlear implant users regarding the effectiveness, the benefits and risks, of paediatric bilateral cochlear implantation (Brown & Balkany, 2007; Berg, et al., 2007).

The psychoacoustic literature has demonstrated the benefits of binaural hearing for those with normal hearing and for many with bilateral amplification using measures of speech recognition and sound localization (Colburn et al., 2006; Mencher & Davis, 2006). Speech recognition can be measured in noise or in quiet. Speech recognition in noise is typically improved in binaural compared to monaural hearing conditions (Zurek, 1993). Sound localization refers to the ability to identify and locate the source of a sound in space (Brown & Balkany, 2006). Those with available binaural signals take advantage of the head shadow, binaural summation, and binaural squelch effects to improve their speech recognition in noise and sound localization performance (Brown & Balkany, 2007). The benefits of binaural hearing have been observed in normally hearing and in individuals with bilateral hearing aids (Colburn et al., 2006; Mencher & Davis, 2006). As such, it is a logical area for bilateral cochlear implantation research to determine if the same benefits are accrued through the auditory stimulation provided by two cochlear implants.

Recent narrative reviews examining the literature on bilateral cochlear implantation have explored sound localization and speech recognition in noise measures (Brown & Balkany, 2007; Ching et al., 2007; Murphy & O'Donoghue, 2007; Schafer & Thibodeau, 2006). These reviews included studies examining the effectiveness of bilateral cochlear implant use

(BICIs) compared to unilateral CI use in adults and children. All four reviews (Brown & Balkany, 2007; Ching et al., 2007; Murphy & O'Donoghue, 2007; Schafer & Thibodeau, 2006) concluded that there are benefits for patients receiving bilateral stimulation compared to use of a single CI on measures of speech recognition in noise and sound localization. Children with lesser degrees of hearing loss may receive benefit with a hearing aid in their second ear (Ching et al., 2007; Schafer & Thibodeau, 2006). For children who can not use hearing aids in their second ear due to severity of hearing loss, this bilateral stimulation would only occur through the use of a second CI.

As BICIs become increasingly available in paediatric CI centres (Berg et al., 2007), it is essential that up-to-date research information is available for parents and clinicians on a variety of outcomes of interest. Sound localization and speech recognition in noise measures are important and have been explored in the literature (Brown & Balkany, 2007; Ching et al., 2007; Murphy & O'Donoghue, 2007). Speech, language, education, and quality of life outcomes are also important to those making decisions about bilateral implantation and have not been included in published reviews. This review was conducted as a means of providing readers with a replicable, systematic, catalogue, and synthesis of the paediatric BICI literature. Recent audiology and otolaryngology literature has included papers on the need for evidence for parents, researchers, and clinicians about bilateral implantation (Gregoret, 2003; Berg et al., 2007). The objective of this review is to provide clinicians, families, and researchers with updated, comprehensive information on the available paediatric BICI literature to aid them in their decision-making.

The primary research questions addressed by this review are: 1) What is the available

research on bilateral paediatric cochlear implantation?, 2) Does the use of bilateral CIs lead to improvements in communication and/or quality of life compared to use of a unilateral CI?, 3) Are there additional documented or perceived benefits associated with paediatric BICIs?

Methods

Selection criteria for studies

All research articles investigating paediatric bilateral cochlear implantation were included. Studies were included if they examined the outcomes of children with significant congenital or pre-lingual hearing loss who have undergone bilateral CI surgery and received multi-channel devices prior to age 18. Commentaries, position, and opinion papers were not included. All types of research evidence may be relevant to this relatively new clinical topic. Randomized controlled trials, prospective cohort studies, retrospective cohort studies, case studies and series, reviews, and qualitative studies were eligible for inclusion. No exclusion criteria were set based on the number of participants. Publication in a peer-reviewed journal was required.

Time frame

The review included studies published between 2000 and January 2008. Due to the lack of bilateral paediatric CI surgeries occurring before 2000, it was reasonable to commence the search from this time forward.

Outcome measures

Outcome measures of interest included auditory processing, speech recognition in quiet and in noise, sound localization, electrophysiological measures, oral receptive and expressive language, vocabulary, and communicative competence. Literature exploring other outcome measures including social and psychological functioning, educational placement, family satisfaction, and quality-of-life were sought to add additional information regarding the potential benefits of bilateral implantation. Surgical complications related to the CI surgery itself- such as facial palsy, vestibular problems, device failure, and negative reactions to anaesthetic- were not included in this review.

Search strategy

A pre-determined search strategy was used to search electronic databases Medline, PsycINFO, CINAHL, Web of Science, and ERIC. The primary search terms were "cochlear implant$" and "bilateral." These terms were combined in the database searches and included other additional secondary search terms. The identified articles were downloaded into reference management software for the application of the inclusion/exclusion criteria based on title and abstract. Articles were downloaded (and ordered where necessary) so that inclusion criteria could be applied based on the complete article. Those articles that met the inclusion criteria had their reference lists examined for additional relevant publications. Hand-searching of the tables of contents of journals *Ear and Hearing*, *Otology and Neurotology*, *American Journal of Audiology*, and *International Journal of Audiology* were also undertaken.

Inclusion/Exclusion criteria

Inclusion/exclusion criteria for this critical review are presented in Table 1. The inclusion criteria were applied to the results of the database search by two researchers as a means of assessing reliability. The two had 95.7% agreement and a Kappa of 0.862 (*Confidence Interval =0.77-0.95*) indicating very strong agreement.

Data abstraction

Articles that met inclusion criteria were read and examined for relevant information. A spreadsheet was used to organize information on each of the included articles. All outcome and secondary variables were entered into the spreadsheet. Table 2 provides a summary of the included articles, the characteristics of each study, their results, and conclusions based on the outcomes of interest in the original article.

Assessment of study quality

No assessment of study quality was undertaken. The Jadad Scale was deemed inappropriate due to the lack of randomized trials (Jadad & Moore, 1996). The Newcastle-Ottawa Scale (Wells et al., 2005) for grading the internal validity of studies was considered as this scale was developed as a means of assessing the quality of non-randomized studies during systematic reviews. Studies are evaluated on three criteria: "the selection of the study groups, the comparability of the groups, and the ascertainment of either the exposure or outcome of interest for case-control or cohort studies respectively." Due to the nature of current CI literature, the lack of blinding of study staff and subjects, the selection of convenience

samples, underpowered studies, and often lack of control groups, an assessment of study quality was not done.

Analysis

The heterogeneity of research designs and variability in methods and reporting prevented an overall meta-analysis from being undertaken. A descriptive analysis of the available studies was completed. Results from studies that used similar testing methods, outcome measures, and result reporting were compared for sound localization and speech recognition in noise measures. Two-tailed Z-tests of two proportions were used to examine if there was a difference in the proportion of children, using either a CI or BICIs, who were able to perform the tasks at specific levels.

Results

Figure 1 documents the progress through the review and the final number of included studies. Fifteen of the 29 identified papers (52%) were completed by researchers located in the United States. Seven (24%) were completed in Europe. Three of the papers were completed in Canada and the remaining four in Australia.

The trend for publishing on the topic of bilateral paediatric cochlear implantation is stronger in more recent years. Sixteen of the 29 identified studies (55%) were published in 2007 and the first month of 2008. Only one study was published before 2003. This trend reflects the relative newness of this research topic.

The average sample size of included studies was 12 and the range was from 1 subject to 46. Thirteen of the 25 primary research studies (52%) had ten or fewer subjects in their sample. Only six studies had 20 or more subjects.

Research Methods

A variety of research methodologies were identified. Four reviews that included the topic of bilateral paediatric CIs in their scope met the inclusion criteria. No randomized control trials were identified. A single national survey of CI centres in the United States was included. Fifteen cohort studies and two case-control studies were identified. Seven case series or case studies were identified. Nine studies had comparison groups. Four studies used a matched or independent unilateral CI comparison group. Two studies compared the results of the bilateral CI group with normally hearing children. Four of the 25 included primary research studies and two of the reviews included children with CIHA as the primary comparison group. Other studies compared the same children in two different testing conditions, once with their CI and secondly in a BICI condition. Table 2 provides a listing of the specific studies, their study design and composition of comparison groups.

The studies had high variability in methods, not only in ages of participants or weeks of device implementation, but in the outcomes measured, testing procedures, and selection criteria of control and experimental groups. As a result, a descriptive analysis of the available studies was completed and follows. Results from studies that used similar testing methods, outcome measures, and result reporting were included in tables in order to compare the performance of children with BICIs to children using CIs. This was possible for one sound

localization measure, the Minimum Audible Angle (MAA) and one speech recognition in noise measure, the Adaptive Spondees Test (AdSpon).

Outcomes of Interest and Reported Results

A variety of outcomes were reported in the identified literature. Sound localization and speech recognition in noise were overwhelmingly the primary outcomes of interest and at least one of these measures was included in 22 of the 29 articles. Five studies included electrophysiological measures making them the next most common outcome of interest. One of the objectives of this review was to identify articles examining outcomes that might describe some of the additional benefits associated with bilateral implantation- beyond some of the psychoacoustic measures that have been reported on previously (Brown & Balkany, 2007; Ching et al., 2007; Murphy & O'Donoghue, 2007; Schafer et al., 2007). Of particular interest were language, communication, and quality of life measures. Seven studies explored at least one of these outcomes: 2 using standardized measures and 5 using qualitative approaches or study designed measures. All of the outcomes and results are described in the following sections by outcome of interest.

Sound Localization and Speech Recognition in Noise. The four identified review and meta-analysis articles focused entirely on sound localization and speech recognition in noise in both children and adults (Brown & Balkany, 2007; Ching et al., 2007; Murphy & O'Donoghue, 2007; Schafer et al., 2007). These reviews and 17 original research articles that studied at least one of these outcomes are listed in Table 2. When individual research studies were examined to explore the results of sound localization in children, six of the twelve

research studies reported improvements in the BICI condition compared to the unilateral condition (Beijen et al., 2007; Litovsky et al., 2006a; Litovsky et al., 2006b; Mok et al., 2007; Senn et al., 2005; Steffens et al. 2007). The studies that did not report improvements in sound localization in the BICI condition had participants who had only recently received the BICI (average 8 months post-activation of second implant) (Grieco-Calub et al., 2008; Litovsky et al., 2004b) or had an extended period of auditory deprivation in the second ear (Galvin et al., 2007a; Galvin et al., 2007b; Litovsky et al., 2006b; Peters et al., 2004). To summarize, there was a longer period of time between the primary CI surgery and the second CI (between 3 and 10 years). One of the studies that reported significant overall improvements in sound localization for the BICI group found that the improvements were not universal across subjects and that improved performance was associated with reduced periods of deafness in the second ear (Litovsky et al., 2006a).

The studies examining sound localization in paediatric users of BICIs used a variety of testing approaches. The most commonly used measures were the minimum audible angle (MAA) and the localization of pink noise as measured in a 2-alternative-forced choice task. Some variations of these measures were used in the twelve identified studies. Differences occurred between studies in the stimulus presentation level (range from 60 to 70dB SPL) and the angle of the stimulus presentation (range from -90° to +90°, to -70° to +70°) (Beijen et al., 2007; Galvin et al., 2007a; Galvin et al., 2007b; Grieco-Calub et al., 2008; Litovsky et al., 2006a; Litovsky et al., 2006b; Litovsky et al., 2004a; Litovsky et al., 2004b; Mok et al., 2007; Peters et al., 2004; Senn et al., 2005; Steffens et al. 2007).

Table 3 provides a comparison of the results of the studies that examined the sound

localization performances of children using a BICI and a CI at various angles of stimulus presentation on the standardized MAA task (Grieco-Calub et al., 2008; Litovsky et al., 2006a; Litovsky et al., 2006b; Peters et al., 2004; Senn et al., 2005). Normally hearing children typically perform this task at thresholds below 20° (Litovsky, 1997). Twenty degrees was chosen as the target performance level for children using cochlear implants. In total, 31 children participated in these five studies that used the MAA testing paradigm. Results indicated that 46% (17/37) of the children using BICIs performed the task at the 20° threshold compared to 14% (5/35) of children using one CI. The proportion of children with BICIs who performed this task at levels comparable with children with normal hearing was significantly higher than the proportion of children with one CI who could perform the task at this level according to a 2-tailed, Z-test of two proportions ($Z=3.22$, $p=0.001$). The Peters et al. (2004) study did not demonstrate any improvement in the BICI condition. These children were all 8 - 13 years of age at the time of testing and had experienced extended periods of deafness in their second ear prior to receiving their second implant.

Thirteen primary research studies explored speech recognition in noise performance in children using BICIs (Galvin et al., 2007a; Galvin et al., 2007b; Kuhn-Inacker et al., 2004; Litovsky et al., 2006a; Litovsky et al., 2004a; Litovsky et al., 2004b; Mueller et al., 2000; Peters et al., 2004; Peters et al., 2007; Schafer & Thibodeau, 2006; Senn et al., 2005; Steffens et al. 2007; Wolfe et al., 2007). Some of the identified studies used standardized measures such as the Children's Realistic Index of Speech Perception (CRISP) (Litovsky et al., 2006a; Litovsky et al., 2004a; Litovsky et al. 2004b; Peters et al., 2004; Peters et al., 2007) or the AdSpon (Galvin et al., 2007a; Galvin et al., 2007b). The other studies used a variety of study-designed measures that included word and/or sentence recognition in various

noise types (Kuhn-Inacker et al., 2004; Mueller et al. 2000; Schafer & Thibodeau, 2006; Senn et al., 2005; Steffens et al. 2007; Wolfe et al., 2007). Improvements in speech recognition in noise in the BICI condition were reported in eleven of the thirteen studies (Galvin et al., 2007a; Kuhn-Inacker et al., 2004; Litovsky et al., 2006a; Litovsky et al., 2004a; Litovsky et al., 2004b; Mueller et al., 2000; Peters et al., 2004; Peters et al., 2007; Senn et al., 2005; Steffens et al. 2007; Wolfe et al., 2007). Similar to the sound localization results, the two studies that did not report improvements in speech recognition in noise in the BICI condition had participants that had experienced an extended period of time between the initial and the second CI surgery (Schafer & Thibodeau, 2006; Galvin et al., 2007b). As with the sound localization studies, the improvements were not universal across subjects. Improved speech recognition in noise performance was associated with reduced periods of deafness or auditory deprivation in the second ear in some studies (Peters et al., 2007; Litovsky et al., 2004a; Steffens et al. 2007) but not all (Kuhn-Inacker et al., 2004; Litovsky et al., 2006a; Wolfe et al., 2007).

Table 4 summarizes the results from the two identified studies that examined the speech recognition in noise performances of children using a BICI and a unilateral CI measured by the AdSpon in the ipsilateral noise presentation condition (Galvin et al., 2007a; Galvin et al., 2007b). Normally hearing children typically perform this task at signal-to-noise ratios of -11.5 dB (Rance et al., 2007). A total of 16 children participated in the two studies that used the AdSpon. None of the children using one CI could perform the task at levels typical of normal hearing children; whereas, 13% (2/16) of children with BICIs could perform the task at this level. The proportion of children with BICIs who performed this task at levels comparable with normally hearing children was not statistically different from the proportion

of children with one CI performing the task according to a 2-tailed, Z-test of two proportions ($Z=0.73$, $p=0.465$).

Electrophysiological Measures. Five of 29 studies reported on electro-physiological measures, and they are summarized in Table 2. Evoked potential measures can also be used to assess the binaural processing in the auditory brainstem using auditory brainstem response (ABR) mean and interaural wave latencies and binaural difference responses and in the cortex using cortical auditory evoked potentials (Bauer et al., 2006, Gordon et al., 2007a; Gordon et al., 2007b; Gordon et al., 2008; Sharma et al., 2005). A child's ability to perform speech recognition in noise and sound localization tasks is highly dependent upon the nature of the interaural auditory brainstem and cortical responses. Interaural timing differences between ears (indicated by response latency differences) make sound localization extremely challenging for children with sequential implantations- even when the BICI processors are programmed in synchrony (Gordon et al., 2007b). The identified studies had not been included in previously published reviews of paediatric bilateral cochlear implantation (Brown & Balkany, 2007; Ching et al., 2007; Murphy & O'Donoghue, 2007). All five studies suggested that there was a sensitive period, from birth to age 3, for central auditory development of bilateral processing. ABR responses showed prolonged wave latencies in the second implanted ear when implanted sequentially compared to simultaneous surgeries (Gordon et al., 2007b; Gordon et al., 2008. Longitudinal studies indicated that wave latencies improved over time- particularly in children implanted before age 3 (Gordon et al., 2007a; Gordon et al., 2007b; Gordon et al., 2008). Cortical auditory evoked potentials, as measured by P1 latency and morphology, were fundamentally different among children implanted before 3.5 years and after 3.5 years (Bauer et al., 2006; Sharma et al., 2005). Early implanted

children showed rapid development of morphology and P1 latency whereas the late implanted children showed malformed wave morphology and slower P1 latency (Bauer et al., 2006; Sharma et al., 2005).

Language Studies. Only one older case study examined speech and language in a bilaterally implanted child (Vermeire et al., 2003). This child received her first implant at 2.5 years and her second at age 4. The child was diagnosed with auditory neuropathy with the etiology of her hearing loss being hyperbilirubinemia. Results indicated that speech and language development at 4.5 years was within the normal range for this child.

Other Measures. A single identified study used a standardized paediatric quality of life measure (PedsQL) in children with bilateral CIs (Beijen et al., 2007). No difference was seen between the bilaterally implanted children and the unilaterally implanted comparison group on the PedsQL measure. Five other studies used a qualitative approach or a study-designed questionnaire to obtain information on parental report of benefits from bilateral CI in addition to the other outcomes of interest (Galvin et al., 2007b; Gregoret, 2003; Kuhn-Inacker et al., 2004; Mueller et al., 2000; Senn et al., 2005). These studies indicated that, in general, parental and teen reports of benefits are positive regarding the perceived benefits from the second CI. Perceived benefits were in the form of sound localization and hearing in noise.

Discussion

Research methodology

Similar to the Thoutenhoofd et al. (2005) review of unilateral paediatric CIs, the variability in the study designs and outcome measures used in the literature published to date is high. There are significant challenges in drawing conclusions about the effectiveness of bilateral CIs from case studies or uncontrolled research studies with small, convenience samples (Oleckno, 2002). While these studies are valuable in the early development stages of an intervention, their value in assessing effectiveness of treatment is limited due to their lack of comparison control groups against which one can evaluate treatment outcomes. Eleven of the 24 primary research studies had samples of fewer than nine participants (Bauer et al., 2006; Beijen, et al., 2007; Galvin et al., 2007b; Gregoret, 2003; Litovsky et al., 2004a; Litovsky, 2004b; Mok, et al., 2007; Mueller et al., 2000; Senn et al., 2005; Sharma et al., 2005; Vermeire, et al., 2003).

Sound localization and speech recognition in noise results

The outcomes that have been studied in the BICI literature focus primarily on binaural hearing ability in children by measuring their performance on sound localization and speech recognition in noise tasks. There was some variability in the measures used for sound localization (MAA and localization of pink noise) and speech recognition in noise (CRISP, AdSpon, study designed measures) and of the testing procedures (stimuli and noise presentation levels, presentation angles). In general, the results from available studies indicated improved performance for children using BICIs compared to using one CI for both sound localization and speech recognition in noise. When similar studies were compared,

results indicated that a greater number of children performed sound localization and speech recognition in noise tasks at levels comparable to normally hearing children using BICIs compared to a single CI. While these differences were not statistically significant for the speech recognition in noise results, the studies were not adequately powered to determine if a difference exists between the children using BICIs compared to a CI. The 13% difference between groups that was obtained would be statistically significant had a sample of 41 children been obtained in the literature, but it is not statistically significant with the current sample of 16.

The results of this review are consistent with other published reviews (Brown & Balkany, 2007; Murphy & O'Donoghue, 2007). These auditory skills are not only important for personal safety (e.g. identifying sounds in one's environment) but for school and social environments. Research on children with unilateral hearing loss has emphasized the importance of binaural input in the classroom and its positive influence on school achievement (Bess et al., 1998).

The improvements in sound localization and speech recognition in noise for children using BICIs were more consistent for those children receiving their second implant within four years of the onset of hearing loss in some (Peters et al., 2007; Litovsky et al., 2006a; Litovsky et al., 2004a; Steffens et al. 2007) but not all studies (Kuhn-Inacker et al., 2004; Litovsky et al., 2006a; Wolfe et al., 2007). It appears that the greatest improvements in speech understanding in noise and sound localization are seen in children who were implanted early and with a short gap between surgeries. Within the unilateral implantation literature, the age of unilateral implantation and duration of deafness have also been

identified as important factors in auditory function and speech and language development in children with unilateral implants (Connor et al., 2006; Harrison et al., 2005).

Electrophysiological results

The authors of the electrophysiological studies explored the auditory brainstem and cortical responses in children with simultaneous and sequential BICIs (Bauer et al., 2006, Gordon et al., 2007a; Gordon et al., 2007b; Gordon et al., 2008; Sharma et al., 2005). They were particularly interested in the influence of the timing of a second implant on either ABR or P1 morphology and latencies. Many of the studies compared children implanted bilaterally with a short delay between first and second implant (1-3.5 years) to children implanted with a longer delay between bilateral devices (greater than 3.5 years). Children with shorter periods of unilateral deafness showed improved morphology and consistent latencies on P1 and ABR recordings (Bauer et al., 2006, Gordon et al., 2007a; Gordon et al., 2007b; Gordon et al., 2008; Sharma et al., 2005). Unlike the behavioural measures of speech recognition in noise and sound localization that showed variability in the influence of age of second implantation, the improvements in wave morphology and latency among early implanted children were consistent in all identified studies that used electrophysiological measures (Bauer et al., 2006, Gordon et al., 2007a; Gordon et al., 2007b; Gordon et al., 2008; Sharma et al., 2005). One would expect greater variability in the behavioural responses due to the confounding influence of attention, memory, and recall (Harrison et al., 2005).

Additional research to examine the influence of time between implant surgeries on both electrophysiological and behavioural measures should be conducted. Such research provides

important information regarding auditory plasticity and critical periods for auditory development. However, based on the available studies of electrophysiological, speech recognition in noise, and sound localization measures there appears to be support for reduced periods of time between cochlear implant surgeries (and therefore reduced periods of auditory deprivation for the second ear).

Speech, language, and quality of life results

Less information is available on the longer-term outcomes of speech, language, and quality of life. One of the objectives of this review was to identify, beyond speech recognition in noise and sound localization studies, what additional benefits might be accrued through bilateral implantation. As the bilateral implantation procedure is relatively recent, little research on these outcomes is available. The single case study examining language in a child with a BICI (Vermeire et al., 2003) cannot be extrapolated to other children due the sample of one and lack of a control group. No other identified studies explored speech, language, or other psycho-educational outcomes in children with bilateral CIs. As more children are implanted bilaterally, studies with larger sample sizes will likely emerge as has been the case with the unilateral CI literature (Moog & Geers, 2003; Thoutenhoofd et al., 2005). Large samples will be essential in identifying if BICIs are one of the factors associated with improved post-implant language outcomes. Moog and Geers (2003) have identified that higher IQ, oral and mainstream classrooms, higher socioeconomic status, and smaller families are among some of the family and education characteristics associated with improved speech perception, production, and language outcomes in children with unilateral implantation. Implant characteristics such as speech coding strategy, more electrodes, larger

dynamic range, and loudness growth are some of the implant characteristics identified associated with better post-implant outcomes. Future large-scale studies will likely include BICIs as one of the implant characteristics that may contribute to positive overall outcomes in children.

Quality of life improvements have been identified in children with unilateral CIs compared to non-implanted children- particularly those children who receive a CI before age 5 (Stacey et al., 2006). It is still unclear if quality of life will be further improved with a second CI. Only a single identified study used a standardized quality of life measure, and there was no significant improvement reported between the children with BICI and the children with one CI (Beijen et al., 2007). However, this measure may not be sensitive to the benefits accrued through a second CI. The other studies that used qualitative or study designed measures to gathering information on parental and teen perceptions of benefit generally reported positive perceptions of improvement with the second CI (Kuhn-Inacker et al., 2004; Senn et al., 2005). In some studies, the benefits were not perceived by all participants (Galvin et al., 2007; Gregoret, 2003; Mueller et al., 2000). In order to understand the benefits received by bilaterally implanted children, exploratory qualitative research may identify some of the important outcomes associated with BICI use. Some of these may include parent child interaction, school performance, or peer relationships.

Other outcomes of interest

The examination of the paediatric BICI literature provides evidence that speech recognition in noise, sound localization, and electrophysiological responses improve with the use of a

second CI. However, Tyler et al. (2003) offer the following caveat, "It should also be apparent that providing a second implant results in additional hardware expenses, surgical risk, and programming time. It will be necessary to evaluate the additional benefit provided by the second device in the context of these additional costs and risks." No cost-benefit or cost-utility analyses for bilateral implantation in children were identified in the literature. A cost-utility analysis of BICIs for post-lingually deafened adults concludes that, "more quality of life is likely to be gained per unit of expenditure on unilateral implantation than bilateral implantation" (Summerfield et al., 2002). The authors did not identify the significant changes in quality of life with the second CI that have been identified with the first. However, these results cannot be extrapolated to children due to the very different nature of providing auditory stimulation to pre-lingually deafened children and the potential for benefit from early language, auditory stimulation, and binaural hearing, particularly in the school environment. It will be important for further research to examine the benefits, costs, and risks associated with the second implant in children in family, school, and social environments.

One way to explore this further is to identify parents' (and older children's) perceptions of costs, risks, and benefits of bilateral cochlear implantation. Only a single study was identified in the literature that provided insight into the needs and concerns of parents when making the BICI choice (Gregoret, 2003). It included the perceptions of four parents and highlighted the variability in parental perceptions of bilateral implantation. With a limited number of CI surgeries available in many sites, it may be important to ensure that those receiving the second implant value the benefits accrued by it and find the risks acceptable. The BICI decision is a choice that carries both harms and benefits that may be differentially viewed depending upon the personal preferences of the client. Incorporating patient

preference into the clinical pathway can be an important next-step in the creation of BICI guidelines (Krahn & Naglie, 2008)

This critical review has provided compelling evidence on the benefits of bilateral cochlear implantation for children on sound localization, speech recognition in noise, and electrophysiological responses. There are still additional research and clinical questions that remain unanswered including: 1) Does the use of BICIs lead to improvements in quality of life compared to use of a single CI in children?, 2) What are the perceived parental benefits and risks associated with bilateral cochlear implantation?, 3) Which children, in terms of age and severity of loss, benefit from bilateral implantation?, 4) Will BICIs facilitate the integration of children into the school system?

Limitations

Systematic reviews can be excellent means of synthesizing research. However, the conclusions that can be arrived at using systematic review methods are entirely dependent upon the quality of research that has been previously conducted. Due to the recent introduction of BICIs into both the clinical and research fields, there are limited articles available on the topic. The available articles include many studies with small samples without comparison groups, varied testing conditions and outcome measures that preclude overall quantitative comparisons.

The BICI topic is a relatively new area of research. As such, it is expected that additional publications will be emerging in the near future. These articles will hopefully provide additional information for parents and clinicians. This review did not seek out unpublished

results or conference proceedings which may have the results of bilateral studies currently underway. Due to the timeliness of this topic, this critical review should be updated within two years in order to include newly emerging studies.

Conclusions

A replicable, critical review of paediatric bilateral cochlear implantation was conducted. When compared with previously published reviews on the topic, additional paediatric studies were identified that examined outcomes of interest beyond speech recognition in noise and sound localization. The available articles on bilateral implantation in children provide evidence that there are benefits to children who receive a second CI. The benefits include improved performance on speech recognition in noise and sound localization tasks, and auditory brainstem and cortical evoked potential responses. Children with BICIs that are activated at earlier ages and with shorter gaps between surgeries appear to receive greater benefit than those implanted later and with longer gaps between surgeries. Well designed and controlled studies that explore a variety of additional outcomes- including cost-effectiveness, quality of life, speech, language and psycho-educational outcomes should be further explored in order to provide additional support for parents and clinicians facing the BICI decision. Identifying the factors that lead to successful bilateral implantation as well as the potential risks associated with a second cochlear implant should also be a priority.

References

Bauer, P.W., Sharma, A., Martin, K. & Dorman, M. 2006. Central auditory development in children with bilateral cochlear implants, *Arch Otolaryngol Head Neck Surg,* 132, 1133-6.

Beijen, J.W., Snik, A.F. & Mylanus, E.A. 2007. Sound localization ability of young children with bilateral cochlear implants., *Otol Neurotol* 28, 479-85.

Berg, A.L., Ip, S.C., Hurst, M. & Herb, A. 2007. Cochlear implants in young children: informed consent as a process and current practices, *Am J Audiol,* 16, 13-28.

Bess, F.H., Dodd-Murphy, J. & Parker, R.A. 1998. Children with minimal sensorineural hearing loss: Prevalence, educational performance, and functional status. *Ear Hear,* 19, 339-354.

Brown, K.D. & Balkany, T.J. 2007. Benefits of bilateral cochlear implantation: a review, *Curr Opin Otolaryngol Head Neck Surg,* 15, 315-8.

Ching, T.Y., van Wanrooy, E. & Dillon, H. 2007. Binaural-bimodal fitting or bilateral implantation for managing severe to profound deafness: a review, *Trends Amplif,* 11, 161-92.

Colburn, H.S., Shinn-Cunningham, B., Kidd, G. & Durlach, N. 2006. The perceptual consequences of binaural hearing, *Int J Audiol,* 45, S34-S44.

Connor, C.M., Craig, H.K., Raudenbush, S.W., Heavner, K. & Zwolan, T.A. 2006. The age

at which young deaf children receive cochlear implants and their vocabulary and speech-production growth: Is there an added value for early implantation? *Ear Hear,* 27, 628-644.

Food and Drug Administration. 2008. Devices at FDA: Nuleus 22 Channel Cochlear Implant System in Children. Retrieved online at: http://www.accessdata.fda.gov/scripts/cdrh/devicesatfda/index.cfm?db=PMA&id=5000 on August 6, 2008.

Galvin, K.L., Mok, M. & Dowell, R.C. 2007a. Perceptual benefit and functional outcomes for children using sequential bilateral cochlear implants, *Ear Hear*, 28, 470-82.

Galvin, K.L., Mok, M., Dowell, R.C. & Briggs, R.J. 2007b. 12-month post-operative results for older children using sequential bilateral implants, *Ear Hear*, 28, 19S-21S.

Geers, A.E., Nicholas, J.G. & Sedley, A.L. 2003. Language skills of children with early cochlear implantation. *Ear Hear,* 24, 46S-58S.

Gordon, K.A., Valero, J. & Papsin, B.C. 2007a. Binaural processing in children using bilateral cochlear implants, *Neuroreport*, 18, 613-17.

Gordon, K.A., Valero, J. & Papsin, B.C. 2007b. Auditory brainstem activity in children with 9-30 months of bilateral cochlear implant use, *Hear Res*, 233, 97-107.

Gordon, K.A., Valero, J. & Papsin, B.C. 2008. Abnormal timing delays in auditory

brainstem responses evoked by bilateral cochlear implant use in children, *Otol Neurootol*, 29, 193-8.

Gregoret, L. 2003. Bilateral cochlear implants: are they in your child's future?, *Volta Voices*, 10, 10-5, 34.

Grieco-Calub, T.M., Litovsky, R.Y. & Werner, L.A. 2008. Using the observer-based psychophysical procedure to assess localization acuity in toddlers who use bilateral cochlear implants, *Otol Neurootol*, 29, 235-9.

Harrison, R.V., Gordon, K.A. & Mount, R.J. 2005. Is there a critical period for cochlear implantation in congenitally deaf children? Analyses of hearing and speech perception performance after implantation. *Dev Psychobiol,* 46, 252-261.

Jadad A. & Moore A. 1996. Assessing the quality of reports of randomized clinical trials: Is blindness necessary? *Control Clin Trials*, 17, 1–12.

Krahn, M. & Naglie, G. 2008. The next step in guideline development: Incorporating patient preferences. *JAMA*, 300, 436-438.

Kuhn-Inacker, H., Shehata-Dieler, W., Muller, J. & Helms, J. 2004. Bilateral cochlear implants: a way to optimize auditory perception abilities in deaf children?, *Int J Pediatr Otorhinolaryngol*, 68, 1257-66.

Litovsky, R.Y. 1997. Developmental changes in the precedence effect: Estimates of minimal audible angle. *J Acoust Soc Amer*, 102, 1739-1745.

Litovsky, R.Y., Johnstone, P.M. & Godar, S.P. 2006a. Benefits of bilateral cochlear implants and/or hearing aids in children, *Int J Audiol*, 45, S78-91.

Litovsky, R.Y., Johnstone, P.M., Godar, S., Agrawal, S., Parkinson, A. et al., 2006b. Bilateral cochlear implants in children: localization acuity measured with minimum audible angle, *Ear Hear*, 27, 43-59.

Litovsky, R.Y., Johnstone, P.M., Parkinson, A., Peters, R. & Lake, J. 2004a. Bilateral cochlear implants in children. *Intl Congress Series*, 1273, 451-454.

Litovsky, R.Y., Parkinson, A., Arcaroli, J., Peters, R., Lake, J., et al. 2004b. Bilateral cochlear implants in adults and children, *Arch Otolaryngol Head Neck Surg*, 130, 648-55.

Mencher, G.T. & Davis, A. 2006. Bilateral or unilateral amplification: is there a difference? A brief tutorial, *Int J Audiol, 45, S3*-S11.

Mok, M., Galvin, K.L., Dowell, R.C. & McKay, C.M. 2007. Spatial unmasking and binaural advantage for children with normal hearing, a cochlear implant and a hearing aid, and bilateral implants, *Audiol Neurootol,* 12, 295-306.

Moog, J. S. & Geers, A. E. 2003. Epilogue: Major findings, conclusions and implications for

deaf education. *Ear Hear,* 24, 121S-125S.

Mueller, J., Schoen, F. & Helms, J. 2000. Bilateral cochlear implant--new aspects for the future?, *Adv Otorhinolaryngol*, 57, 22-7.

Murphy, J. & O'Donoghue, G. 2007. Bilateral cochlear implantation: an evidence-based medicine evaluation, *Laryngoscope*, 117, 1412-18.

Nucleus ® 24 Candidacy Criteria. 2004. Retrieved online at http://www.cochlear.com/Support/631.asp on February 9, 2008

Oleckno, W.A. 2002. *Essential Epidemiology*. Prospect Heights, IL: Waveland Press Inc.

Peters, B.R., Litovsky, R., Lake, J. & Parkinson, A. 2004. Sequential bilateral cochlear implantation in children, *Intl Congress Series*, 1273, 462-5.

Peters, B.R., Litovsky, R., Parkinson, A. & Lake, J. 2007. Importance of age and postimplantation experience on speech perception measures in children with sequential bilateral cochlear implants, *Otol Neurotol*, 28, 649-57.

Rance, G., Barker, E., Mok, M., Dowell, R., Rincon, A., et al. 2007. Speech Perception in Noise for Children with Auditory Neuropathy/Dys-Synchrony Type Hearing Loss, *Ear Hear,* 28, 351-360.

Schafer, E.C., Amlani, A.M., Seibold, A. & Shattuck, P.L. 2007. A meta-analytic comparison of binaural benefits between bilateral cochlear implants and bimodal stimulation. *J Am Acad Audiol,* 18, 760-776.

Schafer, E.C. & Thibodeau, L.M. 2006. Speech Recognition in noise in children with cochlear implants while listening in bilateral, bimodal, and FM-system arrangements, *Am J Audiol*, 15, 114-26.

Senn, P., Kompis, M., Vischer, M. & Haeusler, R. 2005. Minimum audible angle, just noticeable interaural differences and speech intelligibility with bilateral cochlear implants using clinical speech processors, *Audiol Neurootol*, 10, 342-52.

Shafir, D.N. 2007. Study researches bilateral cochlear implants, *ASHA Leader*, 5, 19.

Sharma, A., Dorman, M.F. & Kral, A. 2005. The influence of a sensitive period on central auditory development in children with unilateral and bilateral cochlear implants, *Hear Res*, 203, 134-43.

Stacey, P.C., Fortnum, H.M., Barton, G.R. & Summerfield, A.Q. 2006. Hearing implaired children in the United Kingdom, I: auditory performance, communication skills, educational achievements, quality of life, and cochlear implantation. *Ear Hear*, 27, 161-86.

Steffens, T., Lesinski-Schiedat, A., Strutz, J., Aschendorff, A., Klenzner, T., et al. 2007. The benefits of sequential bilateral cochlear implantation for hearing-impaired children. *Acta*

Oto-Laryngologica, 128, 164-176.

Summerfield, A.Q., Marshall, D.H., Barton, G.R. & Bloor, K.E. 2002. A cost-utility scenario analysis of bilateral cochlear implantation, *Arch Otolaryngol Head Neck Surg*, 128, 1255-62.

Thoutenhoofd, E., Archbold, A., Gregory, S., Lutman, M., Nikolopoulos, T. et al. 2005. *Paediatric Cochlear implantation: Evaluating Outcomes*. London: Whurr.

Tyler, R.S., Dunn, C.C., Witt, S.A. & Preece, J.P. 2003. Update on bilateral cochlear implantation. *Curr Opin Otolaryngol Head Neck Surg*, 11, 388-93.

Vermeire, K., Brokx, J.P., Van de Heyning, P.H., Cochet, E. & Carpentier, H. 2003. Bilateral cochlear implantation in children, *Int J Pediatr Otorhinolaryngol*, 67, 67-70.

Wells, G.A., Shea, B., O'Connell, D., Peterson, J., Welch, V. et al. 2005. The Newcastle-Ottawa Scale (NOS) for assessing the quality of nonrandomised studies in meta-analyses. Retrieved online at: http://www.ohri.ca/programs/clinical_epidemiology/oxford.htm. on November 16, 2005.

Wolfe, J., Baker, S., Caraway, T., Kasulis, H., Mears, A., et al. 2007. 1-year postactivation results for sequentially implanted bilateral cochlear implant users, *Otol Neurotol,* 28, 589-96.

Zurek, P.M. 1993.Binaural advantages and directional effects in speech intelligibility. In: Studebaker GA, Hochberh I, eds. *Acoustical Factors Affecting Hearing Aid Performance*. 2nd

ed. Boston, MA: Allyn & Bacon; 1993:255-276.

Table 1: Paediatric bilateral cochlear implantation review inclusion/exclusion criteria

Component	Inclusion	Exclusion
Patient population	Children and Youth (0-18 yrs) Published in English or French	Only adults
Intervention (or Exposure)	Bilateral Cochlear Implant Surgery	Other types of auditory, ear, or head surgeries
Method	Published in peer-reviewed source, Systematic Reviews, Meta-analyses, Randomized Controlled Trials, Cohort Studies, Case Controls, Cross-sectional, Retrospective and Prospective Case Studies, Qualitative Studies	Theoretical papers Opinion-based Editorial Commentaries
Outcomes	Quality of Life Auditory, Audiology, Language, Speech, Social Development and Skills, Learning and School Achievement, Electrophysiological, Economic	Medical and Surgical

Table 2: A summary of the included articles, the characteristics of each study, their results, and conclusions by outcomes of interest in the studies.

Authors and Location	Title *Journal* (Year)	# of Paediatric Studies Included	Population	Outcomes Included in Review	Results and Authors' Conclusions
Paediatric Review Articles					
Brown & Balkany FL, USA	Benefits of bilateral cochlear implantation: a review *Curr Opin Otolaryngol Head Neck Surg* (2007)	5	Children and adults	Sound localization Speech Recognition in noise Qualitative improvement	- Speech Recognition in noise and sound localization seems improved in children receiving BICI compared to CI - The head shadow effect provides greater benefit to bilateral users than binaural squelch or summation
Ching, van Wanrooy, & Dillon Australia	Binaural-bimodal fitting or bilateral implantation for managing severe to profound deafness: a review *Trends Amplif* (2007)	6	Children and adults	Sound localization Speech perception in quiet and in noise	- Both BICI and CIHA methods of bilateral stimulation reported some binaural benefits when compared with CI - No available controlled comparisons between CIHA and BICI
Murphy & O'Donoghue UK	Bilateral cochlear implantation: An evidence-based medicine evaluation *The Laryngoscope* (2007)	9	Children and adults	Sound localization Speech Recognition in noise	- Improvements in sound localization and understanding of speech in noise for children with BICI - Simultaneous or sequential surgeries with short intervals seem most advantageous
Schafer, Amlani, Seibold & Shattuck TX, USA	A meta-analytic comparison of binaural benefits between bilateral cochlear implants and bimodal stimulation J Am Acad Audiol (2007)	4	Children and adults	Binaural summation and squelch Head-shadow effect	- Improvements in outcome measures for BICI and CIHA conditions compared to CI condition - No significant differences between BICI or CIHA conditions

Authors and Location	Title *Journal* (Year)	Study Design	Comparison Group	Sample Size	Sequential / Simultaneous	Population Study Age	Outcomes measured	Results and Authors Conclusions
Electrophysiological Studies								
Bauer, Sharma, Martin & Dorman TX, USA	Central auditory development in children with bilateral cochlear implants *Arch Otolaryngol Head Neck Surg* (2006)	Case series	No	N=4	2 / 2	Children < 2 years	P1 response latencies	- In 2 pts with sequential BICI, P1 latencies were WNL within 3-6 months of 2nd implantation - In 2 pts receiving simultaneous BICI, P1 latencies were WNL by 1 month post implant.
Gordon, Valero & Papsin Toronto, Canada	Auditory brainstem activity in children with 9-30 months of bilateral cochlear implant use. *Hear Res* (2007)	Cohort study	No	N=13	13 / 2	Children 2-12 years	Auditory Brainstem Response (ABR) mean latencies, inter-wave latencies Waves I-V	- Prolonged wave latencies in children implanted after a delay - Reduced wave latencies over time- particularly in children implanted before age 3. - ABR results suggest a negative impact of unilateral implant use on bilateral auditory brainstem plasticity
Gordon, Valero & Papsin Toronto, Canada	Abnormal timing delays in auditory brainstem responses evoked by bilateral cochlear implant use in children *Otol Neurotol* (2008)	Cohort study	No	N=46	31 / 15	Children < 3 years	Same as above	Same as above
Gordon, Valero & Papsin Toronto, Canada	Binaural processing in children using bilateral cochlear implants *Neuroreport* (2007)	Cohort study	age match ed CI group	N=40 BICI N=40 CI	30 / 10	Children Mean = 6 years	Same as above	- Shorter wave latencies in children with experienced versus naive implanted ear - Prolonged latencies in those implanted sequentially compared to simultaneously
Sharma, Dorman & Kral TX, USA	The influence of a sensitive period on central auditory development in children with unilateral and bilateral cochlear implants *Hear Res* (2005)	Cohort study	Early vs late CI group	N=2 BICI N=21 CI	All sequential	Children 1 and 9 years	cortical auditory evoked potentials (CAEP) P1 response latencies	- CAEP morphology development is different among early (< 3.5 yrs) and late implanted children (3.5-7 yrs) - Early implanted children showed rapid development of morphology and P1 latency, late implanted children show malformed wave morphology and slower P1 latency

Authors and Location	Title *Journal* (Year)	Study Design	Comparison Group	Sample Size	Sequential /Simultaneous	Population Study Age	Outcomes measured	Results and Authors Conclusions
Sound Localization and Detection Studies								
Beijen, Snik & Mylanus Netherlands	Sound localization ability of young children with bilateral cochlear implants. *Otol Neurotol* (2007)	Case-control study	CI group	N=5 BICI N=5 CI	1 / 4	Children BICI Mean 3yr, 7mo CI Mean 5yr 3 mo	-% sound localizations correct at 90 and 30 degrees -PedsQL -Speech, Spatial, and Qualities of Hearing Scale (SSQ)	- Significantly better scores on localization test for BICI children - Significantly better scores on parent reported spatial domain of SSQ - No differences in SSQ speech and quality of hearing domains or Peds QL
Grieco-Calub, Litovsky & Werner WI, USA	Using the observer-based psychophysical procedure to assess localization acuity in toddlers who use bilateral cochlear implants *Otol Neurotol* (2008)	Cohort study	normal hearing, CI group	N=10 BICI N=8 normal hearing N=8 CI	All sequential	Children 2-3 years	- Minimum Audible Angle (MAA) as measured by a 2-alternative-forced choice task	- Localization acuity seems to be developing in half of the BICI children - None of the CI children could perform the task above chance
Litovsky, Johnstone, Godar, Agrawal, Parkinson, Peters & Lake WI, USA	Bilateral cochlear implants in children: localization acuity measured with minimum audible angle *Ear Hear* (2006)	Cohort study	CI group	N=13 BICI N=6 CIHA	All sequential	Children 3-16 years	- MAA as measured by a 2-alternative-forced choice task	- In the MAA test of location acuity, 9/13 BICI children were able to discriminate left/right compared to CI condition - High inter-subject variability
Mok, Galvin, Dowell & McKay Australia	Spatial unmasking and binaural advantage for children with normal hearing, a cochlear implant and a hearing aid, and bilateral implants. *Audiol Neurootol* (2007)	Cohort study	NH, CIHA group	N=4 BICI N=10 Normal Hearing N=9 CIHA	All sequential	Children 7-12 years	- Loudness balancing - Sound detection thresholds measured in noise from 0 and 90 degrees L and R	- Spatial unmasking was present in both BICI and CIHA children - Binaural advantage in both CI and BICI groups - Head shadow was present in both BICI and CIHA, although greater in BICI children

Authors and Location	Title *Journal* (Year)	Study Design	Comparison Group	Sample Size	Sequential /Simultaneous	Population Study Age	Outcomes measured	Results and Authors Conclusions
Speech Recognition or Intelligibility and/or Language Studies								
Kuhn-Inacker, Shehata-Dieler, Muller, & Helms Germany	Bilateral cochlear implants: a way to optimize auditory perception abilities in deaf children? *Int J Pediatr Otorhinolaryngol* (2004)	Cohort study	No	N=18	17 / 1	Children 3-8 years	-Bi-syllabic word recognition in quiet and in noise -Qualitative observations	- Speech in Noise Test- 17/18 scored better with BICI than CI - Qualitative data show BICI improves communicative behaviour
Mueller, Schoen & Helms Germany	Bilateral cochlear implant--new aspects for the future? *Adv Otorhinolaryngol* (2000)	Case series	No	N=3	2 / 1	Children and adults 4-17 years	-Word and sentence recognition in quiet and noise	- Better performance on monosyllabic and sentence discrimination in quiet and noise with BICI - Some children and parents reported improved hearing in noise with BICI
Peters, Litovsky, Parkinson & Lake Multi-site, USA	Importance of age and post-implantation experience on speech perception measures in children with sequential bilateral cochlear implants. *Otol Neurotol* (2007)	Cohort study	No	N=30	All sequential	Children 3-13 years	-Speech Recognition: -in quiet (MLNT, LNT, HINT-C) -in noise (CRISP)	- Significant improvement in speech perception in 2nd ear over the first year following the 2nd CI surgery - Significant improvements in BICI noise condition, not in quiet - Biggest improvements in children who received the 2nd implant earlier
Schafer & Thibodeau TX, USA	Speech Recognition in noise in children with cochlear implants while listening in bilateral, bimodal, and FM-system arrangements *Am J Audiol* (2006)	Cohort study	CIHA group and CIFM group	N=12 BICI N=10 CIHA, CIFM	All sequential	Children 3-12 years	-Speech Recognition in noise thresholds with study created phrases	- Speech in noise thresholds did not improve with a 2nd CI or HA relative to a single CA - FM system thresholds were up to 20 dB lower than in all other conditions.
Vermeire, Brokx, Van de Heyning, Cochet & Carpentier	Bilateral cochlear implantation in children *Int J Pediat Otorhinolaryngol* (2003)	Case study	No	N=1	All sequential	Children 4.5 years	-Sentence identification and recognition in quiet -Speech and Language Scores	- Speech and Language scores are within the normal range for a child her age - Children with Auditory Neuropathy may receive benefit from cochlear implantation.

Belgium								
Wolfe, Baker, Caraway, Kasulis, Mears, Smith, Swim, & Wood OK, USA	1-year post-activation results for sequentially implanted bilateral cochlear implant users. *Otol Neurotol* (2007)	Cohort study	No	N=12	All sequential	Children 3-10 years	-Speech Recognition: -in quiet (MLNT, ESPT) -in noise (Spondee recognition threshold in speech weighted noise at 45dB HL)	- Speech recognition in quiet was improved in the early-implanted children (< 4 yr of age) with BICI - All children possessed better speech recognition scores in noise in the BICI relative to CI
Authors and Location	Title *Journal* (Year)	Study Design	Comparison Group	Sample Size	Sequential /Simultaneous	Population Study Age	Outcomes measured	Results and Authors Conclusions
Studies with Speech Recognition or Intelligibility and Sound Localization Combined								
Galvin, Mok & Dowell Australia	Perceptual benefit and functional outcomes for children using sequential bilateral cochlear implants. *Ear Hear* (2007)	Cohort study	No	N=11	All sequential	Children 4-15 years	-Localization of pink noise at 70dBA -Speech Recognition in noise (AdSpon) - Parental report - SSQ	- Speech recognition indicated improvement with BICI in ipsi-lateral noise, No benefit contra-lateral noise - No improvement in localization with BICI - SSQ showed improvement for the spatial subscale - 6/10 parents report improved performance in daily life
Galvin, Mok, Dowell & Briggs Australia	12-month post-operative results for older children using sequential bilateral implants. *Ear Hear* (2007)	Case series	No	N=6	All sequential	Children 5-15 years	-Localization of pink noise at 70dBA -Speech Recognition in noise (AdSpon)	- No significant improvement in localization or Spondee Recognition in BICI compared to CI condition - Parent and self-report of perceived BICI benefits varied and was consistent with objective measures

Litovsky, Johnstone, Parkinson, Peters & Lake WI, USA	Bilateral cochlear implants in children *Intl Congress Series* (2004)	Case series	No	N=3	All sequential	Children 8-13 years	-Localization of pink noise at 60dB SPL using a 2-alternative-forced choice task -Speech Recognition (CRISP)	- Speech Recognition in noise improves with bilateral experience - No improvement in speech recognition in quiet or in sound localization with BICI
Litovsky, Johnstone & Godar WI, USA	Benefits of bilateral cochlear implants and/or hearing aids in children *Int J of Audiol* (2006)	Cohort study	CI group	N=10 BICI N=10 CIHA	All sequential	Children 4-14 years	-MAA as measured by a 2-alternative-forced choice task -Speech Recognition (CRISP)	- Improved MAA and speech reception thresholds - High individual variability - Benefits from binaural fittings-whether with BICI or HA - Children with BICI outperform the CIHA children in speech recognition and localization acuity.
Litovsky, Parkinson, Arcaroli, Peters, Lake, Johnstone & Yu WI, USA	Bilateral cochlear implants in adults and children *Arch Otolaryngol Head Neck Surg* (2004)	Case series	No	N=3	All sequential	Children and adults 8-12 years	-Localization of pink noise at 60dB SPL using a 2-alternative-forced choice task -R/L discrimination -Speech recognition (CRISP)	- No significant improvement in localization and R/L discrimination with BICI - Speech tasks slightly improved in 2/3 children with BICI - The subjects in this study were 3 months post 2nd CI and had little time to fully adapt to the BICI
Peters, Litovsky, Lake & Parkinson Multi-site, USA	Sequential bilateral cochlear implantation in children *Intl Congress Series* (2004)	Cohort study	No	N=25	All sequential	Children 3-13 years	-MAA as measured by a 2-alternative-forced choice task -Speech Recognition: -in quiet (MLNT, LNT, HINT-C) -in noise (CRISP)	- Sound localization was not improved - Improved speech recognition in quiet and in noise with BICI

Senn, Kompis, Vischer, & Haeusler Germany	Minimum audible angle, just noticeable inter-aural differences and speech intelligibility with bilateral cochlear implants using clinical speech processors *Audiol Neurootol* (2005)	Case-control study	NH matched for age	N=2 BICI	All sequential	Children and adults 14 years	-MAA as measured by a 2-alternative-forced choice task -Speech Recognition in quiet and in noise -Questionnaire Re: benefit of BICI	- Improved sound localization with BICI - Speech recognition in noise was significantly improved with BICI- no change in quiet - Self-reports indicate that teens see some - much improvement with BICI
Steffens, Lesinski-Schiedat, et al. Germany	The benefits of sequential bilateral cochlear implantation for hearing-impaired children *Acta Oto-Laryngologica* (2008)	Cohort study	No	N=20 BICI	All sequential	Children 3-10 years	-Localization of white noise at 60dB SPL using a 2-alternative-forced choice task -Speech Recognition in noise	- Improved sound localization with BICI - Speech recognition in noise was significantly improved with BICI condition

Authors and Location	Title *Journal* (Year)	Study Design	Comparison Group	Sample Size	Sequential /Simultaneous	Population Study Age	Outcomes measured	Results and Authors Conclusions
Other Outcomes								
Berg, Ip, Hurst & Herb NY, USA	Cochlear implants in young children: informed consent as a process and current practices. *Am J of Audiol* (2007)	National Survey	No	47% response rate from CI centres	17% of CI centres reported performing simultaneous BICI	Paediatric CI centres	-Information provided to parents -Consent process -Bilateral implant process	- Differing rates of bilateral services reported across the country - Decisions should be based on informed risk-benefit analysis by parents and professionals working together
Gregoret MA, USA	Bilateral cochlear implants: are they in your child's future? *Volta Voices* (2003)	Case series	No	N=4	sequential /simultaneous	Children 5-6 years	-Parental perceptions	- Varying parental requests for and reactions to 2nd cochlear implant-generally positive

ABR: auditory brainstem response, AdSpon: Adaptive Spondee Test, BICI: bilateral cochlear implant, CAEP: cortical auditory evoked potentials, CI: cochlear implant, CIHA: cochlear implant and hearing aid, CRISP: Children's Realistic Index of Speech Perception, ESPT: Early Speech Perception Test, FM: frequency modulation, HA: Hearing Aid, HINT: Hearing in Noise Test, LNT: Lexical Neighbourhood Test, MAA: Minimum Audible Angle, MLNT: Multisyllabic Lexical Neighbourhood Test, N: number of participants, NH: normal hearing, PedsQL: Pediatric Quality of Life Measure, R/L: right/left, SRM: Spatial Release from Masking, SSQ: Speech, Spatial, and Qualities of Hearing Scale, WNL: within normal levels

Table 3: Sound localization performance as measured by the Minimum Audible Angle (MAA) at various thresholds for children using a single cochlear implant (CI) and bilateral cochlear implants (BICI)

Study	BICI Subjects	Thresholds for sound localization	Number of kids performing task at or below this threshold with CI	Number of kids performing task at or below this threshold with BICI
Litovsky, Johnstone, Godar, et al. (2006)	N=13	+ or - 60°	9/13	9/13
		+ or - 40°	6/13	9/13
		+ or - 20°	2/13	8/13
Grieco-Calub, Litovsky & Werner (2008)	N=10 BICI N=8 CI	+ or - 60°	0/8	5/10
		+ or - 40°	0/8	5/10
		+ or - 20°	0/8	3/10
Litovsky, Johnstone & Godar (2006)	N=6	+ or - 60°	5/6	6/6
		+ or - 40°	3/6	6/6
		+ or - 20°	1/6	4/6
Senn, Kompis, Vischer et al. (2005)	N=2	+ or - 60°	2/2	2/2
		+ or - 40°	2/2	2/2
		+ or - 20°	2/2	2/2
Peters, Litovsky, Lake & Parkinson (2004)	N=6	+ or - 60°	0/6	0/6
		+ or - 40°	0/6	0/6
		+ or - 20°	0/6	0/6
Total	N=31	+ or - 60°	16/35	22/37
		+ or - 40°	11/35	22/37
		+ or - 20°	3/35	15/37

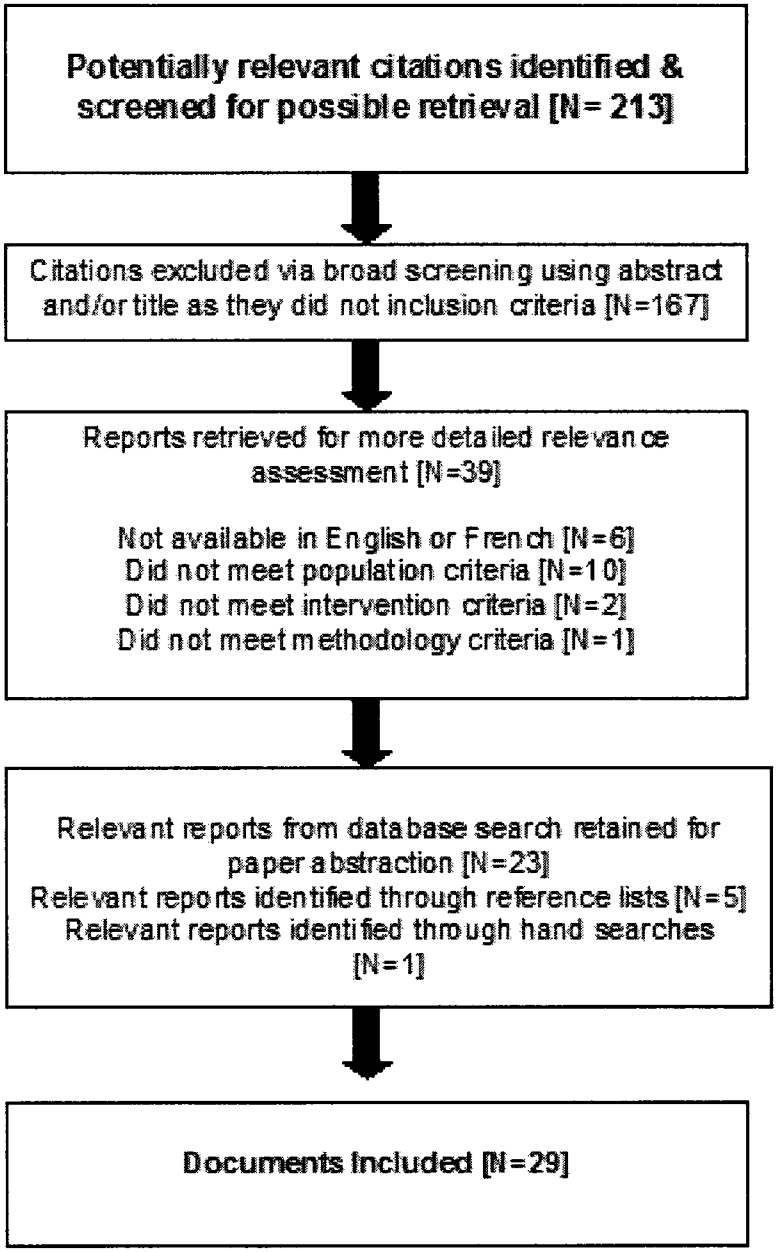
N: number of participating subjects

Table 4: Speech recognition in noise performance as measured by the Adaptive Spondee Test (AdSpon) at various signal-to-noise ratios for children using a single cochlear implant (CI) and bilateral cochlear implants (BICI)

Study	BICI Subjects	Signal to Noise Ratio	Number of Children performing task at this threshold with CI	Number of Children performing task at this threshold BICI
Galvin, Mok & Dowell (2007)	N=10	Normal Hearing Level = -11.5	0/10	1/10
		Sensorineural HL Level = -5.4	5/10	8/10
Galvin, Mok, Dowell & Briggs (2007)	N=6	Normal Hearing Level = -11.5	0/6	1/6
		Sensorineural HL Level = -5.4	5/6	4/6
Total	N=16	Normal Hearing Level = -11.5	0/16	2/16

N: number of participating subjects

Figure 1: Progress through the review



N: number of articles

CHAPTER 4

Estimation of risks associated with paediatric cochlear implantation

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Estimation of risks associated with paediatric cochlear implantation

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Abbreviations: BICI: bilateral cochlear implant, CI: cochlear implant, dB HL: deciBel at Hearing Level, USFDA: United States Food and Drug Administration

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Abstract

Objectives: To estimate the rates of complications associated with paediatric cochlear implantation use: a) at one Canadian cochlear implant (CI) centre, b) in the published literature.

Design: A retrospective hospital-based chart review and a concurrent review of complications in the published literature

Participants: 224 children who had undergone surgery from 1994 to June 2007.

Results: Results of this study indicated that the rates of complications at the local Canadian paediatric CI centre are not significantly different from the literature rates for all examined complication types.

Conclusion: This hospital-based retrospective chart review and review of the literature provides readers with an estimation of the risks to aid in evidence-based decision making surrounding paediatric cochlear implantation.

Strong research evidence has consistently indicated that unilateral cochlear implants (CIs) improve hearing sensitivity levels, speech perception, language and literacy in children (Thoutenhoofd et al., 2005; Moog et al., 2003). Due to the positive research and clinical results associated with implantation, the criteria for CI use in children that were established in 1990 have been expanded (Candidacy Criteria, 2008; United States' Federal Drug Administration [USFDA], 1990). Originally implantation was considered only in children with profound hearing loss, now children with lesser degrees of hearing loss are eligible for CIs. In addition, children may be implanted with CIs at one year of age or earlier (particularly for some cases of meningitis) (Thoutenhoofd et al., 2005). The expanded criteria and positive outcomes associated with CIs have made it the standard of care for children with severe to profound hearing loss (Berg et al., 2007).

Recently, bilateral cochlear implantation has become more commonly used with children (Berg et al., 2007). Four reviews have concluded that there are benefits for patients receiving bilateral stimulation compared to monaural stimulation with a single CI as measured by speech recognition in noise and sound localization (Brown et al., 2007; Ching et al., 2007; Murphy et al., 2007; Schafer et al., 2006). For children who cannot use hearing aids in their second ear due to severity of hearing loss, bilateral stimulation can only occur through the use of a second CI. As the majority of children implanted to date have received one CI, this change in clinical practice toward bilateral implantation is prompting parents to consider an additional CI for their children.

While CIs can provide significant benefits for many children, there is also the potential for complications or adverse events with implant surgery and/or device use. There are both minor and major risks associated with CI surgery (Cohen et al., 1991). Major surgical complications are defined as those that require additional surgery or medical

management in a hospital setting. Examples include facial nerve paralysis, or flap infection requiring hospitalization. Minor complications are defined as those that are treated with medical management on an outpatient basis. These complications can include flap infections requiring oral antibiotics and haematoma.

The possible adverse effects due to implantation use can include tinnitus or imbalance. Increased post-surgical risk of bacterial meningitis among children who have received an implant has been highly publicized and is considered a major risk of the device (Health Canada, 2003; USFDA, 2006). Recent work has identified that the increased risk of meningitis has been linked, in part, to a device with a positioner (a small piece of silicon inserted separate from the electrode array of the CI) that has since been removed from the market (Biernath et al., 2006; USFDA, 2006). Vaccinations to prevent pneumococcal meningitis continue to be recommended for the entire CI recipient population due to the increased risk.

Postauricular edema and/or erythema, clinically presenting as mastoiditis, otitis media presenting as may also occur in some patients with otitis media due to the removal of the mastoid during CI surgery. The predisposition for this condition is a consequence of the surgical access necessary for insertion of the cochlear implant. This adverse event can be major, requiring intravenous antibiotics and hospitalization, or minor requiring oral antibiotics on an outpatient basis.

With less than 20 years of clinical experience with multichannel cochlear implants, long-term device reliability is unknown. It is not realistic to expect that a 12 month old child will be implanted with a device that will function optimally for their entire lifetime. However, some children will experience premature device failure. Device failure can occur due to an internal defect in the device or due to trauma to the device from an impact to the

head (Battmer et al., 2007). In both cases, the device malfunction requires surgery to explant the defective device and reimplant a functional device.

Current estimates of complications of CI surgery available in the published literature are derived primarily from hospital-based retrospective studies (Arnoldner et al., 2005; Bhatia et al., 2004; Black et al. 2007; Cunningham et al., 2004; Gysin et al., 2000; James et al., 2004; Kandogan et al., 2005; Maurer et al., 2005; Migrov et al., 2006a; Migirov et al., 2006b; Parisier et al., 2001; Postelmans et al., 2006; Weise et al., 2005; Stratigouleas et al., 2006; Venail et al., 2007; Yu et al., 2001). Some studies examine the complications of the entire CI population while others explore complications among specific sub-groups such as infants or otitis-prone children. The available studies report sample sizes ranging from 25 participants (James et al., 2004) to as many as 705 (Fayad et al., 2003). There is also considerable variation in the complication rates reported in these studies. For example, some studies report no minor complications in their results (James et al., 2004) while others report rates as high as 18.75% (Postelmans et al., 2006).

Accurate information on the risks and benefits of paediatric cochlear implantation is important for parents, clinicians, and surgeons. When parents make the decision to undergo unilateral or bilateral cochlear implantation for their child, it is essential that "comprehensive and bias-free" information is provided before and following surgery (Sorkin et al., 2008). Providing parents and clinicians with accurate complication rates associated with paediatric cochlear implantation is essential in the CI decision-making and follow-up stages.

Purpose and Objectives

The primary purpose of this project was to ascertain paediatric CI complication rates for use by parents, surgeons, and clinicians. There are two objectives associated with this

project: 1) to estimate the rates of complications associated with paediatric cochlear implantation use at one Canadian CI centre; 2) to estimate the rates of complications of paediatric cochlear implantation in the published literature.

Chart Review

Methods

A retrospective hospital-based chart review was conducted to examine the CI complication rates within one regional paediatric hospital setting that serves a population between 0 and <19 years of age. The medical records of children who had undergone unilateral, bilateral, and reimplantation CI surgery between 1994 and June 2007 were examined for intra- and post-operative surgical and medical complications. Demographic (age at implantation, gender, aetiology), audiological (pure tone average at time of implantation), and medical complication data were extracted from medical records. The complications of interest included, but were not limited to: flap infections, clinical mastoiditis, facial palsy, bacterial meningitis, and device failure. Pre- and post-operative assessment reports were examined for complications. This included operative and discharge reports as well as follow-up audiological and surgical notes. Post-surgical visits to the paediatric hospital emergency room were also examined for relevancy to the CI surgery. Once a complication was identified, it was defined as either major or minor based on the classification by Cohen et al. (1991).

Analysis

Descriptive statistics including frequencies, means, and ranges were used to describe the demographic characteristics of children who had received CIs between 1994 and June

2007 at a single Canadian CI centre. Crude rates or overall rates were calculated for negative medical and surgical outcomes among the population of children who had received CIs. Crude rates are determined by dividing the total number of events of interest by the defined overall population. The crude rate was calculated for minor and major complications and separately for facial nerve palsy. As these types of complications typically occur during the surgery or recovery period the time frame for these calculations was the first year post-CI. Therefore each surgery is included in the denominator for these rate calculations. The crude rate was an appropriate measure as it gives an overall summary of the actual CI surgical experience (Oleckno, 2002).

Complication rates for bacterial meningitis and device failure were calculated slightly differently as these potential risks can occur shortly following the surgery as well as in the years of continued use. For these risks, cumulative incidence rates were calculated. Cumulative incidence rates report the risk of a complication in a defined population, over a specified time frame (Oleckno, 2002). The rates for bacterial meningitis have typically been calculated as the number of negative events by the number of person-years (PY) (Reefhuis et al., 2003; Summerfield et al., 2004). The total number of years during which all children in the sample had worn their devices was used as the denominator to calculate the number of bacterial meningitis events per 1000 PY. Simlarly, 5 year cumulative incidence rates for device failure were calculated for the sample as the number of device failures by the number in the sample who wore the device for at least 5 years. An overall failure rate was also calculated (number of negative events by total implants) as many of the available articles report such rates. Ninety-five percent (95%) confidence intervals were calculated for the local database rates. A binomial distribution was used for the calculation of the confidence

interval of the facial paralysis and facial palsy rates and a poisson distribution was used for the calculation of all other confidence intervals (Last, 2001).

Results

Included surgeries were all those completed between 1994 and June 2007. All surgeries were conducted in the same hospital and by the same surgeon. During this timeframe there were 229 children who underwent 242 surgeries. Eight of the additional surgeries were re-implantations on the same side due to device failure and 5 were to implant a bilateral device. The mean age of implantation during this period was 5.46 years (SD = 4.5, range = 9 months to 19 years). More boys underwent CI surgery (57.6% of surgeries) compared to girls (42.5%). The mean pure tone average of CI recipients in their better (non-implanted) ear was 103 dB HL (SD= 12.9, range = 51.7 to 120 dB HL). These data include children with hearing losses due to auditory neuropathy. Aetiology of the children's hearing losses is presented in Figure 1.

Chart reviews to examine medical complications were possible for 224 children (236 surgeries) of the 229 children (242 surgeries) who had undergone surgery during this timeframe. Therefore, data were available for 97.8% of children and 97.5% of surgeries. Two percent (2.2%) of the charts were not available on the multiple dates on which the review was conducted.

Approximately 38% of the implanted children came from outside of the immediate area and 18% of the children received services from the hospital for only the surgical and early post-surgical period. They received post-surgical services within their home community. The remaining children received surgical, post-surgical, and some rehabilitative support from the hospital though infrequently.

Table 1 presents the results of the local retrospective chart review. It includes the different types of complications (minor complications, major complications, facial nerve palsy, bacterial meningitis, and device failure), the number of children included in the rate calculation, the number of negative events, and the rate of complications for this sample of children.

The most frequently occurring adverse events were mastoiditis (*N*=9, 3.8% of surgeries), wound infections (*N*=8, 3.4% of surgeries), and disequilibrium (*N*=5, 2.1% of surgeries). Minor complications experienced in this setting included auricular and flap infections treated with oral antibiotics (*N*=6, 2.5% of surgeries) and mild dizziness or disequilibrium (*N*=5, 2.1% of surgeries) following surgery. The most commonly occurring adverse event was mastoiditis that was treated in hospital with intravenous (IV) antibiotics (*N*=9, 3.8% of surgeries). Less frequently occurring major complications were localized infection requiring surgery or IV antibiotics (*N*=2, 0.9% of surgeries), and cellulitis (*N*=1, 0.4% of surgeries). Seven children experienced eight device failures. All complications occurred during the first year of device use except for three of the eight cases of mastoiditis which occurred 2 to 3 years post-surgery. The eight device failures occurred within 1.5 to 5 years post-implantation.

Review of Literature

Methods

A review of complications in the published literature was conducted concurrently with the chart review. Multiple databases (Medline, CINAHL, PsycInfo) were searched for articles on medical and surgical complication rates associated with cochlear implantation. Inclusion criteria for the review included articles published after 2000. This date was chosen

with the hope that more recent studies would include surgical procedures and numbers of surgeries most similar to current procedures and numbers. In addition the published articles must have included some children in their sample. Retrospective and prospective studies were included along with larger national or regional database analyses. Case studies were not included. Once an article was identified as relevant, the reference list was examined for additional relevant publications. Included articles were examined for complication rates. The complications of interest were the same as those used in the chart review. The same classification system for the complications was applied (Cohen et al., 1991).

Of the identified articles, two were from the same sample of children but from two different time periods (Migrov et al., 2006a; Migirov et al., 2006b). The article with the longer time frame and larger sample size was included in this analysis (Migrov et al., 2006a).

Analysis

Extracted information from each identified article included the type of complication, the number of complications and the overall sample size. Time frame of the study, characteristics of the sample (age, population), and study design were all extracted from each study. In order to calculate a crude rate based on the available literature, the number of complications in each category from the included studies was divided by the total sample size of all included studies combined. Studies that calculated cumulative incidence rates for meningitis and device failure were included in the review in order to arrive at a measure of the risk of these complications over a specified time period. Ninety-five percent (95%) confidence intervals were calculated for the rates calculated from the literature. A binomial distribution was used for the calculation of facial palsy rates and a poisson distribution was used for the calculation of all other confidence intervals (Last, 2001).

Results

The initial search for articles on complications of paediatric cochlear implantation identified 116 articles. Figure 2 provides a diagram of progress though the review. Appendix C provides a summary of the included studies and their characteristics by complication type. A number of studies were excluded due to the lack of information on the total sample size for which complications were reported. Other studies were excluded as they reported audiological or psycho-educational outcomes and not medical or surgical outcomes. Many of the studies did not include children or examined only a subset of the population (i.e., otitis prone children). Following the application of the inclusion criteria to the database search, the hand searching, and examination of references, 19 articles were identified for inclusion.

Since only studies published in 2000 or later were examined in this review, the majority of studies (13/19) included children implanted in the 1990's and 2000's and not prior to this (Arnoldner et al., 2005; Black et al. 2007; Cunningham et al., 2004; Gysin et al., 2000; James et al., 2004; Kandogan et al., 2005; Maurer et al., 2005; Migrov et al., 2006a; Parisier et al., 2001; Postelmans et al., 2006; Reefhuis et al., 2003; Summerfield et al., 2004; Weise et al., 2005;Yu et al., 2001). Three studies did not report the timeframe in which the surgeries were completed (Battmer et al., 2007; Bhatia et al., 2004; Stratigouleas et al., 2006). All except four studies involved individual hospital site. Four studies examined the complication rates at multiple CI centres (Battmer et al., 2007; Fayad et al., 2003; Reefhuis et al., 2003; Summerfield et al., 2004). Three of the studies examined and reported complications for both adults and children together (Stratgouleas et al., 2006; Fayad et al., 2003; Yu et al., 2001). All of the other studies separated their results or examined only the outcomes of a paediatric population.

Table 2 provides a summary of the complication types, their definitions, the studies identified for each topic, the combined sample size of all included studies, and the combined complication rate of the identified studies for each type of complication. It also provides the 95% confidence intervals for the local retrospective chart review rates and the calculated complication rates from the literature. As the confidence intervals from the retrospective chart review and the literature review for each complication type overlap, they were not significantly different from each other. Due to the larger sample sizes of the combined literature review, the confidence intervals are narrower for these results.

Some studies classified minor complications as those that could be treated medically and major complications as requiring additional surgery or device extrusion (Postelmans et al., 2007). Interestingly, the Postelmans et al. study classified an electrode malfunction, which required additional surgery, as a minor complication. The current study used the definitions by Cohen et al. (1991) which reclassified this complication as major for comparison purposes with other studies. Another study (Migirov et al., 2006a) included all mastoiditis cases (with or without surgical intervention) as a minor complication. Again, these adverse events were reclassified for comparison purposes in the current study.

The risks of bacterial meningitis and facial nerve palsy were the smallest risks associated with paediatric cochlear implantation in the published literature, as they were in the retrospective chart review. The most frequently occurring complications in the literature were wound infections and abscesses (2.7% of surgeries), disequilibrium (1.9% of surgeries), cerebral spinal fluid gushers, and mastoiditis (both occurring in 0.8% of surgeries). These results are similar to the most frequently occurring complications in the local study: mastoiditis, wound infections, and disequilibrium.

Discussion

There are some risks of adverse events associated with cochlear implantation surgery. Results of this study indicated that the rates of complications at the local Canadian paediatric centre examined in this paper are not significantly different from the rates from the literature for all examined complication types.

Major and Minor Complications

Some variability was noted in the classification of complications in the literature. The variability in classifications across the literature makes comparisons challenging for parents and clinicians seeking consistent reporting of complication rates. A consistent approach to reporting and classifying cochlear implantation complications would be helpful.

Interestingly, mastoiditis in the local Canadian centre examined in the retrospective chart review occurred after the first year of device use in 3 of the 8 cases. Parents should be informed about the potential for this risk to continue beyond the period of initial surgery. It is important for children with a history of chronic otitis media to be monitored and treated promptly as chronic otitis media may be associated with increased infectious complications, primarily meningitis (Cunningham et al., 2004).

Temporary and Permanent Facial Nerve Paralysis or Palsy

The risk of permanent facial palsy was the lowest risk of all complication types. Unlike the definitions of minor and major complications, the included studies were consistent in their definitions of facial nerve palsy. The majority of studies that reported cases of temporary facial nerve palsy indicated that these cases resolved after minimal or no intervention (Arnoldner et al., 2005; Bhatia et al., 2004; Black et al., 2007; Fayad et al.,

2003; Gysin et al., 2000; Kandogan et al, 2005; Migrov et al., 2006a). The resolution of these temporary cases is important for parents and clinicians to note. Only two studies reported permanent facial palsy in three children (Stratigouleas et al., 2006; Venail et al., 2007). While there was some variation in the literature in the rates of facial nerve palsy (0 to 2.27/100 for temporary palsy, 0 to 0.74/100 for permanent palsy), these rates were not statistically different from study to study.

Meningitis

The risk of bacterial meningitis for CI users is the complication that has received the most attention from federal health authorities due to the higher occurrence in the CI recipient population compared to the general public (Health Canada, 2003; USFDA, 2006). The risk of bacterial meningitis for CI users in the literature was approximately 1.92 per 1000 PY, whereas the risk of meningitis in the general population is 0.0131/1000 PY (Summerfield et al., 2005). This was a statistically significant difference ($Z>36.5$, $p<0.0001$). This increased rate in CI users has been associated with two risk factors, 1) the use of a CI positioner, and 2) a cochlear malformation (Reefhuis et al., 2003). Devices with positioners have been removed from the market however it has not currently been recommended that these devices be removed. As well, it is still recommended that all CI recipients undertake preventative measures, such as vaccination, to avoid bacterial meningitis (USFDA, 2007). A recent survey of US CI recipients' parents and an examination of patient charts indicated that compliance with vaccinations is not always universal. Twenty-nine (29%) of parents of children under 2 years of age were unaware of their child's vaccination status and for children over 2, vaccination status was absent from the child's chart in 43% of cases. Chart reviews revealed that 12% of children were not vaccinated according to recommended

guidelines (USFDA, 2007). Parents and clinicians need to work together to try and reduce this risk for children through vaccination and seeking prompt treatment for otologic infections.

Device Failure

Many of the included studies reported the device failure rate as the number of defective devices (either due to trauma or a malfunction of the device) by the total number of surgeries without accounting for the length of device use (Arnolder et al., 2005; Battmer et al., 2007; Gysin et al., 2000; Kandogan et al., 2005; Parisier et al., 2001; Postelmans et al., 2006; Stratigouleas et al., 2006; Weise et al., 2005). As device failures increase with the length of device use, rates that include both children with recent implants and those with longer use in the denominator tend to underestimate the rate of device failure. Two studies reported cumulative 5 year survival rates which presents a more accurate indication of the device failures over a 5 year time frame as it includes only children with 5 years of CI experience in the denominator (Maurer et al., 2005; Venail et al., 2007).

Manufacturers report device failure rates over various time frames. The variation in rates depends on time frame, type and age of device, as well as types of failures included in the rates. There is considerable variation in the reported rates among manufacturers. Some of the manufacturers include device failures due to medical, surgical, and accident related issues whereas others only report failures related to the integrity of the device exclusive of external factors. The cumulative 5 year device failure rates range from 1% to 10.9% (Advanced Bionics, 2006; Cochlear Corp., 2008; Med El, 2008). It is essential that clinicians and parents are adequately informed about the challenges in interpreting manufacturers' rates due to this variability.

Informed Decision-Making

Research indicated that the risks associated with unilateral cochlear implantation are acceptable to most families due to the significant benefits for children in terms of speech perception, language, and communication outcomes (Johnston et al., 2008). It is still essential, however, that parents have access to clear information on the risks of cochlear implantation as has been presented here.

For those families making the decision regarding bilateral cochlear implantation, these complication rates will also be of interest. Parents will be able to consider the risks and compare them to the benefits associated with bilateral cochlear implantation, which are different from the benefits associated with unilateral implantation. These benefits include improved sound localization and speech recognition in noise (Brown et al., 2007; Ching et al., 2007; Murphy et al., 2007; Schafer et al., 2006). Some evidence exists to indicate that parents perceive the decision to undergo bilateral cochlear implantation differently than the decision to undergo unilateral implantation (Johnston et al., 2008). These differences in perceptions of the two procedures may be due to differential perceptions of the ratio of risks to benefits, or to a lack of information regarding the risks of this procedure. In either case, the current study may provide parents with additional informational support for their bilateral decision-making.

Limitations

A few charts of completed surgeries (N=5 or 2%) were not available for review during the study time-frame. However, this small percentage of surgeries does not represent a strong threat to the validity of the chart review. A number of the children that were implanted at this site resided out of the local area. While most of these children were seen

periodically at the follow-up clinic and would have had any complications documented in the chart, 18% of the children were only seen for surgery and the early post-surgical period. This loss to follow-up may have resulted in a lower estimation of later occurring complication rates. Latent complications could have occurred in all categories, however, any device failures would have been reported and documented in the charts of these patients.

There are limitations to the study due to the small sample sizes and the rare occurrence of some of the complications. The inclusion of 95% confidence intervals provides readers with an indication of the possible variation of rates due to the sample size. The small sample size of the retrospective chart review, particularly for estimating the rare facial palsy and meningitis events, is reflected in the wide confidence intervals for these complications.

Estimations of complication rates from the published literature are dependent upon the quality of the charting and reviewing at numerous CI centres and therefore subject to considerable variability. There is likely also some variability in the classification and reporting of major and minor complications across centres. This was addressed by re-classifying the complications using the same definitions when there was adequate description in the articles (Cohen et al., 1991).

There may also be variation in the treatment approaches from centre to centre that would determine whether a complication is minor or major. For example, a localized infection that is treated in hospital with IV antibiotics is considered a major complication whereas a localized infection treated with oral antibiotics at home is considered a minor complication. Centres may vary in their approaches to treating localized infections. This variation may result in differing rates of minor and major complications across centres for adverse events. Another example may be the reporting of tinnitus or disequilibrium. While some centres may consider some occurrence of these side effects as an expected occurrence

of opening the cochlear during CI surgery, other centres might report these events as a minor complication. Without an examination of the medical practices at different settings it is difficult to determine if this could occur.

By limiting the inclusion criteria to data from the published literature there is a potential for results to be affected by a publication bias from centres that feel they have complication rates that are exemplary. Future work could examine the rates from centres who have not published their rates to determine if there are differences between published and unpublished rates.

Finally, this study focused on identifying the medical and surgical risks of paediatric cochlear implantation. It was beyond the scope of this research to examine if there are any psycho-social risks associated with the procedure. Additional research may provide insight into the potential for cochlear implants to impact children's early or later psycho-social development.

Conclusions

This hospital-based retrospective chart review and review of the literature provides readers with an estimation of the risks associated with paediatric cochlear implantation. These complication rates, together with information on the benefits of CI use, can be used to help parents and clinicians make evidence-based decisions surrounding cochlear implantation.

References

Advanced Bionics (2006). Advanced Bionics Reliability Report. http://www.bionicear.com/printables/reliabilityreport2006.pdf [January 19, 2009].

Arnoldner C, Baumgartner WD, Gstoettner W, Hamzavi J (2005). Surgical considerations in cochlear implantation in children and adults: a review of 342 cases in Vienna. Acta Oto-Laryngologica 125: 228-234.

Battmer RD, O'Donoghue GM, Lenarz T (2007). A multicenter study of device failure in European cochlear implant centers. Ear and Hearing 28: 95S-99S.

Berg AL, Ip SC, Hurst M, Herb A (2007). Cochlear implants in young children: informed consent as a process and current practices. American Journal of Audiology 16: 13-28.

Bhatia K, Gibbin KP, Nikolopoulos TP, O'Donoghue GM (2004). Surgical complications and their management in a series of 300 consecutive pediatric cochlear implantations. Otology and Neurotology 25: 730–739.

Biernath KA, Reefhuis J, Whitney CG, Mann EA, Costa P, Eichwald J, et al. (2006). Bacterial Meningitis among children with cochlear implants beyond 24 months after implantation, Pediatrics 117: 284-289.

Black IM, Bailey CM, Albert DM, Leighton SEJ, Hartley BEJ, Chatrath P, Patel N (2007). The Great Ormond Street Hospital paediatric cochlear implant programme 1992–2004: A review of surgical complications. Cochlear Implants International 8: 53–67.

Brown KD, Balkany TJ (2007). Benefits of bilateral cochlear implantation: a review, Current Opinions in Otolaryngology, Head and Neck Surgery 15: 315-318.

Candidacy Criteria. (2004). http://www.cochlear.com/Support/631.asp [February 9, 2008].

Ching TY, van Wanrooy E, Dillon H (2007). Binaural-bimodal fitting or bilateral implantation for managing severe to profound deafness: a review. Trends in Amplification 11; 161-192.

Cochlear Corporation (2009). Hear now and always, our commitment to you. http://www.cochlearamericas.com/products/1789.asp [January 19, 2009].

Cohen NL, Hoffman RA (1991). Complications of cochlear implant surgery in adults and children. Annals of Otorhinolaryngology 100: 708-711.

Cunningham CD, Slattery WH, Luxford WM (2004). Postoperative infection in cochlear implant patients. Otolaryngology - Head & Neck Surgery 131: 109-114.

Gysin C, Papsin BC, Daya H, Nedzelski J (2000). Surgical outcome after paediatric cochlear implantation: Diminuation of complications with the evolution of new surgical techniques. The Journal of Otolaryngology 29: 285-289.

Health Canada (2003). Cochlear implant recipients may be at greater risk for meningitis-Information update. http://www.hc-sc.gc.ca/dhp-mps/medeff/advisories-avis/prof/_2003/implant_cochle_2_nth-ah-eng.php, [October 17, 2008].

Fayad JN, Wanna GB, Micheletto JN, Parisier SC (2003). Facial nerve paralysis following cochlear implant surgery. Laryngoscope 113:1344–1346.

James AL, Papsin BC (2004). Cochlear implant surgery at 12 months of age or younger. Laryngoscope 114: 2191-2195.

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. Canadian Journal of Speech-Language Pathology and Audiology, 32,169-82.

Kandogan T, Levent O, Gurol G (2005). Complications of paediatric cochlear implantation: experience in Izmir. Journal of Laryngology & Otology 119: 606-610.

Last JM (2001). A dictionary of epidemiology 4th Ed. New York, NY: Oxford University Press.

Maurer J, Marangos N, Ziegler E (2005). Reliability of cochlear implants. Otolaryngology, Head and Neck Surgery 132: 746–750.

Med El (2008). Cochlear Implant Reliability Data. http://www.medel.com.ar/ENG/US/20_Products/10_Cochlear_Implants/999_reliability.asp [October 28th, 2008].

Migirov L, Muchnik C, Kaplan-Neeman R, Kronenberg J (2006a). Surgical and medical complications in paediatric cochlear implantation: A review of 300 cases. Cochlear Implants International 7: 194-201.

Migirov L, Yakirevitch A, Kronenberg J (2006b). Surgical and medical complications following cochlear implantation: comparison of two surgical approaches. ORL 68: 213-219.

Moog JS, Geers AE (2003). Epilogue: Major findings, conclusions and implications for deaf education. Ear Hear 24: 121S-125S.

Murphy J, O'Donoghue G (2007). Bilateral cochlear implantation: an evidence-based medicine evaluation, Laryngoscope 117: 1412-1418.

Oleckno WA (2002). Essential Epidemiology. Prospect Heights, IL: Waveland Press Inc.

Parisier SC, Chute PM, Popp AL, Suh GD (2001). Outcome analysis of cochlear implant reimplantation in children. Laryngoscope 111: 26-32.

Postelmans JTF, Cleffken B, Stokroos RJ (2007). Post-operative complications of cochlear implantation in adults and children: Five years' experience in Maastricht. The Journal of Laryngology and Otology 121: 318-323.

Reefhuis J, Honein MA, Whitney CG, Chamany S, Mann EA, Biernath KR, Broder K (2003). Risk of bacterial meningitis in children with cochlear implants. The New England Journal of Medicine 349: 433-443.

Schafer EC, Thibodeau LM (2006). Speech Recognition in noise in children with cochlear implants while listening in bilateral, bimodal, and FM-system arrangements. *American Journal of Audiology* 15: 114-126.

Sorkin DL, Zwolan TA (2008). Parental perspectives regarding early intervention and its role in cochlear implantation in children. Otology and Neurootology 29: 137-141.

Stratigouleas ED, Perry BP, King SM, Syms CA (2006). Complication rate of minimally invasive cochlear implantation. Otolaryngology - Head & Neck Surgery 135: 383-386.

Summerfield AQ, Cirstea SE, Roberts KL, Barton GR, Graham JM, O'Donoghue GM (2004). Incidence of meningitis and of death from all causes among users of cochlear implants in the United Kingdom. Journal of Public Health 27: 55-61.

Thoutenhoofd, E., Archbold, A., Gregory, S., Lutman, M., Nikolopoulos, T. et al. (2005). Paediatric Cochlear implantation: Evaluating Outcomes. London: Whurr.

United States Food and Drug Administration (1990). Devices at FDA: Nuleus 22 Channel Cochlear Implant System in Children. http://www.accessdata.fda.gov/scripts/cdrh/devicesatfda/index.cfm?db=PMA&id=7987 [August 6, 2008].

United States Food and Drug Administration (2006) FDA Public Health Notification: Continued Risk of Bacterial Meningitis in Children with Cochlear Implants with a Positioner Beyond Twenty-Four Months Post-Implantation. http://www.fda.gov/cdrh/safety/020606-cochlear.html [January 19, 2009].

United States Food and Drug Administration (2007) FDA Public Health Notification: Importance of Vaccination in Cochlear Implant Recipients http://www.fda.gov/cdrh/safety/101007-cochlear.html [January 19, 2009]

Venail F, Sicard M, Piron JP, Artiered F, Uziel A, Mondain M (2007). Cochlear implantation safety and reliability in paediatric cases. Audiological Medicine 5: 210-217.

Weise JB, Muller-Deile J, Brademann G, Meyer JE, Ambrosch P, Maune S (2005). Impact to the head increases cochlear implant reimplantation rate in children. Auris Nasus Larynx 32: 339-343.

Yu KCY, Hegarty JL, Gantz BJ, Lalwani AK (2001). Conservative management of infections in cochlear implant recipients. Otolaryngology-Head and Neck Surgery 125: 66-70.

Figure 1: Number of children with cochlear implants at one cochlear implant centre with various aetiologies of hearing loss of children (*N*=229).

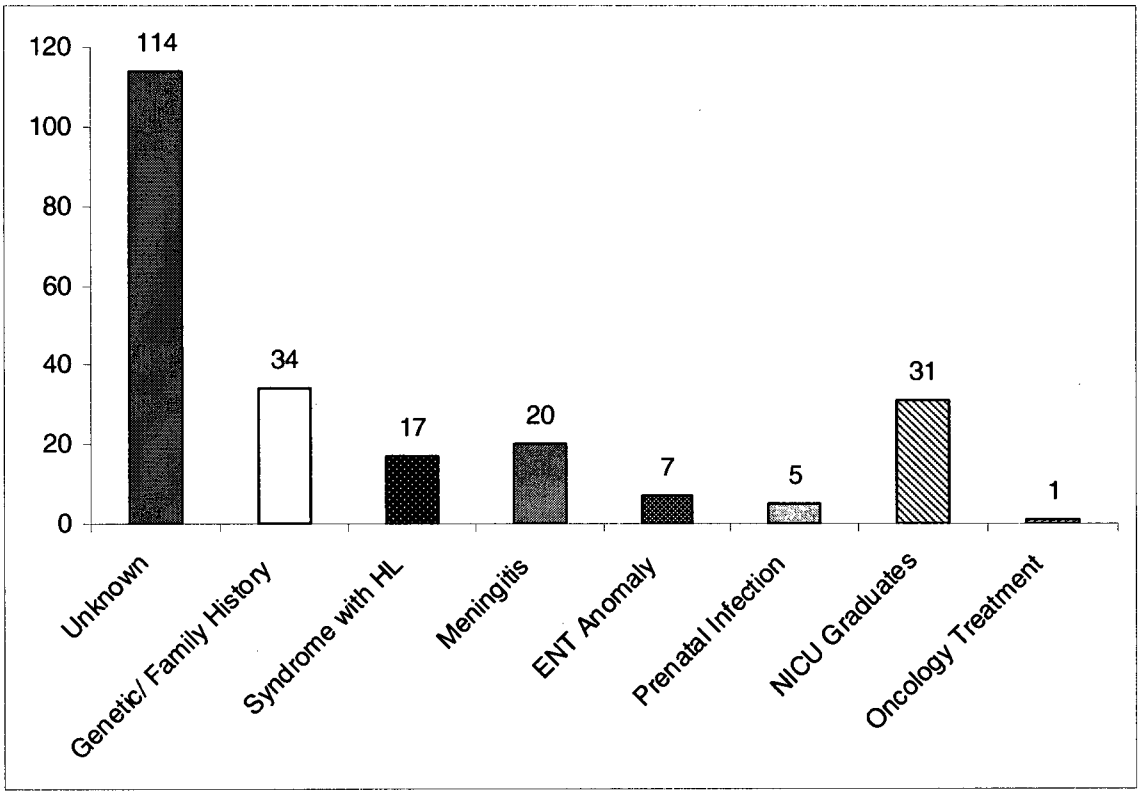

Table 1: Complication rates from local retrospective chart review.

Complication	Definition	Sample Size	Description of Events	Crude Complication Event Rate	Standardized Complication Event Rate
Minor Complications	Managed conservatively on an ambulatory basis	N= 236	Flap or auricular infection treated with oral antibiotics (*N*=6) Dizziness / disequilibrium (*N*=5)	11/236	4.66/100
Major Complications	Required hospitalization, explantation, or IV antibiotics and doesn't fall into other category	N= 236	Mastoiditis (*N*=9) localized infection requiring surgery or IV antibiotics (*N*=2) cellulitis (*N*=1)	12/236	5.08/100
Temporary and Permanent Facial Palsy	Facial nerve weakness or paralysis occurring following CI surgery	Temporary		Temporary	Temporary
		N= 236		0/236	0/1000
		Permanent		Permanent	Permanent
		N= 236		0/236	0/1000
Bacterial Meningitis	Bacterial Meningitis	N= 1286 PY	One child suffered from Meningitis twice	2/1286 PY	1.55/1000 PY
Device Failure	Hard failures of cochlear implants indicated by abnormal integrity test and/or trauma to the CI site	Overall		Overall Crude	Overall Standardized
		N= 236		8/236	3.39/100
		Cumulative 5 year Failure rate		Crude Cumulative 5 year Failure rate	Standardized Cumulative 5 year Failure rate
		N=146		6/146	4.1/100

Figure 2: Progress through the review of the literature

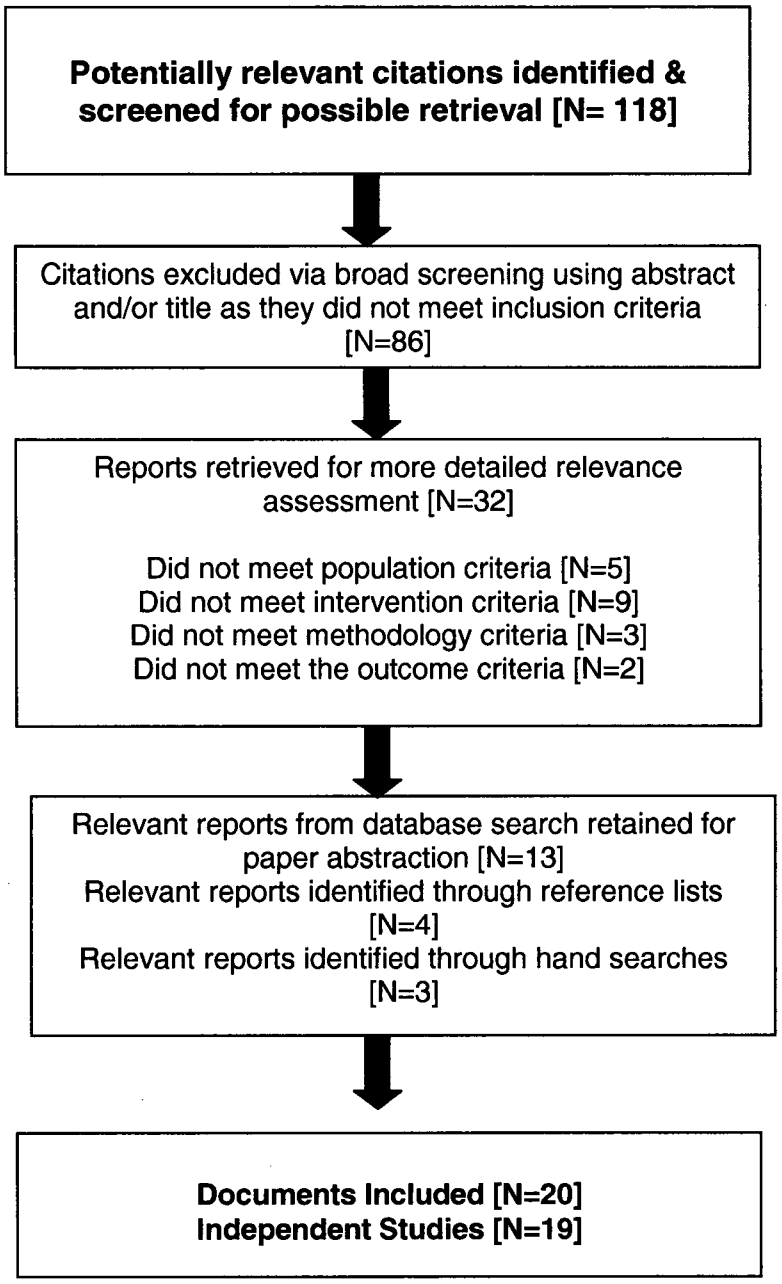

Table 2: Summary of complication rates from the identified literature and comparison with local retrospective chart review.

Complication	Definition	Included Studies		Sample Size of Literature	Crude and **Standardized Rate** from Literature (95% CI)	**Standardized Rate** from Retrospective Chart Review (95% CI)
Minor Complications	Managed conservatively on an ambulatory basis	Stratigouleas, et al. (2006) Cunningham, et al. (2004) Migrov, Muchnik, et al. (2006) Gysin, et al. (2000) Arnoldner, et al. (2005) James & Papsin (2004)	Postelmans. et al. (2006) Venail, et al. (2007) Black, et al. (2007) Kandogan, et al. (2005) Bhatia, et al. (2004)	N=2171	189/2053 **9.21/100** (7.94 – 10.62/100)	**4.66/100** (2.32 - 8.34/100)
Major Complications	Required hospitalization, explantation, or IV antibiotics and not in other category	Stratigouleas, et al. (2006) Cunningham, et al. (2004) Migrov, Muchnik, et al. (2006) Gysin, et al. (2000) James & Papsin (2004)	Bhatia, et al. (2004) Venail, et al. (2007) Yu, et al. (2001) Kandogan, et al. (2005) Black, et al. (2007)	N=2161	73/2161 **3.38/100** (2.64 – 4.25/100)	**5.08/100** (2.62 – 8.88/100)
Temporary and Permanent Facial Palsy	Facial nerve weakness or paralysis occurring following CI surgery	Fayad, et al. (2003) Venail, et al. (2007) Stratigouleas, et al. (2006) Migrov, Muchnik, et al. (2006)	Arnoldner, et al. (2005) Black, et al. (2007) Kandogan, et al. (2005) Bhatia, et al. (2004) Gysin, et al. (2000)	Temporary		
				N=2450	17/2450 **6.94/1000** (4.04 - 11.09/1000)	**0/1000** (0.11 – 23.4/1000)
				Permanent		
				N=2450	3/2450 **1.22/1000** (0.25 - 3.57/1000)	**0/1000** (0.11 – 23.4/1000)
Bacterial Meningitis	Bacterial Meningitis	Summerfield, et al. (2004) Reefhuis, et al. (2003)		N= 13441 PY	26/13441 PY **1.93/1000 PY** (1.26 – 2.8/1000 PY)	**1.55/1000 PY** (0.188-5.6/1000PY)
Device Failure	Hard failures of the device evidenced by abnormal integrity test and/or trauma to CI site	Overall Complication Rate				
		Weise, et al. (2005) Stratigouleas, et al. (2006) Kandogan, et al. (2005) Postelmans, et al. (2006)	Arnoldner, et al. (2005) Parisier.et al. (2001) Gysin, et al. (2000) Battmer, et al. (2007)	N= 14032	550/14032 **3.92/100** (3.60 – 4.26/100)	**3.39/100** (1.46 – 6.68/100)
		Cumulative 5-year failure rate				
		Venail, et al. (2007) Maurer, et al. (2005)		N= 464	31/464 **6.68/100** (4.54 – 9.48/100)	**4.1/100** (1.51 -8.94/100)

CHAPTER 5

The development and piloting of a decision aid for parents considering sequential bilateral cochlear implantation for their child with hearing loss

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The development and piloting of a decision aid for parents considering sequential bilateral cochlear implantation for their child with hearing loss

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Abbreviations: BICI: bilateral cochlear implant, CI: cochlear implant, ODSF: Ottawa Decision Support Framework

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Abstract

Objective: To develop and pilot a decision aid for parents considering sequential bilateral cochlear implantation for their children with severe to profound hearing loss.

Design: Pre-test, post-test pilot using the decision aid.

Participants: Eight parents of children currently using one cochlear implant who faced the decision regarding a second cochlear implant. Five clinicians involved in the bilateral cochlear implantation decision.

Intervention: Self-administered sequential bilateral cochlear implantation decision aid that used available local and published evidence and conformed to international patient decisional aid standards.

Main outcome measures: Acceptability of the decision aid among parents and clinicians; changes in baseline decisional conflict; knowledge, choice predisposition.

Results: The decision aid was found to be acceptable to both parents and clinicians. Parents had significantly increased knowledge of the bilateral cochlear implant options, risks, and benefits following completion of the decision aid. Decisional conflict did not significantly decline.

Conclusion: A decision aid for parents considering sequential bilateral cochlear implantation has the potential as a decision support tool. Future work could examine the influence of the decision aid on decisional conflict of various subgroups of parents as well as their expectations of child outcomes.

The Development and Piloting of a Decision Aid for Parents Considering Sequential Bilateral Cochlear Implantation for Their Child with Hearing Loss

Almost immediately after the diagnosis of a hearing loss, parents of children with bilateral, permanent, sensorineural hearing loss are required to make a number of important decisions regarding the communicative rehabilitation of their child. These decisions include the choice of amplification or cochlear implantation, and the communication approach for their child. Most of these parents have never experienced hearing loss themselves which may make the decisions overwhelming (Northern & Downs, 1991). Parents must absorb significant amounts of technical and scientific information during a period of grief about their child's hearing loss (Anagnostou, Graham, & Crocker, 2007; Kurtzer-White & Luterman, 2003). Decisions about treatment vary depending on the severity, characteristics of the child's hearing loss, the information received, as well as the needs of the family.

Cochlear Implantation

A child with a bilateral severe to profound hearing loss may require considerable intervention in the form of amplification and aural rehabilitation in order to develop spoken communication (Samson-Fang, Simons-McCandless, & Shelton, 2000). For these children, cochlear implants (CIs) are one of the available options. CIs are available for use in children one year of age (sometimes younger) with severe to profound losses (Candidacy Criteria, 2008; Thoutenhoofd, Archbold, Gregory, Lutman, Nikolopoulos, & Sach., 2005). Recently, due to the increased awareness of the importance of binaural hearing for sound localization and speech intelligibility in noise, bilateral CIs have become available in some paediatric centres although not universally (Berg, Ip, Hurst, & Herb, 2007).

Research indicates that there are benefits for patients receiving bilateral stimulation

compared to the use of a single CI, demonstrated on measures of speech recognition in noise and sound localization (Brown & Balkany, 2007; Ching, van Wanrooy, & Dillon, 2007; Johnston, Durieux-Smith, Angus, O'Connor, & Fitzpatrick, in press; Murphy & O'Donoghue, 2007; Schafer & Thibodeau, 2006). However, along with the benefits associated with bilateral devices, there are some risks associated with the CI surgery and the implanted device. Although infrequent, facial nerve paralysis, vestibular problems, and infection are some of the complications of CI surgery (Fayad, Wanna, Micheletto, & Parisier, 2003; Fina, Skinner, Goebel, Piccirillo, Neely, & Black, 2003; Gysin, Papsin, Daya, & Nedzelski, 2000). Another major risk is the post-surgical complication of meningitis among children who have received an implant. Recent work has attributed the increased risk of meningitis, in part, to a particular positioner device that has since been withdrawn from the market (U.S. Food and Drug Administration, 2007; Biernath, Reefhuis, Whitney, Mann, Costa, P., Eichwald, et al., 2006).

When parents are considering sequential bilateral cochlear implantation for their child, they are interested in both the benefits and the risks associated with the devices. The rates of these benefits and risks and parents' perception of these risks may influence parental decision-making.

Parental Decision-making

The challenges parents face in deciding on unilateral cochlear implantation for their children have been documented in the literature (Sorkin & Zwolan, 2008; Sach & Whynes, 2005; Li, Bain, & Steinberg., 2004; Incesulu, Vural, & Erkam, 2003; Most & Zaidman-Zait, 2003; Peters, 2000; Steinberg, Brainsky, Bain, Montoya, Indenbaum, & Potsic., 2000). Incesulu et al. (2003) report that 81% of parents responding to a survey indicated that the

unilateral CI decision was the most difficult aspect of the implantation process for them. Most and Zaidman-Zait (2003) also describe the high parental stress during the implantation decision-making process and the specific parental needs for information to aid in the process.

In contrast, Sach and Whynes (2005) of the United Kingdom report that most of the 216 interviewed families found the decision regarding implantation to be straightforward. They did, however, describe the overall stress for families undergoing cochlear implantation. A very recent survey of parents in the United States indicated that those who chose the CI for their child felt that they lacked "comprehensive and bias-free" information when making the decision (Sorkin & Zwolan, 2008). Including patients or their families in their health decisions is an important component of knowledge translation (Holmes-Rover, Llewellyn-Thomas, Entwistle, Coulter, O'Connor, & Rovner, 2001; Coulter, 1997).

Recent work indicates that while parents report that the unilateral cochlear implant decision was straightforward and consistent with their value regarding spoken language and oral communication, they are much more uncertain regarding the sequential bilateral cochlear implant decision (Johnston, Durieux-Smith, Fitzpatrick, O'Connor, Benzies, & Angus., 2008). Parents reported being uncertain of the benefits and risks associated with the second procedure for their child. Similar to the requests from parents from the Sorkin and Zwolan (2008) study for additional information when making the unilateral decision, parents and clinicians interviewed in the Johnston, et al. (2008) study requested additional evidence-based information on the benefits and risks of bilateral implantation to help with decision-making.

Decision Aids for Supporting Decision-making

One approach to providing evidence-based information for health-care consumers is through the use of decision aids (O'Connor & Edwards, 2001). Decision aids are, "tools designed to help people participate in decision making about health care options. They provide information on the options and help patients clarify and communicate the personal value they associate with different features of the options." (International Patient Decision Aid Standards [IPDAS], 2008). Decision aids are particularly important in health situations where there is a choice between two or more options and there is no clear standard of care based on the available evidence (O'Connor & Edwards, 2001). Wennberg (2002) distinguishes between "effective" and "preference sensitive" care. When adequate scientific evidence documenting clear benefits and few harmful effects associated with one health choice is available (i.e. a clear standard of care or "effective" care), there is often little conflict regarding the choice that is to be made. However, many health decisions are not based on clear evidence or the evidence that is available indicates that the choices carry both harms and benefits that may be differentially viewed depending upon the personal preferences of the client (O'Connor, Legare, & Stacey, 2003; Wennberg, 2002). Decision aids are tools designed to help patients become informed regarding preference sensitive options.

A Cochrane systematic review of the effectiveness of decision aids shows that the decision-making quality and process is improved with their use (O'Connor, Stacey, Entwistle, Llewellyn-Thomas, Rovner, Holmes-Rovner, et al., 2002). Improved knowledge and reduced decisional conflict were found among those who use decision aids compared to those who do not. Decision aids were also shown to increase participation in decision-

making without increasing anxiety as well as creating more realistic expectations of outcomes. The effectiveness of decision aids in improving the decision-making process and in appropriately translating research knowledge to consumers makes them applicable in many healthcare situations.

Based on parents' responses, the paediatric bilateral cochlear implant decision appears to be one of these preference sensitive decisions (Johnston, et al., 2008). In order for parents to make a decision that is well informed and consistent with their personal values it is essential that the available relevant research knowledge is provided to them. Decision aids are the most relevant tool for disseminating the current state of knowledge, explaining the choices, and exploring the personal preferences of parents (O'Connor & Edwards, 2001). As such, a decision aid was developed for parents on the topic of sequential paediatric bilateral cochlear implantation.

Purpose

The primary purpose of the study was to develop and pilot a decision aid for parents considering a second cochlear implant for their children with severe to profound hearing losses. Secondly, the purpose was to explore the knowledge and decisional conflict among parents before and after use of the decision aid. There were two research questions for the piloting of the decision aid: 1) Is the decision aid acceptable to parents and clinicians? 2) How does exposure to the decision aid change knowledge and/or decisional conflict among parents?

Method

Decision Aid Development

A needs assessment was previously conducted in order to assess the needs of parents considering cochlear implantation (Johnston, et al., 2008). In addition to showing that parents wanted more info on CIs generally, the work also indicated that parents lacked information on the benefits and risks associated with bilateral cochlear implantation. In order to address this need for additional information, the Ottawa Decision Support Framework (ODSF; O'Connor, Tugwell, Wells, Elmslie, Jolly, Hollingworth, et al., 1998) was chosen as the theoretical framework to guide the decision aid's development and piloting. A standardized Ottawa Patient Decision Aid template that conforms to IPDAS standards (Elwyn, O'Connor, Stacey, Volk, Edwards, Coulter, et al., 2006) was adapted for the bilateral cochlear implant decision.

The objective of the decision aid was to provide parents with information on the options, the risks, and the benefits associated sequential bilateral cochlear implantation. The tool was designed to help people participate in decision-making with their clinicians by clarifying and communicating their personal values and any additional needs.

The benefits associated with bilateral cochlear implantation were systematically gathered from the available published literature and synthesized (Johnston et al., in press). Risks of cochlear implant surgery were also systematically gathered from the literature and local databases (Johnston et al., unpublished research). These documents were used to create a draft decision aid. This document was reviewed by an expert panel comprised of research and clinical audiologists, a cochlear implant surgeon, and decision aid methodologist. The decision aid was developed iteratively based on responses from the expert panel until an

acceptable decision aid was created. The resulting decision aid and background documents including references from the literature that were used to inform the benefit and risk rates are available in Appendix F.

It is important to note that the risks and benefits presented in the decision aid reflect those for a child who has already undergone one cochlear implant surgery. This is particularly relevant for the surgical risks associated with "not accepting a second cochlear implant". As the child has already received one cochlear implant and either experienced complications or not for their first surgery, there is no increased surgical risk when "not accepting the second cochlear implant". The surgical risks that are presented for the acceptance of the second cochlear implant are the same as the risks associated with the first surgery based on expert opinion.

Decision Aid Pilot Study

In order to assess the acceptability of the decision aid to parents and clinicians, an exploratory research pilot study was undertaken. Acceptability of the decision aid was assessed using a post-test research design. A pre-test, post-test research design, was used to examine the decisional conflict and knowledge of parents before and after using the decision aid.

Participants and recruitment. A convenience sample of parents who had indicated an interest in receiving additional information about bilateral CIs was recruited to participate in the study. Eligible participants were parents who had children with one cochlear implant who were considering an implant for their child's second ear.

Hospital CI audiologists and rehabilitation therapists were also invited to participate in the study. Consent for participation was obtained from both parents and clinicians prior to

study commencement. Ethical approval was received from the Children's Hospital of Eastern Ontario and the University of Ottawa, Research Ethics Boards.

Procedure and outcome measures. Parents were asked, by a member of the research team, to complete a questionnaire eliciting the parent's education level, child's and parent's current age, age at identification of hearing loss, age at first implantation, and additional clinical characteristics.. Parents were then asked to complete baseline measures of knowledge and decisional conflict.

The knowledge test was a 5-item questionnaire developed for this study used a standardized template and was endorsed for content validity by the expert panel used during the decision aid's development. Response categories (true, false, or unsure) were coded as correct or incorrect. Content in the knowledge test was reflective of topics covered in the decision aid. It is available in Step 3 of the decision aid (Appendix F)

A previously validated decisional conflict scale was also used (O'Connor, 1995). The decisional conflict scale is a 16-item questionnaire. Test-retest and α coefficients exceed 0.81. Other research indicates that the measure is sensitive to change (O'Connor et al., 1998).

Following the completion of the baseline measures, parents were asked to read the decision aid and to complete the steps of decision making as set out in the decision aid. The decision aid leads readers through four steps that include an introduction of the options, the benefits and risks of the options, exercises for readers to identify the reasons to choose each option that matter most to them, and to identify any additional decisional needs. Following the reading of the decision aid, parents were asked to complete the final knowledge and decisional conflict measures. Finally, parents were invited to complete an acceptability questionnaire which consisted of both open- and closed-ended questions. Closed-ended questions included items that evaluated the amount, length, clarity, and helpfulness of the

information in the decision aid. Other items examined whether the information was presented in a balanced manner and whether they would recommend it to others. This acceptability tool has been used in studies examining decision tools developed for osteoporosis, hormone replacement therapy, lung cancer, and prenatal testing.

The research procedure lasted between 30 and 40 minutes and was conducted at a mutually agreeable location, either in the parents' home or at the clinic. Clinicians were also asked to review the decision aid and to complete the acceptability measure following their review of the decision aid.

Analysis and Interpretation

Frequencies, counts, and averages were used, where appropriate, to summarize parent and child characteristics. For the acceptability of the decision aid, the frequency of parents and clinicians who recommended the decision aid, found it to be the right length, and found that it was helpful, was analysed. A pre-determined acceptability level of at least 70% for the various elements of the decision aid is recommended in the Ottawa Toolkit for Decision Aid Development (O'Connor, Brehaut, Tugwell, Santesso, Bennett, & Stacey, 2006). Any suggested changes were examined closely during this pilot phase as the purpose was to improve the decision aid prior to widespread use.

The parents' correct answers to each item were summed to create an overall knowledge score. The mean overall knowledge score was also calculated. The pre- and post-decision aid overall knowledge scores were examined using a paired t-test.

The decisional conflict measure consists of a total score (ranging from 1-100, with 0 being no decisional conflict to 100 meaning extremely high decisional conflict) and five subscales: uncertainty, informed, values clarity, support, and effective decision. The mean

total and subscale scores were calculated for the pre- and post-decision aid periods to explore any possible changes. The total pre- and post-decision aid decisional conflict scores were examined using a paired t-test.

Results

Characteristics of Participants

Eight parents of nine children eligible for a second cochlear implant participated in the pilot study. There were seven mothers and one father among the parents. Parents ranged in age from 32 to 42 (*Mdn*=37.5 years). All parents had completed high school and seven of the eight had some post-secondary education or a completed bachelor's degree. All families spoke some English in their homes, four of the families also spoke one or more additional languages: French (2), Arabic (1), American Sign Language (1). The children of these parents ranged in age from 2 to 8 (*Mn*=3.9 years). Seven of the children had been identified with their hearing loss prior to 7 months of age. Two of the children were identified at age 2. The average age at first cochlear implantation was 2.4 years of age (*Range*=1-5years). All children used auditory verbal therapy (AVT) as their primary communication approach. Two children used American Sign Language as a secondary communication approach and one child used pictograms as well.

Five clinicians (audiologists and auditory-verbal therapists) participated in the study. All of these participants have a wide range of experience with children with cochlear implants and their parents.

Acceptability

Figure 1 presents the percentage of respondents, both parents and clinicians (N=13), reporting acceptability of various elements of the decision aid. All of the included elements were acceptable to over 70% of participants. Participants were asked to rate, on a four-point scale from "poor" to "excellent", the way the various information sections of the decision aid was presented. These ratings are seen in Figure 2. No participants rated any section as, "poor." The area rated as "fair" by the largest number of participants was the area describing the criteria for bilateral cochlear implants.

Parents and clinicians were also given an opportunity to provide written comments on what they liked about the decision aid as well as suggestions for improvement. Five of eight parents commented directly on the benefits of seeing the benefits and risks clearly displayed. Comments included, "The facts are very clear and concise," and "The benefits and risks were great to see." Other comments, from both parents and clinicians spoke to the positive elements of the decision aid in general, such as, "written information to take away in a rational format." Parents appreciated receiving additional information, "It's more information that I did not have before."

The suggestions for improvement were consistent with the rating from participants on the various sections of the decision aid. "It needs more regarding candidacy criteria," was a comment received from a parent. Others suggested more information on the effect of the timing of the surgery and the importance of a backup device for some families.

Knowledge

The average knowledge score of participating parents before reading the decision aid was 2.65/5 (Mdn=3, $Range$=0-5, SD=1.60). Following the reading of the decision aid, the

average score was 4.25/5 (*Mdn*= 4.5, *Range*=3-5, *SD*=0.89). Parents showed a statistically significant increase in the knowledge of the bilateral cochlear implant options, risks, and benefits following the reading of the decision aid (t=-3.87, p=0.0061).

Decisional Conflict

The average total decisional conflict score of participating parents before reading the decision aid was 14.26 (*Mdn*= 11.72, *Range*=0-34.38, *SD*=13.16). Following the reading of the decision aid, the average total decisional conflict score was 6.44 (*Mdn*=5.47, *Range*=0-15.63, *SD*=5.87). There was a trend toward a reduction in decisional conflict among participants but it was not statistically significant (t=1.82, p=0.111).

While there was not a statistically significant reduction of decisional conflict scores for the group, there were a number of meaningful changes in decisional conflict among two participants. A total decisional conflict score below 25 is associated with the implementation of a decision and lower decisional conflict. Six of the eight participants had decisional conflict scores below 25 at the onset of the study. The two parents with decisional conflict scores above 25 (34.38 and 32.81) reduced their scores below the 25 point threshold following the reading of the decision aid (15.63 and 0).

Discussion

Previous research indicated that parents found the bilateral cochlear implantation decision challenging. In particular, parents requested additional information on the risks and benefits of bilateral cochlear implantation due to their lack of knowledge about the procedure and its outcomes (Johnston et al., 2008). The decision aid in the current study was developed in order to meet this decisional support need of parents. While there are some small changes

suggested by parents and clinicians who have piloted this decision aid, the tool is acceptable to these users. This feedback provided by parents is important and will be examined closely by the expert panel when they meet to incorporate these results into the final decision aid. The significant improvement in knowledge among users of the decision aid indicates that the tool is meeting an informational and a decision-making need of parents.

The results of this pilot are similar to those of other studies evaluating the use of decision aids in clinical contexts. A systematic review of the effectiveness of decision aids indicates that improved knowledge and reduced decisional conflict are common among those who use decision aids compared to those who do not (O'Connor, et al, 2002). The results of this pilot indicated a significant increase in knowledge of participating parents. While decisional conflict did not significantly decline in the small group of participants in this pilot, there were some meaningful decisional conflict changes for two of the eight participants. A larger-scale evaluation is required in order to determine if the sequential bilateral cochlear implant decision aid is effective in reducing decisional conflict.

Previous research indicates that decision aids increase participation in decision-making without increasing anxiety as well as creating more realistic expectations of outcomes (O'Connor, et al, 2002). These outcomes were not examined in this pilot study but may be important for future research and of interest in a larger-scale evaluation of this decision aid. Parents of children with hearing loss most often have not experienced a hearing loss themselves and they are often faced with a great deal of new scientific and technical information (Northern & Downs, 1991). Finding ways to help parents absorb this information without increasing anxiety could be an important clinical tool.

This is the first decision aid developed for a paediatric cochlear implant topic. It is also the first decision aid developed for an audiology topic (Cochrane Inventory of Patient

Decision Aids, 2008). This decision aid for parents deciding about sequential bilateral cochlear implants is evidence-based and conforms to IPDA Standards (IPDAS, 2008). It will join the 255 other decision aids, for a variety of topics, that have been previously developed to help provide decisional support for preference sensitive health care decisions.

Limitations

As the primary purpose of this study was to examine the acceptability of the decision aid before a larger-scale evaluation of the effects of the decision aid, this study was underpowered to detect a significant change in decisional conflict. Between 51 and 68 parents are required to determine an effect size of .40 on the decisional conflict scale with a type 1 error of 0.05 and a power between 80% and 90%. Previous literature indicates that effect sizes on the decisional conflict scale have been in the range of 0.43-0.82 (O'Connor et al., 1998). Future evaluation of the decision aid should aim for this number of parents. This number of parents would have sufficient power to detect before-after changes in subgroups who differ in baseline predisposition towards options (e.g. unsure versus leaning toward a specific option), education level (e.g. above or below the median numbers of years of education of father), knowledge (e.g. above and below the baseline median knowledge score), and parent (e.g. mother or father).

The decision aid and pilot were designed and piloted at a single cochlear implant centre in Canada. The rates of benefits and risks were developed from literature in the public domain and a standard decision aid template was used in order to make the decision aid as universal as possible. However, as the decision aid development team was from a single Canadian centre it is possible that there are elements of the decision aid that may not apply to other centres.

Conclusions

A decision aid was developed for parents making the sequential bilateral cochlear implant decision. The decision aid used available local and published evidence and conformed to International Patient Decision Aid Standards. The decision aid was acceptable to both parents and clinicians. The decision aid has the potential to become a useful decisional support tool as users of the decision aid showed increased knowledge of the bilateral cochlear implant options, risks, and benefits. Some participants experienced meaningful reductions of their decisional conflict surrounding the sequential bilateral cochlear implant decision after reading the decision aid. However, a more representative evaluation is needed. Future work could examine the influence of the decision aid on decisional conflict for various subgroups and the influence that the decision aid has on expectations of child and family outcomes.

References

Anagnostou, F., Graham, J., & Crocker, S. (2007). A preliminary study looking at parental emotions following cochlear implantation. *Cochlear Implants International, 8,* 68-86.

Berg, A.L., Ip, S.C., Hurst, M. & Herb, A. (2007). Cochlear implants in young children: informed consent as a process and current practices, *American Journal of Audiology, 16,* 13-28.

Biernath, K. A., Reefhuis, J., Whitney, C. G., Mann, E. A., Costa, P., Eichwald, J., et al. (2006). Bacterial meningitis among children with cochlear implants beyond 24 months after implantation. *Pediatrics, 117,* 284-9.

Brown, K.D. & Balkany, T.J. (2007). Benefits of bilateral cochlear implantation: a review, *Current Opinions in Otolaryngology Head and Neck Surgery, 15,* 315-8.

Candidacy Criteria. (2004). Retrieved February 9, 2008 from http://www.cochlear.com/Support/ 631.asp

Ching, T.Y., van Wanrooy, E. & Dillon, H. (2007). Binaural-bimodal fitting or bilateral implantation for managing severe to profound deafness: a review, *Trends in Amplification, 11,* 161-92.

Cochrane Inventory of Patient Decision Aids. (2008). Retrieved June 10, 2008 from http://decisionaid.ohri.ca/cochinvent.php

Coulter, A. (1997). Partnerships with patients: The pros and cons of shared decision making. *Journal of Health Services Research and Policy, 2,* 112-21.

Elwyn, G., O'Connor, A., Stacey, D., Volk, E., Edwards, A., Coulter, A. et al. on behalf of the International Patient Decision Aids Standards (IPDAS) Collaboration. (2006).

Developing a quality criteria framework for patient decision aids: online international Delphi consensus process. *British Medical Journal, 333*, 417.

Fayad, J. N., Wanna, G. B., Micheletto, J. N., & Parisier, S. C. (2003). Facial nerve paralysis following cochlear implant surgery. *Laryngoscope, 113*, 1344-6.

Fina, M., Skinner, M., Goebel, J. A., Piccirillo, J. F., Neely, J. G., & Black, O. (2003). Vestibular dysfunction after cochlear implantation. *Otoogy and Neurotology, 24*, 234-42.

Gregoret, L. (2003). Bilateral cochlear implants: are they in your child's future? *Volta Voices, 10*, 10-15, 34.

Gysin, C., Papsin, B. C., Daya, H., & Nedzelski, J. (2000). Surgical outcomes after paediatric cochlear implantation: diminution of complications with the evolution of new surgical techniques. *Journal of Otolaryngology, 29*, 285-9.

Holmes-Rovner, M., Llewellyn-Thomas, H., Entwistle, V., Coulter, A., O'Connor, A., & Rovner, D.R. (2001). Patient choice modules for summaries of clinical effectiveness: A proposal. *British Medical Journal, 322,* 664-7.

Incesulu, A., Vural, M., & Erkam, U. (2003). Children with cochlear implants: Parental perspective. *Otology and Neurotology, 24,* 605-11.

International Patient Decision Aid Standards. (2008). What are decision aids? Retrieved June 10, 2008 from http://ipdas.ohri.ca/what.html

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. *Canadian Journal of Speech-Language Pathology and Audiology, 32,*169-82.

Johnston, J.C., Durieux-Smith, A., Angus, D., O'Connor, A., & Fitzpatrick, E. (in press) Bilateral paediatric cochlear implants: A critical review. *International Journal of Audiology.*

Kurtzer-White, E., & Lutterman, D. (2003). Families and children with hearing loss: Grief and coping. *Mental Retardation and Developmental Disability Research Review, 9,* 232-5.

Li, Y., Bain, L., & Steinberg, A. G. (2004). Parental decision-making in considering cochlear implant technology for a deaf child. *International Journal of Pediatric Otolaryngology, 68,* 1027-38.

Most, T., & Zaidman-Zait, A. (2003). The needs of parents of children with cochlear implants. *Volta Review, 103,* 99-113.

Murphy, J., & O'Donoghue, G. (2007). Bilateral cochlear implantation: an evidence-based medicine evaluation, *Laryngoscope, 117,* 1412-8.

Northern, J., & Downs, M. P. (1991). *Hearing in children* (4th ed.). Baltimore, MD: Williams & Wilkings.

O'Connor, A. M. (1995). Validation of a decisional conflict scale. *Medical Decision Making, 15,* 25-30.

O'Connor, A., Brehaut, J., Tugwell, P., Santesso N., Bennett, C., & Stacey, D. (2006). *Toolkit for Applying IPDAS Standards in Developing and Evaluating Ottawa Patient Decision Aids.*

O'Connor, A., & Edwards A. (2001). The role of decision aids in promoting evidence-based patient choice. In A. Edwards, G. Elwyn (Eds.), *Evidence-based patient choice* (pp. 220-42). Oxford, UK: Oxford University Press.

O'Connor, A. M., Legare, F., & Stacey, D. (2003). Risk communication in practice: the contribution of decision aids. *British Medical Journal, 327*, 736-40.

O'Connor, A.M., Stacey, D., Entwistle, V., Llewellyn-Thomas, H., Rovner, D., Holmes-Rovner, M., et al. (2002). Decision aids for people facing health treatment or screening decisions (Cochrane Review). Retrieved on July 18, 2008 from http://www.cochrane.org/reviews/en/ab001431.html

O'Connor, A. M., Tugwell, P., Wells, G. A., Elmslie, T., Jolly, E., Hollingworth, G., et al. (1998). A decision aid for women considering hormone therapy after menopause: Decision support framework and evaluation. *Patient Education and Counseling, 33*, 267-79.

Peters, E. (2000). Our decision on a cochlear implant. *American Annals of the Deaf, 145*, 263-7.

Sach, T. H., & Whynes, D. K. (2005). Paediatric cochlear implantation: The views of parents. *International Journal of Audiology, 44,* 400-7.

Samson-Fang, L., Simons-McCandless, M., & Shelton, C. (2000). Controversies in the field of hearing impairment: early intervention, educational methods, and cochlear implants. *Infants and Young Children, 12*, 77-88.

Schafer, E.C. & Thibodeau, L.M. (2006). Speech Recognition in noise in children with cochlear implants while listening in bilateral, bimodal, and FM-system arrangements, *American Journal of Audiology*, *15*, 114-26.

Sorkin, D. L., & Zwolan, T. A. (2008). Parental perspectives regarding early intervention and its role in cochlear implantation in children. *Otology and Neurootology, 29*, 137-41.

Steinberg, A., Brainsky, A., Bain, L., Montoya, L., Indenbaum, M., & Potsic, W. (2000).

Parental values in the decision about cochlear implantation. *International Journal of Pediatric Otorhinolaryngology, 55,* 99-107.

Thoutenhoofd, E., Archbold, A., Gregory, S., Lutman, M., Nikolopoulos, T., & Sach, T.H. (2005). *Paediatric cochlear implantation: Evaluating outcomes*. London: Whurr.

United States Food and Drug Administration. (2007). Advice for patients with cochlear implants: New information on meningitis risk. Retrieved May 18, 2008 from http://69.20.19.211/cdrh/medicaldevicesafety/atp/101007-cochlear.html

Wennberg, J. E. (2002). Unwarranted variations in healthcare delivery: Implications for academic medical centres. *British Medical Journal, 325*, 961-4.

Figure 1: Number of respondents, both parents and clinicians, reporting acceptability of various elements of the decision aid (*N*=13)

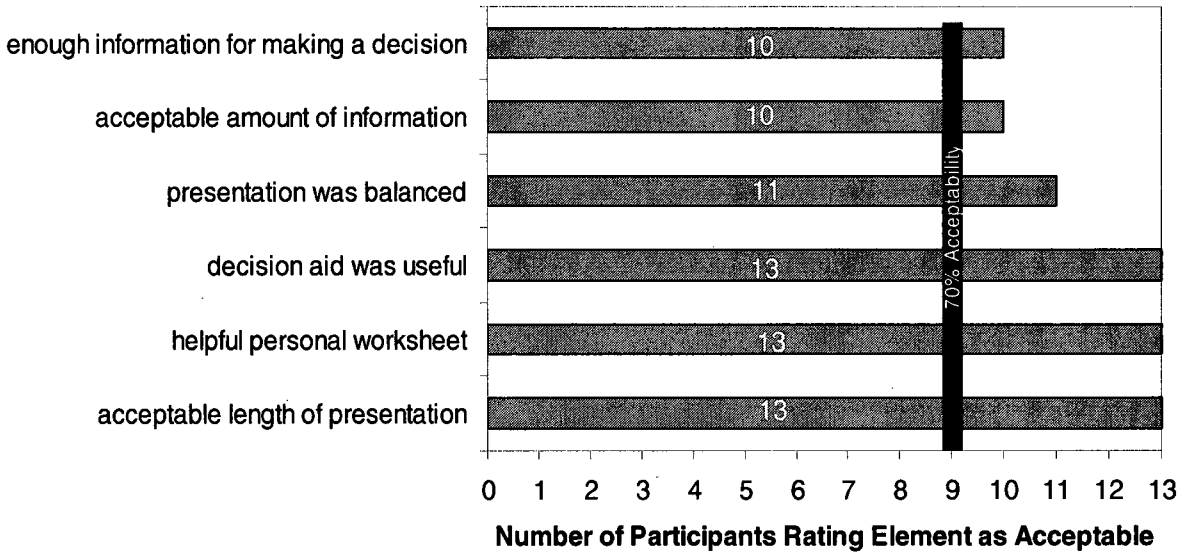
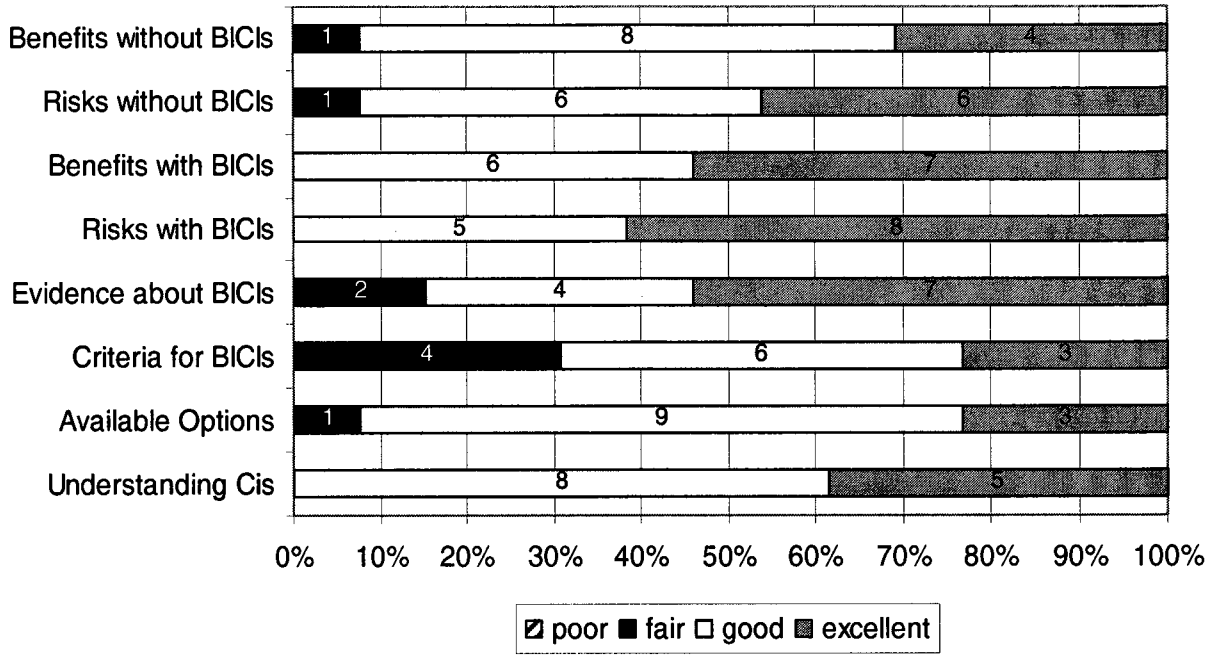


Figure 2: Participants' ratings of various sections of the decision aid (N=13).



CHAPTER 6:

Summary and Integration of Thesis Research

Objectives of the Research

There were five objectives for this doctoral research. All of these objectives were accomplished during the thesis research program. The five objectives were:

1) To explore the decision-making needs of parents regarding unilateral and BICIs, including their: (a) perceived options, advantages, and disadvantages; (b) parental perceptions of their knowledge and expectations, values, support, and resources available to them; (c) decisional conflict and contributing factors when making the decisions; and (d) need for a decision aid to support parents and clinicians
2) To provide clinicians, families, and researchers with updated, comprehensive information on the available paediatric BICI literature
3) To estimate risks for cochlear implantation at a local CI center and in the published literature
4) To develop a decision aid for sequential bilateral paediatric cochlear implantation that is acceptable to clinicians and parents, using IPDAS standards, the Ottawa Decision Aid Template, and available data
5) To pilot a decision aid and describe changes among parents in knowledge and decisional conflict after using a patient decision aid.

Context

Bilateral CIs (BICIs) had only recently been offered as a clinical option in a small number of clinics at the commencement of the thesis research (Berg et al., 2007). At that time BICIs were not, nor are they currently, offered as the standard of care for children with bilateral hearing loss. At the onset of the research there were no clinical practice guidelines on this new procedure nor was there any information on why parents were choosing or

declining this option. There was also limited literature on the outcomes associated with the procedure.

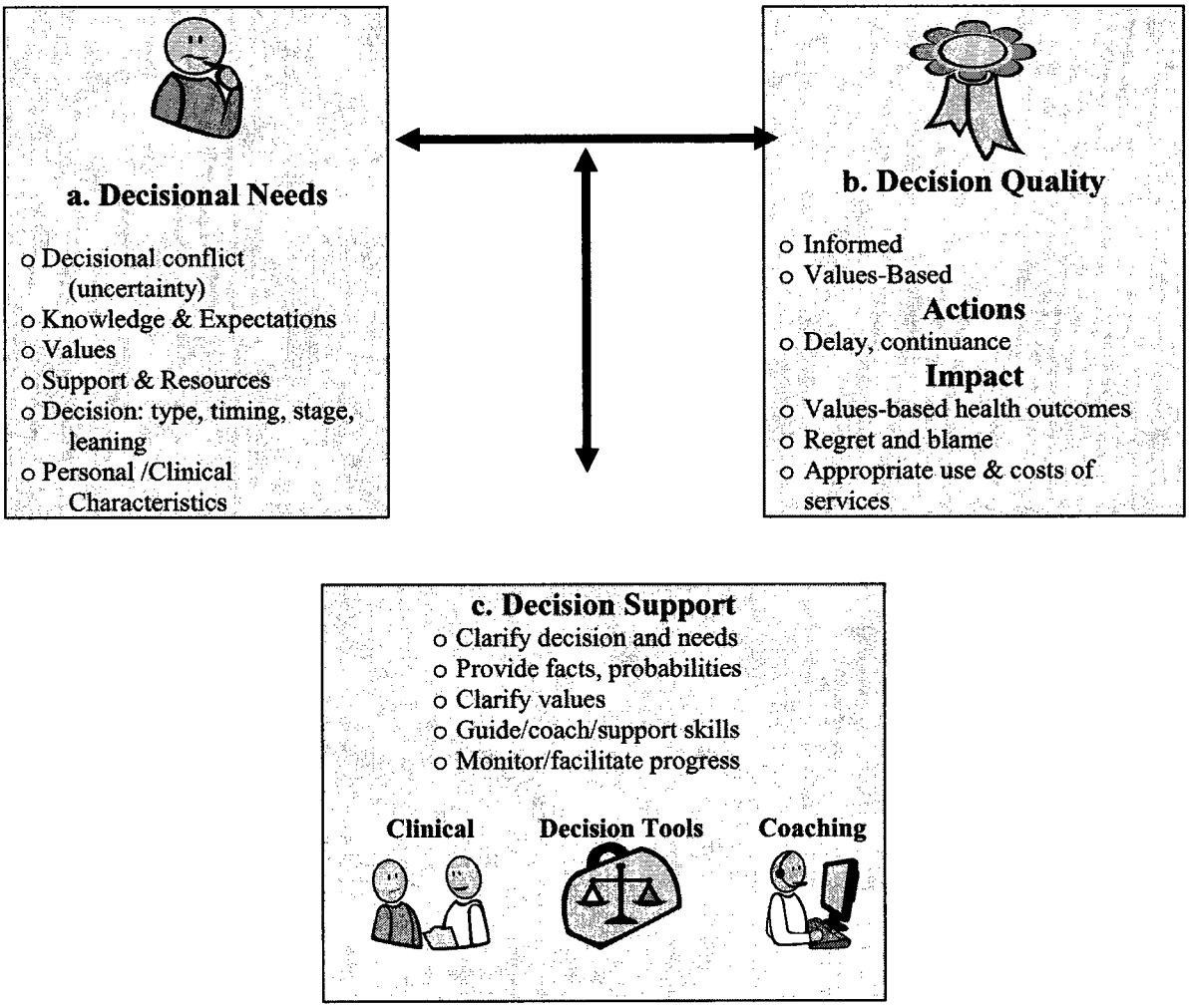
In the cochlear implant centre where the research took place, a paediatric cochlear implant (CI) centre in Eastern Ontario that completes 20-24 CI surgeries per year, the bilateral option was offered only to parents of children with hearing loss as a result of meningitis who are at risk for cochlear ossification following the disease. This ossification can prohibit or impede the implantation of the CI electrode. Now, upon research completion, parents of children with hearing loss of any etiology may opt to be considered for BICI surgery. Provincial funding of CI devices currently limits the number of CI surgeries (including bilaterals) that are completed each year. The changing of the candidacy criteria for BICI mid-project may partly explain parents' need for additional information on the candidacy criteria for BICI.

The BICI research context has also changed in tandem with the clinical changes over the duration of this thesis research. During the period of thesis research there have been numerous publications on bilateral cochlear implantation, including reviews, primary research, and clinical practice guidelines (Brown & Balkany, 2007; Ching et al., 2007; Murphy & O'Donoghue, 2007; Perreau, Tyler, Witt et al., 2007; William House Cochlear Implant Study Group, 2008). No other studies have examined how parents are making the BICI decision for either sequential or simultaneous implantation. Keeping up to date with the rapidly changing state of literature and needs for the BICI decision has been an ongoing challenge of the project.

Theoretical Basis of the Research

The Ottawa Decision Support Framework (ODSF, O'Connor et al., 1998) and the Population Health Promotion Model (Health Canada, 2005) both served as theoretical frameworks for this thesis research. The ODSF (O'Connor et al., 1998) provided the primary theoretical basis for the research. This theoretical framework (presented in Figure 1) depicts how a family's decisional needs and quality influence each other and how decision-making support can be used to address unmet needs to improve the quality of decisions. This

Figure 1: Ottawa Decision Support Framework



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AM O'Connor, Ottawa Decision Support Framework to Address Decisional Conflict © 2006

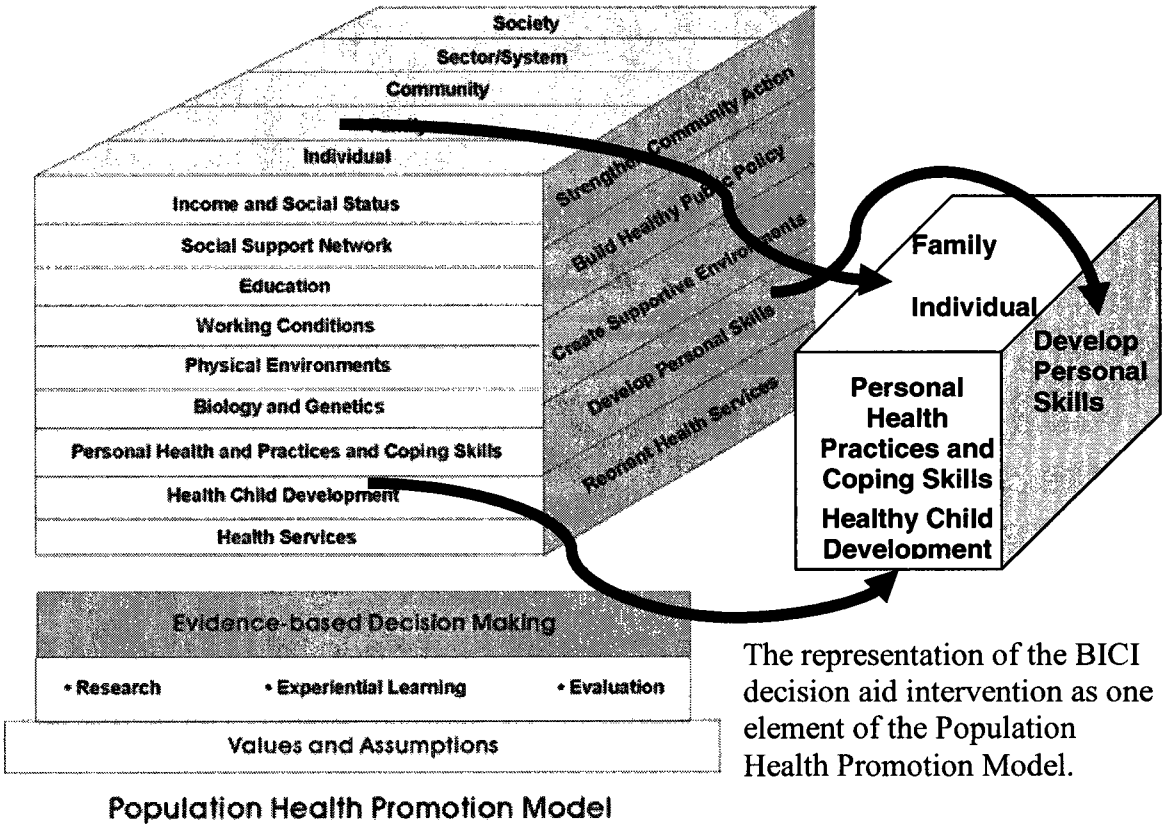
theoretical framework was essential in all stages of the thesis work. It was used in assessing the needs of parents, developing the decision aid, and in the pilot study of the tool. In particular, it was used in the design of the research program and the sequence of studies, the study objectives, the framework for qualitative analysis, and the outcomes of interest.

The first paper of thesis research addressed the first objective of the thesis project. It focused on understanding the decision and parents' needs around the decision (elements of the decision, decisional conflict, knowledge and expectations, and values) (component [a] of the ODSF). The second and third research papers addressed objectives two and three of the thesis project and focused on developing evidence for providing appropriate decisional support (component [c] as defined by the ODSF). The fourth research paper addressed objectives four and five of the thesis research and described how the earlier gathered information was used to provide decisional support through the development and piloting of the decision aid. This was accomplished by 1) providing access to information on the health situation, options, and outcomes, 2) re-aligning expectations by presenting probabilities of outcomes, 3) clarifying personal values for outcomes, 4) providing guidance in decision making. This paper also examined the acceptability of the newly developed tool as well as some elements of the quality of the decision (knowledge, decisional conflict) (component [b] of the ODSF).

The Population Health Promotion Model (Health Canada, 2005) is presented in Figure 2 along with a representation of how the thesis work fits within the framework. This three dimensional model of health represents how the health of a population can be influenced by the determinants of health through the use of a variety of health promotion strategies. The decision aid intervention developed in this thesis research fits into the population health model by developing personal skills for the family or individual with the

objective of improving their personal health practices and coping skills as well as encouraging healthy child development. It has been demonstrated in this thesis research that this population health intervention has improved parent knowledge and the potential to

Figure 2: Population health promotion model and a representation of the thesis research within this model.



The representation of the BICI decision aid intervention as one element of the Population Health Promotion Model.

improve decisional conflict regarding a decision important to the parent/child interaction. Further research is required to identify if parents who use the decision aid for the bilateral cochlear implantation decision will apply these decision-making skills to other health decisions for themselves or their children in the future.

The evidence base of the Population Health Promotion Model was an essential component of this thesis research. Many different forms of evidence were used in designing the decision aid. Evidence from systematic reviews was essential in identifying the benefits and risks of the bilateral implantation and evidence from parent and clinician experience was necessary for identifying the needs of these stakeholders.

Method

The methods used in this thesis research have been described in full in other sections of this document. Table 1 provides a summary of the methods used in each project.

Table 1

Major Projects and Research Design	Method
Needs Assessment • Semi-structured interviews with parents currently making the decision and with parents post-decision using the ODSF • Semi-structured interviews with clinicians	**Participants:** 3 families awaiting their CI surgery, 5 families post-CI, 9 clinicians **Measures:** Semi-structured interview guide based on standard Ottawa template **Analysis:** Content analysis for interview data using a deductive coding strategy based on ODSF, descriptive statistics for structured interview responses
Development of resources for the decision aid • Systematic Review • Secondary analysis of existing local database of patient cohort for estimating probabilities of risk • Systematic standardized methods for development of decision aid with expert panel	**Procedure:** • Critical review of benefits to ensure evidence-based development of decision aid • Estimating probabilities of harms using local databases and available literature • Development of a decision aid that used the Ottawa decision aid Template and applied International Patient Decision Aid Standards (IPDAS) standards
Description of parents' and practitioners' responses to the decision aid • Pre-test, post-test design in which parents' knowledge and decisional conflict is measured at the introduction of the topic, and upon completion of the decision aid • Acceptability questionnaire and semi-structured interviews with clinicians who have used the decision aid	**Participants:** 8 Parents undergoing BICI decision-making, 5 clinicians **Measures:** Standardized measures of knowledge and decisional conflict taken pre-, and post- decision aid. Acceptability questionnaire completed post-decision aid **Analysis:** Descriptive statistics, paired t-tests, content analysis of interviews using an inductive coding strategy

A mixed methods approach was used for the research design of the study. This approach seeks to use both qualitative and quantitative research methods as needed to answer research questions (Tashakkori & Teddlie, 2003). In a mixed method approach, a researcher is required to competently move between qualitative and quantitative research approaches. The three studies and four papers demonstrate my ability to complete research using both qualitative and quantitative methods.

Summary and Interpretation of the Findings

Needs Assessment

In the first study of the thesis, interviewed parents reported that their decision to undergo cochlear implantation for their children with significant hearing loss was related to the value that their specific family placed on oral communication. When asked about the information that they would have liked during CI decision-making, parents reported that additional information on the risks and benefits associated with cochlear implantation as well as information on what others have chosen should be consistently offered to all families making the CI decision. Based on the interviews with parents and clinicians during the study, it appears that the choice for a single CI is a value-based, informed decision for most parents and seemed to present little decisional conflict. This research was similar to other research that has examined the challenges of making decisions for others that has indicated that parents find surrogate decision-making difficult (Fagerlin et al., 2001; Tait et al., 2001).

The situation was somewhat different for those parents contemplating bilateral cochlear implantation for their child. The need for information about bilateral cochlear implantation decision-making is important due to the lack of existing resources for parents. Results of the study indicated that there appears to be increased variability in parental values

of the benefits and risks associated with bilateral cochlear implantation. This study was the first in the published literature to document the decision-making of parents considering bilateral cochlear implantation.

The critical review of the risks and benefits of cochlear implantation and the local examination of risks were undertaken to gather additional information on bilateral cochlear implantation and to meet the identified needs of parents from the first study. These benefits and risks were also essential in developing a patient decision aid for the bilateral cochlear implantation decision.

Development of Resources for the Decision Aid

The replicable, critical review of paediatric bilateral cochlear implantation was conducted in order to identify and document any benefits associated with BICI. The review was a significant contribution to the field as it expanded the information from other previously published reviews on the topic. In the thesis review of BICIs, additional paediatric studies were identified that examined outcomes of interest beyond speech recognition in noise and sound localization. The available articles on bilateral implantation in children provided evidence that there are benefits to children who received a second CI. The benefits included improved performance on speech recognition in noise and sound localization tasks, and auditory brainstem and cortical evoked potential responses. In addition, children with BICIs that were activated at earlier ages and with shorter durations between surgeries appear to receive greater benefit than those implanted later and with longer periods between surgeries (Grieco-Calub et al., 2008; Litovsky et al., 2004; Galvin et al., 2007a; Galvin et al., 2007b; Litovsky et al., 2006; Peters et al., 2004).

The review also highlighted the gaps in the literature. There were a variety of

outcomes of interest that were not available in the published literature. It is hoped that in the future, studies that explore a variety of additional outcomes- including cost-effectiveness, quality of life, speech, language and psycho-educational outcomes will provide additional support for parents and clinicians facing the BICI decision. Identifying the factors that lead to successful bilateral implantation as well as the potential risks associated with a second CI should also be a priority and is critical for decision-making.

Following the examination of benefits associated with BICIs, it was important to gather the risks associated with the CI surgery in order to provide a balanced representation of the procedure. A hospital-based retrospective chart review and review of the literature were conducted to provide parents and clinicians with estimations of the risks associated with paediatric cochlear implantation both locally and in the available literature.

The risks study identified that there are some risks of adverse events associated with cochlear implantation surgery. Results of this study indicated that the rates of complications at the local Canadian paediatric centre examined in this paper are not significantly different from the rates from the literature for all examined complication types. To parents who have opted for the surgery, the complication rates are acceptable, in light of the benefits, (Johnston et al., 2008).

The complication rates, together with information on the benefits of CI use, were information that the parents interviewed in the needs assessment had requested. These rates of risks and benefits were used in the next stage of research, the development of the decision aid.

Development and Description of Responses to the Decision Aid

A decision aid was developed for parents making the sequential BICI decision. The decision aid used the benefit and risk data collected in the reviews of the published literature and from local databases. The decision aid also conformed to IPDAS. An iterative development approach was used with feedback provided by research and clinical audiologists and a CI surgeon. This iterative approach ensured that the decision aid was acceptable to various clinicians.

The pilot of the decision aid identified that the tool was acceptable to both parents and clinicians. The decision aid has the potential to become a useful decisional support tool as users of the decision aid showed increased knowledge of the BICI options, risks, and benefits. Two of the eight participants experienced meaningful reductions of their decisional conflict surrounding the sequential BICI decision after reading the decision aid, however the sample was too small to identify if these changes would occur consistently among most parents or if only among different subgroups of parents.

Integration of Findings

There are a number of studies in the literature that have examined the decision-making of parents with regards to cochlear implantation (Incesulu, et al., 2003; Li, et al., 2004; Most & Zaidman-Zait, 2003; Peters, 2000; Sach & Whynes, 2005; Sorkin & Zwolan, 2008; Steinberg, et al., 2000). These studies have typically used study-designed questionnaires or interview guides to examine various elements of the parental decision-making process. This is the first study in the available Audiology literature to contextualize a decision from the field in the larger decision-making literature context. All aspects of this thesis study used standardized tools that have been used in the wider decision-making

literature, from the needs assessment questions of Jacobson and O'Connor (2006) to the IPDAS for the development of the decision aid.

These are also the first articles in the Audiology literature that provide an intervention for improving the decision-making of either parents or patients. The positive effect of decision aids on knowledge and decisional conflict is well documented in the literature (O'Connor et al., 2002). There is considerable evidentiary support for the types of decision aids that have similar criteria to the decision aid designed in this study. It is anticipated that by using these evidence-based tools from another, related field, audiologists can improve their clinical service delivery.

Relevance to Population Health

Permanent hearing loss is the most common congenital disorder (American Academy of Pediatrics, 2003). It occurs in 1 to 4 per 1000 children. Permanent hearing loss in children, unless very minimal, requires intervention in order for normal oral language development to occur (Samson-Fang, Simons-McCandless, & Shelton, 2000). These children, particularly those with severe and profound losses, are at risk for language delays, low literacy levels, and poor educational and employment opportunities (Mohr et al, 2000). The population health approach, as described by Health Canada (2005) is, "an approach to health that aims to improve the health of the entire population and to reduce health inequities among population groups." Children with hearing loss are a unique population. This thesis research developed and piloted an intervention designed to improve the decisions of a special population of parents and children with severe and profound hearing loss.

Many of the key components of a population health approach as defined by Health Canada (2005) were used in this thesis research. This research addressed the determinants of

health of child development and personal health practices and coping skills, used an evidence-based approach, engaged multiple stakeholders in the development of intervention, applied multiple strategies to improve decision-making, and intervened upstream in the decision-making process.

One of the determinants of health that is addressed by this thesis is "healthy child development". Parents are key contributors to the health of their children. Parent/child interactions are essential for language, cognitive, and social development. The decisions that parents make for their children with regards to cochlear implantation may have a long-lasting influence on the relationship that they will have with each other. Providing a tool that helps parents make the best choice for their family and child that is consistent with their values is important for the parent/child relationship and the development of their child.

Another determinant of health addressed by this project is, "personal health practices and coping skills" of the population (Health Canada, 2001). In order to positively influence the personal health practices of the population it is important that evidence of positive health practices be made available to health care consumers. Patients or health care consumers, require research dissemination for making optimal health decisions and improving their personal health practices. This thesis work provided evidence in an appropriate format for parents in order for them to have the appropriate information on a medical procedure for their children. This decision aid has been shown to improve parents' knowledge about the bilateral cochlear implantation procedure. It is hoped that this increase in knowledge will improve the parents' ability to make decisions for their children.

The evidence-based foundation of the population health perspective is highly relevant to the field of Audiology (Fitzpatrick, Johnston, Angus, & Durieux-Smith, 2006). This thesis work has increased the available evidence, as well as provided a tool for sharing research

knowledge with parents. The CI decision aid will act as a model for the development of decision aids in the field of Audiology and for the use of an evidence-based approach in general.

Overall Strengths and Limitations of the Research

The greatest strength of this project was the systematic approach, centered in the available published literature that was used for all stages of the thesis research. The rates of benefits and risks were developed from literature in the public domain and a standard decision aid template was used in order to make the decision aid as universal and evidence-based as possible. This evidence-based approach was essential as only a single Canadian CI site was included in this study for the needs assessment and the design and piloting of the decision aid.

Unlike some other CI centres, the centre used in this research offers auditory-verbal therapy as the only treatment option for children and families. Families and clinicians from other centres may have different perceptions of the CI decision-making process and different needs based on the information shared with them by the clinicians in their local centres. It is possible that there are elements of the decision aid that may not apply to other centres. However the evidence-base used for its development will allow other centres to explore the potential for the decision aid to be used with their populations.

A limitation of the research was that the sample sizes for the needs assessment, local risk estimation, and the decision aid pilots were relatively small. As only 20-24 children are implanted each year in the study CI centre, the population of families making the decision and pursuing implantation is also small. Data gathered in the needs assessment indicated that data saturation was achieved even with the small sample. As the primary purpose of this

study was to examine the acceptability of the decision aid before a larger-scale evaluation of the effects of the decision aid, this study was underpowered to detect a significant change in decisional conflict. Between 51 and 68 parents are required to determine an effect size of 0.40 on the decisional conflict scale with a type 1 error of 0.05 and will give a power between 80% and 90%. Previous literature indicates that effect sizes on the decisional conflict scale have been in the range of 0.43-0.82 (O'Connor et al., 1998). Future evaluation of the decision aid should aim for this number of parents. This number of parents would have sufficient power to detect before-after changes in subgroups who differ in baseline predisposition towards options (e.g. unsure versus leaning toward a specific option), education level (e.g. above or below the median numbers of years of education of father), and knowledge (e.g. above and below the baseline median knowledge score).

The included reviews of the benefits and risks of bilateral cochlear implantation provide important sources of information for clinicians and parents. However, the conclusions that can be arrived at using systematic review methods are entirely dependent upon the quality of research that has been previously conducted. Due to the nature and very recent use of bilateral implantation, there are methodological limitations to the body of research currently available on the topic. This includes small samples without control groups, differing testing conditions and outcome measures that often preclude quantitative comparisons between studies. As such, no meta-analysis was possible at this time. It is also expected that additional publications will emerge in the near future. These articles will hopefully provide additional information for parents and clinicians. This review did not seek out unpublished results or conference proceedings which may include the results of bilateral studies currently underway. As with any health research, but particularly for this rapidly

evolving field, there will be a need to update the reviews and the decision aid as new research is published.

While the review of benefits tried to include additional outcomes of interest to parents beyond speech perception and sound localization, no additional literature on quality of life, language development, school outcomes, or social development were identified. Only medical, surgical, and device failure risks were examined in the review of risks. There was no inclusion in the literature review of possible social or cultural risks associated with cochlear implantation. These possible social or cultural risks would often only become apparent as children became adolescents or adults and became involved with the Deaf community. An additional review of the literature of adolescent and adult CI users would have to be included in future research to explore these possible risks. With regards to the relevance for this research, it is unlikely that there would be a significant difference between users of unilateral and bilateral CIs for these social outcomes but rather for CI users in general.

Implications of the Research

This thesis research has a number of clinical implications. The entire research process has identified informational needs in the delivery of paediatric cochlear implantation services and designed an intervention to address this need. The decision aid provides a clinical tool that, due to the use of evidence from the published literature, may be of use in all paediatric CI centres. While there may be variations from site to site in parents' preferences in choosing CI surgery for their children and in the values associated with the benefits and risks, the tool will be helpful in ensuring the informational needs of parents are met. As this is the first example of the decision aid developed in Audiology and the field is generally unfamiliar

with the tools, there may be reticence among clinicians to adopt its use. It is important to emphasize that the decision aid is a clinical tool to be used to complement clinical counseling and information sharing rather than to replace it. The decision aid has been shown to improve parents' knowledge of bilateral cochlear implantation and potentially improve decisional conflict. All CI clinicians may be interested in a tool that has the potential to improve parents' decision making.

The needs assessment identified information gaps for some parents facing the cochlear implantation decision at one Canadian CI centre. These gaps were in the information associated with the benefits and risks and the choices that other families have made. It will be important for clinicians in other centres to consider the information needs of parents at their centres and to provide appropriate support. Clinicians at all CI centres should consistently provide evidence on the risks and benefits of cochlear implantation and information about the choices of other families to all parents.

This research has also identified that bilateral cochlear implantation is a preference sensitive decision for parents. It may be important for clinicians to partner with researchers to identify other preference sensitive decisions and to provide appropriate supports for these families. The research process used in this thesis research has the potential for use in other Audiology decisions. If clinicians identify that parents they are working with are having challenges in making decisions for their children, there is a generic Ottawa Personal Decision Guide (2008) available to help guide patients or their family members through difficult decisions.

Future Research

A variety of future research programs could be developed to build upon this thesis research. One study that could directly follow this thesis research is a large-scale evaluation of the decision aid (either randomized trial or pre-post study design with a larger sample). This could be undertaken in order to ascertain the effectiveness of the tool in reducing decisional conflict. A larger sample would also provide adequate power for evaluating the role of the decision aid in reducing decisional conflict. A large-scale study would also allow for the exploration of changes of knowledge and decisional conflict in subgroups of users. Subgroups could include parents with different baseline predisposition towards options (e.g. unsure versus leaning toward a specific option), education level (e.g. above or below the median numbers of years of education of father), knowledge (e.g. above and below the baseline median knowledge score), and parent (e.g. mother or father). This type of study may help to identify those individuals making BICI decisions that require special decisional support from clinicians.

A larger sample that was randomly assigned to either receive the decision aid or receive the current clinical practice would also allow for the identification of how the decision aid influences the surgical choices of parents (those choosing CI surgery vs. those who do not). A larger sample of parents would help to identify if the use of the decision aid influences parents' surgical options in the case of paediatric bilateral cochlear implantation.

It would also be interesting to examine how the decision aid influences parent expectations of outcomes. The current study examined knowledge and decisional conflict in parents. Parents' expectation of outcomes, and the influence of the decision aid on their

expectations, would be of interest to clinicians and parents. A randomized trial would be required to explore this.

Of additional interest is a research program that would follow children longitudinally after using a decision aid, and to examine how using a decision aid might have an impact on the children's and parents' future decision making. Parents examined in this thesis research had children ranging from one to five years of age. As these children grow, enter elementary, junior, and senior high schools it is certain that there are additional educational, rehabilitation, and amplification choices that the parents (and later children) must make. It would be important to identify if parents who have used a decision tool then apply the decision-making process to other decisions, such as seeking the information resources they need and considering their personal values when making future decisions for their children. Such research could identify the role that decision aids have on personal health practices and coping skills of the population.

It may also be important to explore some of the direct financial and non-financial costs associated with the unilateral cochlear implantation. Financial costs might include, but are not limited to, warranty and repair costs, while non-financial costs could include inclusion or exclusion from the Deaf community associated with the choice to pursue cochlear implantation.

In addition to the topic of paediatric bilateral cochlear implantation, it would also be fascinating to apply the ODSF to other relevant preference sensitive such as adult cochlear implantation, amplification choices (digital vs. programmable), and paediatric auditory rehabilitation options. Interest in developing a decision aid for adults making the CI decision has been indicated by some clinicians working with this population. In addition, there may

be some preference sensitive decisions in Speech Language Pathology and other related fields that could be addressed with the approach used in this project.

Summary

The individual studies conducted in this thesis work (as well as the thesis research as a whole) provides new contributions to the fields of Audiology and Population Health. This thesis research examined the decision-making of parents considering unilateral and bilateral cochlear implantation and identified the need for additional decisional support for some parents, particularly for bilateral cochlear implantation. The benefits and risks of bilateral implantation were systematically identified, documented, and estimated. A decision tool was designed to provide information about the options, their risks and benefits, as well as tools for parents to clarify and communicate the value they attribute to the various risks and benefits. The decision aid, that was acceptable to parents and clinicians, was piloted and improvements in parents' knowledge were noted. To summarize, this thesis work provides an example of evidence-based research that has identified a clinical gap and provided an intervention to improve the knowledge of parents in making decisions about BICIs for their children with significant hearing loss.

Statement of Contributions

Paper 1: An assessment of parents' decision-making regarding paediatric cochlear implants
Cyne Johnston was responsible for the ethics applications, updates, and final reports. She was also responsible for all data collection, analysis of qualitative and quantitative data, and original draft of paper. Transcriptions of the interviews were completed by a research assistant, Jennifer Vuong. Andrée Durieux-Smith, Annette O'Connor, Doug Angus, and Ian

Graham all provided support regarding research design. Andrée Durieux-Smith, Annette O'Connor, Doug Angus, Karen Benzies, and Elizabeth Fitzpatrick all provided feedback and suggestions for edits to drafts of the paper.

Paper 2: Bilateral paediatric cochlear implantation: A critical review

Cyne Johnston was responsible for executing the database searches, retrieving the articles, applying inclusion criteria, coding of the articles, and synthesizing the coded date. She was also responsible for the original draft of paper. A research assistant provided reliability data of the inclusion criteria. Andrée Durieux-Smith, Annette O'Connor, Doug Angus, and Ian Graham all provided support regarding review design. Andrée Durieux-Smith, Annette O'Connor, Doug Angus, Karen Benzies, and Elizabeth Fitzpatrick all provided feedback and suggestions for edits to drafts of the paper.

Paper 3: Estimation of risks associated with paediatric cochlear implantation

Cyne Johnston was responsible for the ethics applications, updates, and final reports. She was responsible for executing the database searches, retrieving the articles, applying inclusion criteria, coding of the articles, and synthesizing the coded date. She was also responsible for the original draft of paper. Andrée Durieux-Smith, Annette O'Connor, Doug Angus, and Ian Graham all provided support regarding study design. Andrée Durieux-Smith, Annette O'Connor, Doug Angus, Karen Benzies, Elizabeth Fitzpatrick and David Schramm all provided feedback and suggestions for edits to drafts of the paper.

Paper 4: The development and piloting of a decision aid for parents considering sequential bilateral cochlear implantation for their child with hearing loss

Cyne Johnston was responsible for the ethics applications, updates, and final reports. She was also responsible for all data collection, analysis of data, and original draft of paper. Andrée Durieux-Smith, Annette O'Connor, Doug Angus, and Ian Graham all provided support regarding research design. Andrée Durieux-Smith, Annette O'Connor, Doug Angus, Karen Benzies, and Elizabeth Fitzpatrick all provided feedback and suggestions for edits to drafts of the paper. Andrée Durieux-Smith, Annette O'Connor, Doug Angus, Karen Benzies, Elizabeth Fitzpatrick, Linda Moran, and Dr. David Schramm all provided feedback on the drafts of the decision aid as well.

Knowledge Translation

Traditional approaches to knowledge translation, such as publication and presentation of results at academic conferences have already been undertaken or will be completed over the next six months. Two of the papers (papers 1 and 2) have been published or accepted for publication. The fourth paper has been submitted and is awaiting decision regarding publication and the third paper will be submitted shortly. In January 2008, the needs assessment paper (Paper 1) was chosen by Canadian Association of Speech Language Pathologists and Audiologists (CASLPA) to include on their website as the "Monthly Featured Article" from the Winter 2008 Issue. The selection of this article ensures that the article is available in full to the media, related organizations and the general public from the CASLPA website. This selection offers an additional opportunity for knowledge translation to clinicians, parents, and the general public.

The research has been presented at the 2006 Canadian Academy of Audiology Conference as well as the CASLPA Conference in 2007. Additional conferences that are important for knowledge translation include those of CI clinicians, both audiologists and

surgeons as these are the clinicians most likely to be consistently included in the parental decision-making process. Audiologists are also the first individuals to offer information regarding bilateral cochlear implantation to parents. Ensuring awareness of the decision aid among these clinicians is essential. The CI International Conference may provide an important opportunity to reach these clinicians.

Clinician awareness of the decision aid is important, however it unlikely that awareness is adequate to change practice and increase use of the decision aid. In order to increase clinician use of the decision aid it may be important to identify any barriers to use and to find ways to address these barriers. This may include creating training sessions for interested clinicians. This may be an important knowledge translation strategy. Potential barriers may include but are not limited to a lack of knowledge and awareness of decision aids, attitudes toward decision aids, clinical procedures regarding which clinicians can provide decisional support, and a lack of skill in providing decisional support.

Inclusion of the decision aid among the catalogue of available decision aids will also be undertaken. The decision aid for sequential bilateral paediatric CI will be listed among other available decision aids for those seeking decisional support. Parents and clinicians will be able to retrieve the decision aid from the Internet, which is a key resource for parents. It may be worthwhile to seek support from various parent support groups in placing a link from their site to the online decision aid. This may help direct parents to this evidence-based resource.

References

American Academy of Pediatrics (2003). Infant hearing screening rates increase for America's number one birth defect. Retrieved November 12, 2005 from http://www.aap.org/advocacy/washing/July_8_2003rel.htm.

Berg, A.L., Ip, S.C., Hurst, M. & Herb, A. (2007). Cochlear implants in young children: informed consent as a process and current practices, *American Journal of Audiology,* 16, 13-28.

Brown, K.D. & Balkany, T.J. (2007). Benefits of bilateral cochlear implantation: a review. *Current Opinions in Otolaryngology, Head and Neck Surgery,* 15, 315-318.

Ching, T.Y., van Wanrooy, E. & Dillon, H. (2007). Binaural-bimodal fitting or bilateral implantation for managing severe to profound deafness: a review. *Trends in Amplification, 11,* 161-192.

Fagerlin, A., Ditto, P.H., Danks, J.H., Houts, R.M., & Smucker, W.D. (2001). Projection in surrogate decisions about life-sustaining medical treatments. *Health Psychology, 20,* 166-75.

Fitzpatrick, E., Johnston, J.C., Angus, D., & Durieux-Smith, A. (2006). Fitting audiology within the population health perspective. *Canadian Journal of Public Health, 97,* 153-155.

Galvin, K.L., Mok, M. & Dowell, R.C. (2007a). Perceptual benefit and functional outcomes for children using sequential bilateral cochlear implants, *Ear and Hearing, 28,* 470-82.

Galvin, K.L., Mok, M., Dowell, R.C. & Briggs, R.J. (2007b). 12-month post-operative results for older children using sequential bilateral implants, *Ear and Hearing, 28,* 19S-21S.

Grieco-Calub, T.M., Litovsky, R.Y. & Werner, L.A. (2008). Using the observer-based psychophysical procedure to assess localization acuity in toddlers who use bilateral cochlear implants, *Otology and Neurootology, 29,* 235-9.

Health Canada (2001). *The population health template: key elements and actions that define a population health approach.* Ottawa: Health Canada Population and Public Health Branch Strategic Policy Directorate.

Health Canada (2005). What is the population health approach? Retrieved September 7, 2005 from http://www.phac-aspc.gc.ca/ph-sp/phdd/approach/approach.html#health

Incesulu, A., Vural, M., & Erkam, U. (2003). Children with cochlear implants: Parental perspective. *Otology and Neurotology, 24,* 605-611.

Jacobson, M. J., & O'Connor, A. M. (2006). Population needs assessment: A workbook for assessing patients' and practioners' decision making needs. Retrieved July 18, 2008 from http://decisionaid.ohri.ca/docs/implement/Population_Needs.pdf

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. *Canadian Journal of Speech-Language Pathology and Audiology,* 32,169-82.

Li, Y., Bain, L., & Steinberg, A.G. (2004). Parental decision-making in considering cochlear implant technology for a deaf child. *International Journal of Pediatric Otolaryngology, 68,* 1027-1038.

Litovsky, R.Y., Johnstone, P.M., Godar, S., Agrawal, S., Parkinson, A. et al., (2006). Bilateral cochlear implants in children: localization acuity measured with minimum audible angle, *Ear and Hearing, 27,* 43-59.

Litovsky, R.Y., Parkinson, A., Arcaroli, J., Peters, R., Lake, J., et al. (2004). Bilateral cochlear implants in adults and children, *Archives of Otolaryngology Head and Neck Surgery, 130,* 648-55.

Most, T., & Zaidman-Zait, A. (2003). The needs of parents of children with cochlear implants. *Volta Review, 103,* 99-113.

Mohr, E., Feldman, J., Dunbar, J.L., McConkey-Robbins, A., Niparko, J.K., Rittenhouse, R.K., & Skinner, M.W. (2000). The societal costs of severe to profound hearing loss in the United States. *International Journal of Technology Assessment in Health Care, 16,* 1120-1135.

Murphy, J., & O'Donoghue, G. (2007). Bilateral cochlear implantation: an evidence-based medicine evaluation. *Laryngoscope, 117,* 1412-1418.

O'Connor, A.M., Stacey, D., Entwistle, V., Llewellyn-Thomas, H., Rovner, D., Holmes-Rovner, M., et al. (2002). Decision aids for people facing health treatment or screening decisions (Cochrane Review). Retrieved on July 18, 2008 from http://www.cochrane.org/reviews/en/ab001431.html

O'Connor, A. M., Tugwell, P., Wells, G. A., Elmslie, T., Jolly, E., Hollingworth, G., et al. (1998). A decision aid for women considering hormone therapy after menopause: Decision support framework and evaluation. *Patient Education and Counseling, 33,* 267-279.

Ottawa Personal Decision Guide (2008). Retrieved on January 17, 2009 online at http://decisionaid.ohri.ca/decguide.html.

Perreau, A.E., Tyler, R. S., Witt, S., & Dunn, C. (2007). Selection strategies for binaural and

monaural cochlear implantation. *American Journal of Audiology, 16*, 85-93.

Peters, E. (2000). Our decision on a cochlear implant. *American Annals of the Deaf, 145*, 263-267.

Sach, T. H., & Whynes, D. K. (2005). Paediatric cochlear implantation: The views of parents. *International Journal of Audiology, 44,* 400-407.

Samson-Fang, L., Simons-McCandless, M., & Shelton, C. (2000). Controversies in the field of hearing impairment: early intervention, educational methods, and cochlear implants. *Infants and Young Children, 12*, 77-88.

Sorkin, D. L., & Zwolan, T. A. (2008). Parental perspectives regarding early intervention and its role in cochlear implantation in children. *Otology and Neurootology, 29*, 137-141.

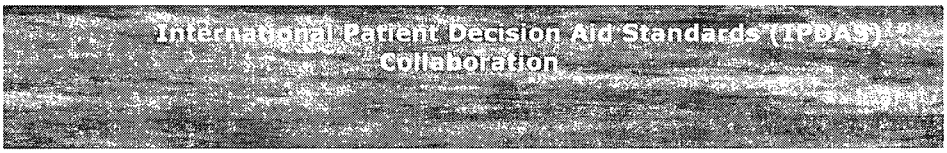
Steinberg, A., Brainsky, A., Bain, L., Montoya, L., Indenbaum, M., et al. (2000). Parental values in the decision about cochlear implantation. *Int J Pediatr Otorhinolaryngol, 55,* 99-107.

Tait, A.R., Voepel-Lewis, T., Munro, H.M., & Malviya, S. (2001). Parent' preferences for participation in decisions made regarding their child's anaesthetic care. *Paediatric Anasthesia, 11*, 283-90.

Tashakkori, A., & Teddlie, C. (2003). *Handbook of mixed methods in social and behavioral research.* Thousand Oaks, CA: Sage.

William House Cochlear Implant Study Group. (2008). Position statement on bilateral cochlear implantation. *Otology and Neurootology*, *29*, 107-108.

Appendix A: International Patient Decision Aid Standards



IPDAS 2005: Criteria for Judging the Quality of Patient Decision Aids

Steering Committee: A O'Connor (CA) & G Elwyn (UK) (co-leaders) with A Barratt (AU), M Barry (US), A Coulter (UK), M Holmes-Rovner (US), N Moumjid (FR), H Llewellyn-Thomas (US), M O'Kane (US), R Thomson (UK), D Stacey (CA), T Whelan (CA) **Methods Group:** G Elwyn (leader, UK) with S Bernstein (US), P Shekelle (US), R Thomson (UK), R Volk (US) **Stakeholder Leader:** A Coulter (UK) **Quality Criteria Panels:** A O'Connor (CA) & H Llewellyn-Thomas (US) (editors) with J Austoker (UK), A Barratt (AU), M Barry (US), H Bekker (UK), J Belkora (US), C Braddock (US), P Butow (AU), E Chan (US), A Charvet (Switz), A Clarke (UK), J Davison (CA), J Dolan (US), A Edwards (UK), V Entwistle (UK), A Fagerlin (US), D Feldman-Stewart (CA), J Fowler (US), D Frosch (US), P Hewitson (UK), M Holmes-Rovner (US), T Hope (UK), MJ Jacobsen (CA), A Kennedy (Switz), S Knight (US), M Kupperman(US), B Ling (US), T Marteau (UK), K McCaffery (AU), N Moumjid (FR), A Mulley (US), M O'Connor (US), E Ozanne (US), M Pignone (US), A Raffle (UK), C Ruland (NO), L Schwartz (US), K Sepucha (US), S Sheridan (US), S Stableford (US), D Stacey (CA), D Stilwell (US), V Tait (CA), D Timmermans (NL), L Trevena (AU), T Whelan (CA), C Wills (US), S Woloshin (US), S Ziebland (UK)

What are patient decision aids and why are they needed?
Patient decision aids are tools to help people participate in their health decisions in ways they prefer. They are used when there is more than one medically reasonable option to diagnose or treat a health problem. Each of the options has good and bad features that people value differently. Even when two people are in the same situation, what is important for one person may be different for another person. Therefore, there is no clear answer that applies to everyone. The best choice involves matching which features matter most to a person with the option that has these features. To make a good decision, you need an expert on the facts (e.g. a health practitioner) and an expert on which features matter most (e.g. the patient) and a way to share their views with each other in ways they prefer.

Patient decision aids aim to do three things to prepare a person for decision making. They provide facts about a person's condition, the options and their features. They help people to clarify their values (the features that matter most to them). They help people to share their values with their health care practitioner and others, so a course of action can be planned that matches their values. Patient decision aids do not advise people to choose one option over another. They do not replace counseling from a health care practitioner. Instead, they prepare people to discuss the options with their health care practitioner.

An international group of researchers, known as the 'Cochrane Review Team of Patient Decision Aids' is compiling decision aids and summarizing the results of research trials. The latest review of 34 studies shows that patients and practitioners who use patient decision aids make better decisions. Patients participate more, know more, and have more realistic expectations of what might happen. They are more likely to receive an option with features they most value (O'Connor et al., *Cochrane Library*, 2003).

The **International Patient Decision Aid Standards (IPDAS)** Collaboration is a group of researchers, practitioners and stakeholders from around the world. The goal is to establish an internationally approved set of criteria to determine the quality of patient decision aids. These criteria will be helpful to a wide variety of individuals and organizations that use and/or develop patient decision aids.

Why are standards needed?
There are over 500 patient decision aids available or being developed by many different individuals and groups around the world. However, people have difficulty knowing whether or not a decision aid is a source of reliable health information that can help in decision making.

How were the standards obtained?

There was a 2-stage evidence-informed Delphi consensus process

1. Participants included 122 people from 14 countries and 4 stakeholder groups [researchers/developers; health professionals/ patient/consumers; policy makers/health plan administrators]

2. A voting document was developed from a series of background papers on 12 quality domains. [The experts who wrote these papers are listed above]. Before voting on the importance of each criterion in judging the quality of a patient decision aid, voters reviewed: definition of decision aids; definition of criterion; theoretical link between criterion and decision quality; and empirical evidence supporting or not supporting its use in decision aids. Evidence was derived from fundamental studies and a Cochrane Collaboration systematic review of randomized trials of patient decision aids.

The standards are summarized in a users' checklist on the next page.

I. Content: Does the patient decision aid ...

Provide information about options in sufficient detail for decision making?

- ☐ describe the health condition
- ☐ list the options [including doing nothing]
- ☐ describe the natural course without options
- ☐ describe positive features [benefits]
- ☐ describe negative features of options [harms / side effects / disadvantages]
- ☐ include chances of positive / negative outcomes
- ☐ describe procedures

Additional items for tests

- ☐ describe what test is designed to measure
- ☐ include chances of true positive, true negative, false positive, false negative test results
- ☐ include chances the disease is found with / without screening
- ☐ describe possible next steps based on test result
- ☐ describe detection / treatment that would never have caused problems if one was not screened

Present probabilities of outcomes in an unbiased and understandable way?

- ☐ use event rates specifying the population and time period
- ☐ compare outcome probabilities using the same denominator, time period, scale
- ☐ is tailored to patient [e.g. age]
- ☐ use visual diagrams
- ☐ use multiple methods [words, numbers, diagrams] to view changes
- ☐ use both positive and negative frames
- ☐ describe uncertainty around probabilities
- ☐ place probabilities in context of other events

Include methods for clarifying and expressing patients' values?

- ☐ describe the procedures and outcomes to help patients imagine what it is like to experience their physical, emotional, social effects
- ☐ ask patients to consider which positive and negative features matter most
- ☐ suggest ways for patients to share what matters most with others

Include structured guidance in deliberation and communication?

- ☐ provide steps to make a decision
- ☐ include tools [worksheet, question list] to discuss options with others

II. Effectiveness: Does the patient decision aid ensure decision making is informed and values based?

Decision processes leading to decision quality. The patient decision aid helps patients to:

- ☐ recognise a decision needs to be made
- ☐ know options and their features
- ☐ be clear about option features that matter most
- ☐ understand that values affect decision
- ☐ discuss values with their practitioner
- ☐ become involved in preferred ways

Decision quality

- ☐ there is a match between the chosen option and the features that matter most to the informed patient

III. Development Process: Does the patient decision aid ...

Present information in a balanced manner?

- ☐ able to compare positive / negative features of options
- ☐ shows negative / positive features with equal detail [fonts, order, display of statistics]

Have a systematic development process?

- ☐ finds out what users [patients, practitioners] need to discuss options
- ☐ has peer review by patient / professional experts not involved in development and field testing
- ☐ includes developers' credentials / qualifications

Field tests with users show the patient decision aid is:

- ☐ acceptable
- ☐ balanced for undecided patients
- ☐ understood by those with limited reading skills

Use up to date scientific evidence that is cited in a reference section or technical document?

- ☐ report steps to summarise evidence
- ☐ describe quality of scientific evidence [including lack of evidence]
- ☐ provides references to evidence used
- ☐ report how patient decision aid is updated
- ☐ report date of last update
- ☐ uses evidence from studies of patients similar to those of target audience

Disclose conflicts of interest?

- ☐ report source of funding to develop and distribute the patient decision aid
- ☐ report whether authors or their affiliations stand to gain or lose by choices patients make after using the patient decision aid

Use plain language?

- ☐ is written at a level that can be understood by the majority of patients in the target group
- ☐ is written at a grade 8 equivalent level or less according to readability score [SMOG or FRY]
- ☐ provides ways to help patients understand information other than reading [audio, video, in-person discussion]

Appendix B: Cochlear Implant Needs Assessment Semi-Structured Interview Guide

Cochlear Implant Needs Assessment Semi-Structured Interview Guide

1) Can you tell me about the first few days and weeks after you found out about your child's hearing loss? What were the most important decisions that you faced early on?

2) Let's focus on the cochlear implant decision. How were you introduced to the topic of cochlear implants?

3) Can you describe your experience in making the decision to implant or not?

4) How did you feel when you had to make this decision? Were you: ☐ unsure about what to do
☐ worried what could go wrong
☐ distressed or upset
☐ constantly thinking about the decision
☐ wavering between choices or changing your mind
☐ delaying the decision
☐ questioning what is important to you
☐ feeling physically stressed–tense muscles, racing heartbeat, difficulty sleeping

5) What made this decision difficult to make? Were you?
☐ lacking information about options, pros and cons
☐ lacking information on the of benefits and harms
☐ unclear about what is important to you
☐ lacking information on what others decide
☐ feeling pressure from others
☐ lacking support from others
☐ not feeling ready to make a decision
☐ lacking the ability to make this type of decision

6) Thinking about the cochlear implant decision, which options were there for your family?

7) What do you see as the main advantages and disadvantages of these options?

8) Who was most involved in helping you make this decision?

9) Thinking about the clinic staff you encountered, how were they usually involved in making this decision? Did they:
☐ make the decision for you,
☐ share the decision with you,
☐ providing support or advice for you to make the decision on your own

10) How did you go about making such a decision? Did you:
☐ get information on choices
☐ get information on how likely the choices are
☐ consider how important choices are,
☐ get information on how others decide
☐ find ways to handle pressure
☐ get support from others

11) What helped you to make this decision?

12) What gets in the way of making this decision?

13) What else is needed?

14) I will list possible ways to help people with decisions, which ones do you think may be useful for you?
☐ Counseling from health practitioner
☐ Discussion groups of people facing the same decisions,

⌊_⌋ Information materials If yes, type of medium---->
⌊_⌋ booklets, pamphlets ⌊_⌋ videos ⌊_⌋ CD ROMS
⌊_⌋ Internet ⌊_⌋ other, specify _____

15) Who do you think should prepare information about this decision?
⌊_⌋ health societies
⌊_⌋ expert medical and health practitioners
⌊_⌋ government
⌊_⌋ consumer associations
⌊_⌋ cochlear implant companies

16) Bilateral cochlear implants are now emerging as an option for children with bilateral hearing loss. Is there anything you would like to add about this issue?

Appendix C: Summary of complication rates from the identified literature and comparison with local retrospective chart review

Minor Complications Not Requiring Hospitalization								
Authors	Definition	Study Design	Sample Size	Time Frame	Population Study Age Sequential/Simultaneous	Event Rate		Rate out of 100
						#	base	
Stratigoulea s, Perry, King, & Syms (2006)	-Managed conservatively on an ambulatory basis -Flap infections, stitch abscess, transient tinnitus and dizziness, otitis externa	Retrospective cohort	N=176	Not reported Follow-up mean 289 days	Adults and Children 1 to 86 years (Mean 27yr) Unilateral CI	12	176	6.82
Cunningham, Slattery & Luxford (2004)	- Postoperative infections including wound infections, complicated otitis media, or meningitis -Minor if requiring local wound care or oral antibiotics	Retrospective cohort	N=272	Implants between 1993 and 2002	Children 1 to 10 years (Mean 4 yr) Unilateral CI	6	272	2.21
Migrov, Muchnik, Kaplan-Neeman & Kronenberg (2006)	-Minor complications defined as: wound infections, stitch granuloma (vestibular problems, perforated eardrum not included)	Retrospective cohort	N=300	Implants between 1993 and 2005	Children 2 to 16 years Unilateral CI	53	300	17.67
Gysin, Papsin, Daya & Nedzelski (2000)	- Minor complications; Flap cellulites, perilymph leakage	Retrospective cohort	N=100	Implants between 1990 and 1998	Children 1 to 17 years (Mean= 6 yr) Unilateral CI	2	100	2.00
Arnoldner, Baumgartner, Gstoettner & Hamzavi (2005)	- Wound complications, keloid scar formation, cholesteatoma	Retrospective cohort	N=128	Implants between 1994 and 2003	Children 0.75 to 14 years (Mean= 5 yr) Unilateral CI	2	128	1.56
James & Papsin (2004)	- surgical complications	Retrospective cohort	N=25	Implants between 2000 and 2003	Children 0.5 to 1 year Unilateral CI	0	25	0
Black, Bailey, Albert,	- wound infections, otorrhoea, scar tissue, etc	Retrospective cohort	N=242	Implants between 1992 and 2004	Children 1 to 17 year s (Mean=5 yr)	44	221	19.94

Leighton, Hartley, Chatrath & Patel (2007)					Unilateral CI			
Kandogan, Levent & Gurol (2005)	- minor complications that settled with conservative treatment, local care or medication	Retrospective cohort	N=227	Implants between 1998 and 2004	Children 1 to 16 years Unilateral CI	15	227	6.60
Bhatia, Gibbin, Nikolopoulos & O'Donoghue (2004)	- swelling, hemotoma, infection settled with conservative treatment	Prospective cohort	N=300	Follow-up time 0.1-14 yrs (mean 4 years)	Children 1 to 16 years Unilateral CI	46	300	15.33
Postelmans, Cleffken & Stokroos (2006)	- mild infection	Retrospective cohort	N=32	Implants between 2000 and 2005	Children 1 to 11 years (mean=4 yr) Unilateral CI	2	32	18.75
Venail, Sicard, Piron et al. (2007)	- Skin flap problems, haematoma, infection	Retrospective cohort	N=272	Implants between 1989 and 2005	Children .9 to 15 years (mean=5 yr) Unilateral CI	7	272	2.57
Total						189	2053	9.21

Complications requiring Hospitalization or Additional Surgery

Authors	Definition	Study Design	Sample Size	Time Frame	Population Study Age Sequential/Simultaneous	Event Rate		Rate out of 100
						#	base	
Stratigoulleas, Perry, King, & Syms (2006)	-Required hospitalization and doesn't fall into other category -Mastoiditis, wound infection, migration	Retrospective cohort	N=176	Not reported Follow-up mean 289 days	Adults and Children 1 to 86 years (Mean 27) Unilateral CI	3	176	1.70

Yu, Hegarty, Gantz & Lalwani (2001)	-Post Operative Infections requiring antibiotics, or drainage, hospitalization	Retrospective cohort	N=108	Implants between 1991 and 2000	Adults and Children Not Reported Unilateral CI	4	108	3.70
Cunningham, Slattery & Luxford (2004)	- Postoperative infections including wound infections, complicated otitis media or mastoiditis -Major if requiring hospitalization, explantation, IV antibiotics	Retrospective cohort	N=272	Implants between 1993 and 2002	Children 1 to 10 years (Mean 4 yr) Unilateral CI	10	272	3.68
Migrov, Muchnik, Kaplan-Neeman & Kronenberg (2006)	-mastoiditis requiring surgery, foreign body reaction, wound breakdown, magnet displacement, stitch granuloma, etc.	Retrospective cohort	N=300	Implants between 1993 and 2005	Children 2 to 16 years Unilateral CI	23	300	7.66
Gysin, Papsin, Daya & Nedzelski (2000)	- Major complications; flap breakdown, perilymph gusher, cholesteatoma	Retrospective cohort	N=100	Implants between 1990 and 1998	Children 1 to 17 years (Mean= 6 yr) Unilateral CI	4	100	4.00
James & Papsin (2004)	- surgical complications	Retrospective cohort	N=25	Implants between 2000 and 2003	Children 0.5 to 1 year Unilateral CI	0	25	0
Black, Bailey, Albert, Leighton, Hartley, Chatrath & Patel (2007)	- mastoiditis, flap breakdown, perilymph gusher, cholesteatoma, failed insertion, electrode migration,	Retrospective cohort	N=221	Implants between 1992 and 2004	Children 1 to 17 year s (Mean=5 yr) Unilateral CI	13	221	5.88
Kandogan, Levent & Gurol (2005)	- CSF gusher	Retrospective cohort	N=227	Implants between 1998 and 2004	Children 1 to 16 years Unilateral CI	5	227	2.20

Bhatia, Gibbin, Nikolopoulos & O'Donoghue (2004)	- Complications requiring surgical intervention or a permanent resulting in disability. Including cholesteatoma, electrode exposure	Prospective cohort	N=300	Follow-up time 0.1-14 yrs (mean 4 years)	Children 1 to 16 years Unilateral CI	7	300	2.33
Arnoldner, Baumgartner, Gstoettner & Hamzavi (2005)	- major complications	Retrospective cohort	N=128	Implants between 1994 and 2003	Children 0.75 to 14 years (Mean= 5 yr) Unilateral CI	0	128	0
Postelmans, Cleffken & Stokroos (2006)	- abscess formation near site	Retrospective cohort	N=32	Implants between 2000 and 2005	Children 1 to 11 years (mean=4 yr) Unilateral CI	1	32	3.13
Venail, Sicard, Piron et al. (2007)	- electrode shifting, receiver displacement,	Retrospective cohort	N=272	Implants between 1989 and 2005	Children .9 to 15 years (mean=5 yr) Unilateral CI	3	272	1.10
Total						73	2161	3.38

Facial Palsy or Facial Nerve Paralysis

Authors	Definition	Study Design	Sample Size	Time Frame	Population Study Age Sequential/Simultaneous	Event Rate # temporary (# permanent)	Event Rate base	Rate out of 100
Fayad, Wanna, Michelet to & Parisier (2003)	- Facial nerve weakness or paralysis occurring following CI surgery -Complication occurred between 18 hrs and 19 days post-implant	Retrospective cohort	N=705	Implants between 1980 and 2002	Adults and Children Not Reported Unilateral CI	5 (0)	705	0.71 (0)

Stratigouleas, Perry, King, & Syms (2006)	-Facial nerve weakness or paralysis occurring following CI surgery	Retrospective cohort	N=176	Not reported Follow-up mean 289 days	Adults and Children 1 to 86 years (Mean 27) Unilateral CI	4 (1)	176	2.27 (0.57)
Migrov, Muchnik, Kaplan-Neeman & Kronenberg (2006)	-Facial nerve paralysis	Retrospective cohort	N=300	Implants between 1993 and 2005	Children 2 to 16 years Unilateral CI	2 (0)	300	0.66 (0)
Gysin, Papsin, Daya & Nedzelski (2000)	- Facial weakness	Retrospective cohort	N=100	Implants between 1990 and 1998	Children 1 to 17 years (Mean= 6 yr) Unilateral CI	1 (0)	100	1 (0)
Arnoldner, Baumgartner, Gstoettner & Hamzavi (2005)	- Transient facial palsy	Retrospective cohort	N=128 (children)	Implants between 1994 and 2003	Children 0.75 to 14 years (Mean= 5 yr) Unilateral CI	0 (0)	128	0 (0)
Black, Bailey, Albert, Leighton, Hartley, Chatrath & Patel (2007)	- facial nerve palsy	Retrospective cohort	N=242	Implants between 1992 and 2004	Children 1 to 17 years (Mean= 5 yr) Unilateral CI	3 (0)	242	1.24 (0)
Kandogan, Levent & Gurol (2005)	- Facial Nerve Paresis	Retrospective cohort	N=227	Implants between 1998 and 2004	Children 1 to 16 years Unilateral CI	1 (0)	227	0.44 (0)
Bhatia, Gibbin,	- Temporary facial weakness	Prospective cohort	N=300	Follow-up time 0.1-14 yrs	Children	2 (0)	300	0.66 (0)

Nikolopoulos & O'Donoghue (2004)				(mean 4 years)	1 to 16 years Unilateral CI			
Venail, Sicard, Piron et al. (2007)	- Facial Stimulation	Retrospective cohort	N=272	Implants between 1989 and 2005	Children 0.9 to 15 years (Mean= 5 yr) Unilateral CI	1 (2)	272	0.36 (0.74)
Total						17 (3)	2450	0.69, [1] (0.12, [0.1])

Bacterial Meningitis

Authors	Definition	Study Design	Sample Size	Time Frame	Population Study Age Sequential/Simultaneous	Event Rate		CI Rate out of 100
						#	base	
Reefhuis, Honein, Whitney, Chamany, Mann, et al. (2003)	- Bacterial Meningitis	Retrospective Cohort and nested case control study	N=4262 (children)	Implants between 1997 and 2002	Children Less than 6 with CI Unilateral CI	26	4264 (10852 PY)	0.240/100 PY
Summerfield, Cirstea, Roberts, et al. (2004)	- Bacterial Meningitis	Retrospective cohort	N=1851	Implants between 1997 and 2002	Children 1 to 16 years Unilateral CI	0	1851 (2589 PY)	0
Total						26	13441 PY	0.193 (0.1)

Device Failure- Overall

	Definition	Study Design	Sample Size	Time Frame	Population Study Age	Event Rate	

Authors					Sequential/Simultaneous	#	base	CI Rate out of 100
Stratigouleas, Perry, King, & Syms (2006)	-Device failure including intermittent locking	Retrospective cohort	N=176	Not reported Follow-up mean 289 days	Adults and Children 1 to 86 years (Mean 27) Unilateral CI	3	176	1.70
Parisier, Chute, Popp & Suh (2001)	-Device failures	Retrospective cohort	N=403	Implants between 1990 and 1999 Follow-up over 6 months	Children 2 to 10 years Unilateral CI	27	403	6.70
Weise, Muller-Deile, Brademann, Meyer, Ambrosch & Maune (2005)	-Device failures by traumatic failure, failed electrode insertion, and infection	Retrospective cohort	N=110	Implants between 1990 and 2003 Follow-up over 6 months	Children 2 to 10 years Unilateral CI	6	110	5.45
Gysin, Papsin, Daya & Nedzelski (2000)	- Device failures	Retrospective cohort	N=100	Implants between 1990 and 1998	Children 1 to 17 years (Mean= 6 yr) Unilateral CI	2	100	2
Arnoldner, Baumgartner, Gstoettner & Hamzavi (2005)	- Device failures	Retrospective cohort	N=128 (children)	Implants between 1994 and 2003	Children 0.75 to 14 years (Mean= 5 yr) Unilateral CI	22	128	17.19
Kandogan, Levent & Gurol (2005)	- Trauma to the device	Retrospective cohort	N=227	Implants between 1998 and 2004	Children 1 to 16 years Unilateral CI	22	227	9.69

Battmer, O'Donoghue, & Lenarz (2007)	- Device Failure	Retrospective review	N=12856	Not Reported	Adults and Children Not Reported All CI Devices	488	12856	3.79
Postelmans, Cleffken & Stokroos (2006)	- Device Failure	Retrospective cohort	N=32	Implants between 2000 and 2005	Children 1 to 11 years (mean=4 yr) Unilateral CI	2	32	6.26
Total						550	14032	3.92

Device Failure- 5- year failure rate

Authors	Definition	Study Design	Sample Size	Time Frame	Population Study Age Sequential/Simultaneous	Event Rate #	Event Rate base	CI Rate out of 100
Venail, Sicard, Piron et al. (2007)	- Device Failure and soft failures	Retrospective cohort	N=272	Implants between 1989 and 2005	Children .9 to 15 years (mean=5 yr) Unilateral CI	17	272	6.25
Maurer, Marangos & Ziegler (2005)	-Hard failures of cochlear implants due to design errors and direct or indirect trauma to the CI site	Retrospective cohort	N=134 (children)	Implants between 1990 and 2001 Follow-up over 2 years	Children Not Reported Unilateral CI	14	192	7.3
Total						31	464	6.68

Appendix D: Research Ethic Board Certificates

Research Ethics Board: Children's Hospital of Eastern Ontario- Studies 1 and 3

Research Ethics Board: University of Ottawa- Studies 1 and 3

Research Ethics Board: Children's Hospital of Eastern Ontario- Study 4

Research Ethics Board: University of Ottawa- Study 4

Children's Hospital of Eastern Ontario
Centre hospitalier pour enfants de l'est de l'Ontario

CHEO RESEARCH ETHICS BOARD APPROVAL – EXPEDITED REVIEW

Principal Investigator:	Ms. Linda Moran/Ms. Cyne Johnston
Proposal Number:	#06/23X
Protocol Title:	The development and exploration of the use of a decision aid for parents making the cochlear implant decision for their child with hearing loss
Department or PSU:	Audiology
Approval date:	June 12, 2006
Valid Until:	June 11, 2007
Documents reviewed and approved:	Research protocol submitted April 19, 2006

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 under the expedited stream, which is reserved for projects that involve no more than minimal risk to human subjects.

During the course of the research, no deviations from, or changes to, the protocol or consent form may be initiated without prior written approval from the REB. Further, investigators are asked to report the following to the REB:

- Proposed changes to the study procedures (including the recruitment strategy, inclusion criteria, etc.);
- Concerns or issues that arise in conducting the research;
- Changes to the consent documents and advertisement notices;
- Changes to the investigators who assume responsibility for the study; &
- An annual report.

Wishing you success in your project.

Chair, Research Ethics Board

CG/smeh 08/06/06
c.c. Pat Brazeau, Manager, CHEO RI
Joanne Whittingham, Research Associate

This is an official document. Please retain the original for your file *version 11/2003*

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Université d'Ottawa University of Ottawa
Service de subventions de recherche et déontologie Research Grants and Ethics Services

August 30, 2006

Andrée Durieux-Smith
School of Rehabilitation Sciences
University of Ottawa
451 Smyth, room 3028F
Ottawa, ON K1H 8M5

Doug Angus
School of Management
University of Ottawa
136 Jean-Jacques Lussier, room 341
Ottawa, ON K1N 6N5

Jennifer Cyne Johnston
47 Glendale Ave
Ottawa, ON K1S 1WS

Object: The Development and Exploration of the Influence of a Decision Aid for Decision-Making among Parents Choosing Cochlear Implantation for their Children with Hearing Loss – Phase 1 (file H 06-06-11)

Dear Professors Durieux-Smith and Angus and Ms. Johnston;

You will find enclosed the Health Sciences and Science REB ethical clearance for the abovementioned study.

During the course of the study, any modifications to the protocol or forms may not be initiated without prior written approval from the REB. You must also promptly notify the REB of any adverse events that may occur.

This certificate of ethical clearance is valid until August 30, 2007. Please submit an annual status report to the Protocol Officer in August 2007 to either close the file or request a renewal of ethics approval. This document can be found at:
http://web9.uottawa.ca/services/rgessrd/ethics/application_dwn.asp

A copy of this approval will be sent to research services, if necessary.
If you have any questions, you may contact the undersigned at the number 562-5387.

Protocol Officer for Ethics in Research
For Dr. Daniel Lagarec, Chair of the Health Sciences and Science REB

550, rue Cumberland 550 Cumberland Street
Ottawa (Ontario) K1N 6N5 Canada Ottawa, Ontario K1N 6N5 Canada

(613) 562-5841 • Télec. Fax (613) 562-5338
http://www.uottawa.ca/services/research/rge/index.html

Université d'Ottawa University of Ottawa

Service de subventions de recherche et déontologie Research Grants and Ethics Services

HEALTH SCIENCES AND SCIENCE RESEARCH ETHICS BOARD

CERTIFICATE OF ETHICAL APPROVAL

This is to certify that the University of Ottawa Health Sciences and Science Research Ethics Board has examined the application for ethical approval of the research project entitled **The Development and Exploration of the Influence of a Decision Aid for Decision-Making among Parents Choosing Cochlear Implantation for their Children with Hearing Loss – Phase 1 (file H 06-06-11)** submitted by Jennifer Cyne Johnston and supervised by Andrée Durieux-Smith of the School of Rehabilitation Sciences and Doug Angus of the School of Management. The Board found that this research project met appropriate ethical standards as outlined in the Tri-Council Policy Statement and in the Procedures of the University of Ottawa Research Ethics Boards, and accordingly gave it a Category 1a (approval). This certification is valid one year from the date indicated below.

August 30, 2006
Date

Protocol Officer for Ethics in Research
For Dr. Daniel Lagarec, Chair of the
Health Sciences and Science REB

Children's Hospital of Eastern Ontario
Centre hospitalier pour enfants de l'est de l'Ontario

CHEO Research Ethics Board Approval – Expedited Review

Principal Investigator:	Ms. Linda Moran
Proposal Number:	#08/57X
Protocol Title:	The exploration of the use of a decision aid for parents making the bilateral cochlear implant decision for their child with hearing loss.
Department or PSU:	Audiology
Approval date:	August 14, 2008
Valid Until:	August 13, 2009
Documents reviewed and approved:	Research protocol submitted June 4, 2008

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 under the expedited stream, which is reserved for projects that involve no more than minimal risk to human subjects.

During the course of the research, no deviations from, or changes to, the protocol or consent form may be initiated without prior written approval from the REB. Further, investigators are asked to report the following to the REB:

- Proposed changes to the study procedures (including the recruitment strategy, inclusion criteria, etc.);
- Concerns or issues that arise in conducting the research;
- Changes to the consent documents and advertisement notices;
- Changes to the investigators who assume responsibility for the study; &
- An annual report.

Wishing you success in your project.

Regards,

CG/smeh 14/08/08
c.c. Pat Brazeau, Manager, CHEO RI
Cyne Johnston, Research assistant
Joanne Whittingham, Audiology Research

This is an official document. Please retain the original for your file *version 11/2003*

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Université d'Ottawa University of Ottawa
Service de subventions de recherche et déontologie Research Grants and Ethics Services

October 20, 2010

Ms. Cyne Johnston
Children's Hospital of Eastern Ontario
Research Institute
Hearing Research Lab
cyneandmark@shaw.ca

Ms. Linda Moran
Professional Practice Leader
CHEO Audiology
moran@cheo.on.ca

Dear Ms. Johnston and Ms. Moran,

re: "The Exploration of the Use of Decision Aid for Parents Making the Bilateral Cochlear Implant Decision for their Child with Hearing Loss" – File no. H08-08-15

Thank you for the information relating to the CHEO REB's approval of the above-referenced project.

This is to confirm that, in accordance with the agreement between Children's' Hospital of Eastern Ontario (CHEO) and the University of Ottawa, the University of Ottawa recognizes the approval of the CHEO Research Ethics Board (REB) for the above-referenced project and has authorized the CHEO REB to act as Board of Record for the review and oversight of this project.

Copies of annual reports and renewals of CHEO Hospital REB approvals must be provided to our office. 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 University Research on demand of the CHEO REB.

If you have any questions, you may contact the ethics office at 562-5841.

If you have any questions, please do not hesitate to contact me at extension 5841.

Sincerely yours,

Director, Research Grants and Ethics
For Daniel Lagarec, Chair of the HSS REB

cc: Dr. Carole Gentile (Chaire, CHEO REB)
Dr. Daniel Lagarec

Appendix E: Informed Consent Forms

Parent Consent Form - Study 1

Clinician Consent Form - Study 1

Parent Consent Form - Study 4

Clinician Consent Form - Study 4

A Needs Assessment for the Development of a Decision Aid to Help Parents of Children with Hearing Loss Decide Between Cochlear Implantation and Continued Hearing Aid Use

We would like to invite you to take part in a research study. This study examines how best to support parents making a decision about cochlear implants for their child. Researchers at the University of Ottawa and CHEO are conducting the study. Cyne Johnston is the University of Ottawa doctoral student undertaking this research. This study will take place in English.

The diagnosis of a hearing loss can be a time of great stress for parents. Many decisions have to be made by these parents. These include the communication approaches they will use and hearing aid options. Parents of children with severe or profound hearing loss may also have to make decisions about cochlear implant use.

Purpose of this study

- To understand the experiences you had in making the cochlear implant decision.
- To explore the possibility of developing a decision aid to help parents of children with significant hearing losses choose an amplification option for their child.

If you choose not to participate in the study, this will in no way affect the care you and your family members receive at CHEO.

Procedure
1) In this study we will be asking you to allow a researcher to interview you about how you made the cochlear implant decision. The researcher will ask you questions about your feelings that you had as you made this decision. They will also ask about the type of information that helped you with decision making. They are interested in knowing about the people who helped you make this decision. The interview will last between 30 and 45 minutes. This interview will be tape recorded.

All information obtained in the interview will be kept confidential. Any tapes, transcripts, or questionnaires from the study will be kept in the research office for five years after the interviews. Only members of the research team will have access to the study information. The information gathered will be summarized and individual answers will not be identified. The results of this study may be used in research papers, conference, or course presentations. Quotes from individuals may be used but no identifying information will accompany them.

Subject description
Parents of 10 children with hearing loss who had to make the cochlear implant decision will be asked to take part in this research.

Are there any risks to participating in this research?
The interviews and questionnaires can be completed at your convenience. They can be conducted at your home or on one of your visits to the audiology department. It will take you about an hour to participate in this study. There are no physical risks associated with this study.

Benefits
This study will help us to identify what parents need to make the cochlear implant decision. It will help us to develop a tool to help other parents with the decision.

Withdrawing from the study
If you choose to withdraw from the study, this will in no way affect the care you and your family members receive. At any time during the study you may decline to answer a question, withdraw

your answer, or stop the interview. This can be done by indicating your choice to withdraw from the study to the interviewer. Any data already gathered will be destroyed if you withdraw from the study.

I acknowledge that the research procedures described above, and of which I have a copy, have been explained to me. I have been given an opportunity to ask questions concerning the study and the interview process and the questions that I have asked have been adequately answered. In addition, I know that I may contact any of the investigators (whose contact information is below) if I have further questions either now or in the future.

I have been assured that personal records relating to this study will be kept confidential except as required or permitted by law. I understand that I am free to withdraw from the study at any time. I understand that I can withdraw my consent and stop my participation in the study at any time and for any reason. I have been reassured that this will not affect the quality of care that I or any member of my family receives at CHEO. I understand that if any knowledge gained from this study becomes available that could influence my decision to continue in this study, I will be promptly informed.

The Research Ethics Board is a group of people from scientific and non-scientific backgrounds that review research studies. Their goal is to ensure the protection of the rights and welfare of people involved in research. You may contact the CHEO Chair of the Research Ethics Board, for information regarding patient s rights in research studies at 737-7600 (3272), although this person cannot provide any health-related information about the study. The Protocol Officer for Ethics in Research at the University of Ottawa can be reached at tel.: (613) 562-5841, email: ethics@uottawa.ca.

I, _____, consent to my participation in the study described above.

Signature _____ Date _____

I would like to obtain the results of study once it is completed. Yes ____ No____

Name of witness
(printed)_____

Signature _____ Date _____

CHEO Site Investigator : Linda Moran Professional Practice Leader CHEO Audiology (613) 737-7600 ext. 2379	Doctoral Student Investigator: J. Cyne Johnston, M.Sc., University of Ottawa Doctoral Student Population Health Ph.D. Program, cyne_johnston@hotmail.com

moran@cheo.on.ca	(613) 738- 3907
Doctoral Thesis Committee: Andrée Durieux-Smith, Ph.D., Children's Hospital of Eastern Ontario Research Institute Hearing Research Lab adurieux@uottawa.ca (613) 738-3907	Annette O'Connor, Ph.D., Tier 1, Canada Research Chair Faculty of Health Sciences, University of Ottawa aoconnor@ohri.ca
Doug Angus, M.A. Institute of Population Health University of Ottawa 562-5800 extension 4545 Angus@management.uottawa.ca	Ian Graham, Ph.D. Faculty of Health Sciences University of Ottawa igraham@ohri.ca
JoAnne Whittingham Children's Hospital of Eastern Ontario Research Institute Hearing Research Lab adurieux@uottawa.ca (613) 738-3907	

A Needs Assessment for the Development of a Decision Aid to Help Parents of Children with Hearing Loss Decide Between Cochlear Implantation and Continued Hearing Aid Use

We would like to invite you to take part in a research study. This study examines how best to support parents making a decision about cochlear implants for their child. Researchers at the University of Ottawa and CHEO are conducting the study. Cyne Johnston is the University of Ottawa doctoral student undertaking this research. This study will take place in English.

The diagnosis of a hearing loss can be a time of great stress for parents. Many decisions have to be made by these parents. These include the communication approaches they will use and hearing aid options. Parents of children with severe or profound hearing loss may also have to make decisions about cochlear implant use.

Purpose of this study

- To understand the experiences you have in helping parents make the cochlear implant decision.
- To explore the possibility of developing a decision aid to help parents of children with significant hearing losses choose an amplification option for their child.

If you choose not to participate in the study, this will in no way affect your position at CHEO.

Procedure
1) In this study we will be asking you to allow a researcher to interview you about how you view the cochlear implant decision. The researcher will ask you questions about parents' feelings as they make this decision. They will also ask about the type of information that you provide to help them with decision making. They are interested in knowing about the people who helped parents make this decision. The interview will last between 30 and 45 minutes. This interview will be tape recorded.

All information obtained in the interviews will be kept confidential. Any tapes, transcripts, or questionnaires from the study will be kept in the research office for five years after the interviews. Only members of the research team will have access to the study information. The information gathered will be summarized and individual answers will not be identified. The results of this study may be used in research papers, conference, or course presentations. Quotes from individuals may be used but no identifying information will accompany them.

Subject description
Members of the CHEO Cochlear Implant Team who help parents to make the cochlear implant decision will be asked to take part in this research.

Are there any risks to participating in this research?
The interviews can be completed at your convenience. They can be conducted at your home or at the CHEO Research Institute. It will take you about a half an hour to participate in this study. There are no physical risks associated with this study.

Benefits

This study will help us to identify what parents need make the cochlear implant decision. It will help us to develop a tool to help other parents with the decision.

Withdrawing from the study

If you choose to withdraw from the study, this will in no way affect your position at CHEO. At any time during the study you may decline to answer a question, withdraw your answer, or stop the interview. This can be done by indicating your choice to withdraw from the study to the interviewer. Any data already gathered will be destroyed if you withdraw from the study.

I acknowledge that the research procedures described above, and of which I have a copy, have been explained to me. I have been given an opportunity to ask questions concerning the study and the interview process and the questions that I have asked have been adequately answered. In addition, I know that I may contact any of the investigators (whose contact information is below) if I have further questions either now or in the future.

I have been assured that personal records relating to this study will be kept confidential except as required or permitted by law. I understand that I am free to withdraw from the study at any time. I understand that I can withdraw my consent and stop my participation in the study at any time and for any reason. I have been reassured that this will not affect the quality of care that I or any member of my family receives at CHEO nor my position at the hospital. I understand that if any knowledge gained from this study becomes available that could influence my decision to continue in this study, I will be promptly informed.

The Research Ethics Board is a group of people from scientific and non-scientific backgrounds that review research studies. Their goal is to ensure the protection of the rights and welfare of people involved in research. You may contact the CHEO Chair of the Research Ethics Board, for information regarding patient s rights in research studies at 737-7600 (3272), although this person cannot provide any health-related information about the study. The Protocol Officer for Ethics in Research at the University of Ottawa can be reached at tel.: (613) 562-5841, email: ethics@uottawa.ca.

I, _____, consent to my participation in the study described above.

Signature _____ Date _____

I would like to obtain the results of study once it is completed. Yes ____ No____

Name of witness
(printed)_____

Signature _____ Date _____

CHEO Site Investigator : Linda Moran Professional Practice Leader CHEO Audiology (613) 737-7600 ext. 2379 moran@cheo.on.ca	Doctoral Student Investigator: J. Cyne Johnston, M.Sc., University of Ottawa Doctoral Student Population Health Ph.D. Program, cyne_johnston@hotmail.com (613) 738- 3907
Doctoral Thesis Committee: Andrée Durieux-Smith, Ph.D.,	Annette O'Connor, Ph.D.,

Children's Hospital of Eastern Ontario Research Institute Hearing Research Lab adurieux@uottawa.ca (613) 738-3907	Tier 1, Canada Research Chair Faculty of Health Sciences, University of Ottawa aoconnor@ohri.ca
Doug Angus, M.A. Institute of Population Health University of Ottawa 562-5800 extension 4545 Angus@management.uottawa.ca	Ian Graham, Ph.D. Faculty of Health Sciences University of Ottawa igraham@ohri.ca
JoAnne Whittingham Children's Hospital of Eastern Ontario Research Institute Hearing Research Lab adurieux@uottawa.ca (613) 738-3907	

Consent Form for Parents

The evaluation of a decision aid for parents deciding about bilateral cochlear implantation for their children with hearing loss

We would like to invite you to take part in a research study. This study examines how best to support parents making a decision about bilateral cochlear implants for their child. Researchers at CHEO and the University of Ottawa are conducting the study. Cyne Johnston is the doctoral student undertaking this research. This study will take place in English.

The diagnosis of a hearing loss can be a time of great stress for parents. Many decisions have to be made by these parents. These include the communication approaches they will use and hearing aid options. Parents of children with severe or profound hearing loss may also have to make decisions about a single or bilateral cochlear implant use. A decision aid was developed to help parents understand the risks and benefits of bilateral cochlear implants. Decision aids help with decision making by giving individuals information on the options available and the possible benefits and consequences. Decision aids also help people participate with health professionals in deciding about options.

Purpose of this study

- To find out if parents who use a decision aid improve their knowledge about bilateral cochlear implants.
- To find out if parents who use the decision aid have fewer conflicting feelings about their decision.
- To find out if parents and clinicians find that the new decision aid is acceptable.

If you choose not to participate in the study, this will in no way affect the care you and your family members receive at CHEO.

Procedure

1) You will be asked to fill out two questionnaires. One is on your bilateral cochlear implant knowledge. The other is on the difficulty that you are having making the decision.
2) Next, you will be given a copy of the decision aid from a member of the research team. After reading this, you will be given three brief questionnaires. One will be about the material in the decision aid. A second will be about your ability to make the decision. A third will look at how acceptable decision aid is to you.

These steps will be done on one visit and will take about an hour to complete.

All information obtained in the interviews will be kept confidential. Any questionnaires from the study will be kept in the research office for five years after the study. Only members of the research team will have access to the study information. The information gathered will be summarized and individual answers will not be identified. The results of this study may be used in research papers, conference, or course presentations. Quotes from individuals may be used but no identifying information will accompany them

Subject description

20 families of children with hearing loss who are making the cochlear implant decision will be asked to take part in this research.

Are there any risks to participating in this research?
The questionnaires and decision aid will be completed at a visit with the Cochlear Implant Team at CHEO. It will take you about an hour to participate in this study. There are no physical risks associated with this study.

Benefits
This study will help us to identify what parents need to make the bilateral cochlear implant decision. It will help us to develop a tool to help other parents with the cochlear implant decision.

Withdrawing from the study
If you choose to withdraw from the study, this will in no way affect the care you and your family members receive. At any time during the study you may decline to answer a question, withdraw your answer, or stop participating. This can be done by indicating your choice to withdraw from the study to the interviewer. Any data already gathered will be destroyed if you withdraw from the study.

I acknowledge that the research procedures described above, and of which I have a copy, have been explained to me. I have been given an opportunity to ask questions concerning the study and the interview process and the questions that I have asked have been adequately answered. In addition, I know that I may contact any of the investigators (whose contact information is below) if I have further questions either now or in the future.

Confidentiality
I have been assured that personal records relating to this study will be kept confidential except as required or permitted by law. I understand that I am free to withdraw from the study at any time. I understand that I can withdraw my consent and stop my participation in the study at any time and for any reason. I have been reassured that this will not affect the quality of care that I or any member of my family receives at CHEO.

If parents require any additional information regarding cochlear implantation, Linda Moran, the professional practice leader in the Audiology clinic is available to answer their questions. Her contact information is available at the bottom of this form.

This study has been reviewed and approved by the CHEO Research Ethics Board. The CHEO Research Ethics Board 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 protection of the rights and welfare of people participating in research. The Board's work is not intended to replace a parent or child's judgment about what decisions and choices are best for them. You may contact the Chair of the Research Ethics Board, for information regarding patient's rights in research studies at (613) 737-7600 (3272), although this person cannot provide any health-related information about the study. The Protocol Officer for Ethics in Research at the University of Ottawa can be reached at tel.: (613) 562-5841, email: ethics@uottawa.ca.

I, _____, consent to my participation in the study described above.

Signature _____ Date _____

Name of witness
(printed)

Signature _____ Date _____

I HAVE EXPLAINED THIS STUDY TO THE PARTICIPANT AND I AM SATISFIED THAT IT IS UNDERSTOOD

Name and Title _____

Signature _____ Date _____

CHEO Site Investigator : Linda Moran Professional Practice Leader CHEO Audiology (613) 737-7600 ext. 2379 moran@cheo.on.ca	Doctoral Student Investigator: J. Cyne Johnston, M.Sc., Doctoral Student Population Health Ph.D. Program, University of Ottawa cyne_johnston@hotmail.com (613) 738- 3907
Doctoral Thesis Committee: Andrée Durieux-Smith, Ph.D., Children's Hospital of Eastern Ontario Research Institute Hearing Research Lab adurieux@uottawa.ca (613) 738-3907	Annette O'Connor, Ph.D., Tier 1, Canada Research Chair Faculty of Health Sciences, University of Ottawa aoconnor@ohri.ca (613) 562-5800 ext 8337
Elizabeth Fitzpatrick, Ph.D. Faculty of Health Sciences, University of Ottawa (613) 738-3907 elizabeth.fitzpatrick@uottawa.ca	Doug Angus, M.A. Institute of Population Health University of Ottawa 562-5800 extension 4545 Angus@management.uottawa.ca
JoAnne Whittingham, M.Sc. Children's Hospital of Eastern Ontario, Research Institute Hearing Research Lab jwhitt@uottawa.ca (613) 738-3907	

Consent Form for Clinicians

The evaluation of a decision aid for parents deciding about bilateral cochlear implantation for their children with hearing loss

We would like to invite you to take part in a research study. This study examines how best to support parents making a decision about bilateral cochlear implants for their child. Researchers at CHEO and the University of Ottawa are conducting the study. Cyne Johnston is the doctoral student undertaking this research. This study will take place in English.

The diagnosis of a hearing loss can be a time of great stress for parents. Many decisions have to be made by these parents. These include the communication approaches they will use and hearing aid options. Parents of children with severe or profound hearing loss may also have to make decisions about a single or bilateral cochlear implant use. A decision aid was developed to help parents understand the risks and benefits of bilateral cochlear implants. Decision aids help with decision making by giving individuals information on the options available and the possible benefits and consequences. Decision aids also help people participate with health workers in deciding about options.

Purpose of this study

- To find out if parents who use a decision aid improve their knowledge about bilateral cochlear implants.
- To find out if parents who use the decision aid have fewer conflicting feelings about their decision.
- To find out if parents and clinicians find that the new decision aid is acceptable.

If you choose not to participate in the study, this will in no way affect the care you and your family members receive at CHEO nor your position at the hospital.

Procedure
You will be asked to fill out a questionnaire that will look at how acceptable decision aid is to you.

These steps will be done on one visit and will take about a half an hour to complete.

All information obtained in the questionnaires will be kept confidential. Any questionnaires from the study will be kept in the research office for five years after the study. Only members of the research team will have access to the study information. The information gathered will be summarized and individual answers will not be identified. The results of this study may be used in research papers, conference, or course presentations. Quotes from individuals may be used but no identifying information will accompany them.

Subject description
10 clinicians in the Audiology Clinic will be asked to take part in this research.

Are there any risks to participating in this research?

It will take you about a half an hour to participate in this study. There are no physical risks associated with this study.

Benefits

This study will help us to identify what parents need to make the cochlear implant decision. It will help us to develop a tool to help other parents with the bilateral cochlear implant decision.

Withdrawing from the study
If you choose to withdraw from the study, this will in no way affect the care you and your family members receive. At any time during the study you may decline to answer a question, withdraw your answer, or stop participating. This can be done by indicating your choice to withdraw from the study to the interviewer. Any data already gathered will be destroyed if you withdraw from the study.

I acknowledge that the research procedures described above, and of which I have a copy, have been explained to me. I have been given an opportunity to ask questions concerning the study and the interview process and the questions that I have asked have been adequately answered. In addition, I know that I may contact any of the investigators (whose contact information is below) if I have further questions either now or in the future.

Confidentiality
I have been assured that personal records relating to this study will be kept confidential except as required or permitted by law. I understand that I am free to withdraw from the study at any time. I understand that I can withdraw my consent and stop my participation in the study at any time and for any reason. I have been reassured that this will not affect the quality of care that I or any member of my family receives at CHEO.

If parents require any additional information regarding cochlear implantation, Linda Moran, the professional practice leader in the Audiology clinic is available to answer their questions. Her contact information is available at the bottom of this form.

This study has been reviewed and approved by the CHEO Research Ethics Board. The CHEO Research Ethics Board 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 protection of the rights and welfare of people participating in research. The Board's work is not intended to replace a parent or child's judgment about what decisions and choices are best for them. You may contact the Chair of the Research Ethics Board, for information regarding patient's rights in research studies at (613) 737-7600 (3272), although this person cannot provide any health-related information about the study. The Protocol Officer for Ethics in Research at the University of Ottawa can be reached at tel.: (613) 562-5841, email: ethics@uottawa.ca.

I, _____, consent to my participation in the study described above.

Signature _____ Date _____

Name of witness
(printed)

Signature _____ Date _____

I HAVE EXPLAINED THIS STUDY TO THE PARTICIPANT AND I AM SATISFIED THAT IT IS UNDERSTOOD

Name and Title _____

Signature _____ Date _____

CHEO Site Investigator : Linda Moran Professional Practice Leader CHEO Audiology (613) 737-7600 ext. 2379 moran@cheo.on.ca	Doctoral Student Investigator: J. Cyne Johnston, M.Sc., Doctoral Student Population Health Ph.D. Program, University of Ottawa cyne_johnston@hotmail.com (613) 738- 3907
Doctoral Thesis Committee: Andrée Durieux-Smith, Ph.D., Children's Hospital of Eastern Ontario Research Institute Hearing Research Lab adurieux@uottawa.ca (613) 738-3907	Annette O'Connor, Ph.D., Tier 1, Canada Research Chair Faculty of Health Sciences, University of Ottawa aoconnor@ohri.ca (613)562-5800 ext 8337
Elizabeth Fitzpatrick, Ph.D. Faculty of Health Sciences, University of Ottawa (613) 738-3907 eliabeth.fitzpatrick@uottawa.ca	Doug Angus, M.A. Institute of Population Health University of Ottawa 562-5800 extension 4545 Angus@management.uottawa.ca
JoAnne Whittingham, M.Sc. Children's Hospital of Eastern Ontario, Research Institute Hearing Research Lab jwhitt@uottawa.ca (613) 738-3907	

Appendix F: Decision Aid and Supporting Documents

Bilateral Cochlear Implant Decision Aid

Facts and Numbers Behind the Decision Aid

Background Document

Should my child have a second cochlear implant?

A decision aid to discuss options with your health care team

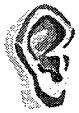
This decision aid is for you if:

- Your child has a cochlear implant.
- You are wondering about a cochlear implant for your child's other ear.

What is a cochlear implant?

- A cochlear implant is a surgically implanted electronic device that provides a sense of sound to a person who is profoundly deaf or severely hard of hearing. The internal component works by directly stimulating the auditory nerves inside the cochlea with electrical impulses. External components include a microphone, speech processor and transmitter.

What are your options?



Accept a second cochlear implant. The procedure is the same as the first. After a general anaesthetic, the device is implanted by surgery. Your child will stay in the hospital for about 2 days. Healing takes about a month and then the device is activated. Your child will have continuing auditory-verbal or other type of therapy. The second implant provides auditory stimulation to your child's second ear. The second implant can also act as a "back-up" in case of damage or malfunction of the first implant.

Decline a second cochlear implant. You continue with the current implant and follow-up therapy. You may wish to consider an implant at a later date. Some research indicates that when children receive the second implant within a shorter period after the first one they see greater benefits than when they receive the implant later.

What other health factors may affect your choice?

Check ☑ any that apply and discuss your concerns with your doctor.

Your child may not be a candidate for a second implant if he or she has the following ……→	☐ Absent auditory nerve in second ear
	☐ Medical reasons for not having surgery

Working through the 4 steps of this decision aid may prepare you for decision making.

Step 1: What are the benefits and risks of each option?

Step 2: Which reasons to choose each option matter most to you?

Step 3: What else do you need to prepare for decision-making?

Step 4: What are the next steps?

Step 1: What are the benefits and risks of each option?

What does the research show? Blocks of 100 faces show a 'best estimate' of what happens to **100 children who already have one cochlear implant** who make different choices over 1 year. Each face (☺) stands for one child. The shaded areas show the number of children affected. The numbers are averaged from more than one study.
There is no way of knowing in advance if your child will be the one who is affected.
Because this is a new procedure, there is not a lot of research on the long-term effects of bilateral implants. The numbers presented here are based on the literature. Your cochlear implant centre may have different rates. Please talk to your clinician about the rates at your centre.

	1 cochlear implant Decline 2nd cochlear implant	2 cochlear implants Accept 2nd cochlear implant	Comments from CHEO clinic
Benefits			
✦ More children will be able to **locate sounds in their environment** with a second implant. For example, a child could hear the direction of a parent's voice more easily →	**17** improve **83** don't improve	**46** improve **54** don't improve	The clinic has not measured sound localization in their patients.
✦ More children will be able to **understand speech in a noisy environment** at normal levels with a second implant. For example, being able to understand people talking in a group setting →	**0** improve **100** don't improve	**13** improve **87** don't improve	The clinic has measured some improvements in understanding of speech in noise for most children with two implants
✦ The same number of children will have **normal receptive vocabulary** with a second implant as those with one implant →	**67** have normal receptive vocabulary **33** are still working on this	**67** have normal receptive vocabulary **33** are still working on this	The clinic's experience is that children who have had more challenges in developing speech and language with one implant will continue to do so with two implants.
Risks			
✦ There is a risk of **surgical complications** with a second cochlear implant just like the first surgery → These may include ear draining or skin infections, or infections in the head area behind the middle ear.	**0** get this **100** avoid this	**12** get complications **88** avoid this	Our clinic has similar rates.
✦ There is a risk of a **permanent facial palsy** with a second cochlear implant just like the first surgery → A facial palsy is an inability to control some muscles in the face.	**0 of 1000 have this** **1000 of 1000 avoid this**	**1 of 1000 have this** **999 of 1000 avoid this**	Our centre has **not had a child experience** this. There is a risk of this in the future.
✦ There is a risk of **meningitis** with a second cochlear implant surgery just like the first surgery → Meningitis is an infection in the fluid around the brain and spinal cord.	**2 of 1000 have this** **998 of 1000 avoid this**	**2 of 1000 have this** **998 of 1000 avoid this**	Our centre has similar rates
Risks over a *FIVE YEAR* period			
✦ There is a risk of a **damaged / defective implant** with a second cochlear implant just like the first implant → Damage to the implant can happen from falling and hitting the implant, or from a problem with the implant itself. The device needs to be replaced in this case.	**7** have a damaged implant **93** avoid this	**14** have a damaged implant **86** avoid this	The rate at our centre is slightly better. About **4/100** children with one implant experience a damaged or defective implant.

Platinum or ✳ Gold symbols mean stronger study results. ✧ Silver or ✦ Bronze symbols mean weaker results

Step 2. Which reasons to choose each option matter most to you?

Common reasons to choose each option are listed below.
Check ✓ how much each reason matters to you on a scale from 0 to 5.
'0' means it is **not** important to you. **'5'** means it is **very** important to you.

 Reasons to **Accept a second cochlear implant**	**Not Important**	**Very Important**
How important is it to improve your child's ability to locate sounds in their environment such as being able to determine the direction of a person's voice?	⓪ ① ② ③ ④ ⑤	
How important is it to improve your child's ability to understand people talking in a noisy setting such as a classroom or restaurant?	⓪ ① ② ③ ④ ⑤	
How important is it to expose your child's second ear to early speech and language stimulation?	⓪ ① ② ③ ④ ⑤	
List other reasons to accept a second cochlear implant:		
	⓪ ① ② ③ ④ ⑤	

 Reasons to **Decline a second cochlear implant**	**Not Important**	**Very Important**
How important is it to avoid the risks of surgery such as ear draining, skin infections, or infections in the head area behind the middle ear?	⓪ ① ② ③ ④ ⑤	
How important is it to wait until there is more scientific evidence on the effects of a second cochlear implant?	⓪ ① ② ③ ④ ⑤	
List other reasons to decline a 2nd cochlear implant:		
	⓪ ① ② ③ ④ ⑤	

Now, think about which option has the reasons that are most important to you.

Which option do you prefer? Check ☑ one
☐ 2 cochlear implants / Accept a second cochlear implant
☐ 1 cochlear implant / Decline a second cochlear implant
☐ I don't know

Step 3: What else do you need to prepare for decision making?

Knowledge

Find out how well this decision aid helped you learn the key facts.
Check ☑ the best answer.

	2 cochlear implants Accept 2nd implant	1 cochlear implant Decline 2nd implant	Both are equal	Don't know
1. Which option has the highest chance of **your child being able to locate sounds in the environment?**	☐	☐	☐	☐
2. Which option has the highest chance of **your child being able to understand speech in a noisy environment?**	☐	☐	☐	☐
3. Which option has an increased chance of **ear draining, skin infections, or infections in the head area behind the middle ear?**	☐	☐	☐	☐
4. Which option improves **receptive vocabulary?**	☐	☐	☐	☐
5. Which option has the most scientific evidence on **long term outcomes?**	☐	☐	☐	☐

Check your answers at the bottom of the page.

Do you know the current evidence on benefits and risks of each option? ☐ Yes ☐ No

Values
Are you clear about which benefits and risks **matter most** to you? ☐ Yes ☐ No

Support
Do you have enough support and advice from others to make a choice? ☐ Yes ☐ No

Uncertainty
Do you feel sure about the best choice? ☐ Yes ☐ No

Step 4: What are the next steps?
List your plans: (for example, discuss the options with your health care team and/or learn more about the options)

This information is not intended to replace the advice of a health care provider.

Answers for the key facts- 1. 2 implants, accept 2nd implant 2. 2 implants, accept 2nd implant 3. 2 implants, accept 2nd implant. 4. Both equal 5. 1 implant, decline 2nd implant

Content Editors: C Johnston, A Durieux-Smith, L Moran, Dr. D Schramm, E. Fitzpatrick
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 Next update due 2009.

Facts and Numbers Behind the Decision Aid.....

Should my child have a second cochlear implant?

A decision aid to discuss options with your health care team

Content Editors:
J.C. Johnston, A. Durieux-Smith, E. Fitzpatrick, Linda Moran, D. Schramm MD FRC

Decision Aid Format Editors:
A. O'Connor

The following pages provide the rationale and references to data regarding benefits and risks of bilateral cochlear
implantation that are provided in the decision aid.

Sound Localization Data

Option	Unilateral Cochlear Implant	Bilateral Cochlear Implants
N Studies (N Participants)	5 (35)	5 (37)
Populations	Children 2-16 years Sequential BICIs	
Type of Study	Retrospective Cohort Studies (3) Case Control Study (1)	
Event Rate	5/35	17/37
Case Definition	Performance of the Minimum Audible Angle (MAA) task at <20°	
Study Time Frame	Range of 3 to 26 months experience with BICI	
Study Rating	Bronze	Bronze
Source	Litovsky, Johnstone, Godar, et al. (2006) Greico-Calub, Litovsky & Werner (2008) Litovsky, Johnstone & Godar (2006) Senn, Kompis, Vischer et al. (2005)	Litovsky, Johnstone, Godar, et al. (2006) Greico-Calub, Litovsky & Werner (2008) Litovsky, Johnstone & Godar (2006) Senn, Kompis, Vischer et al. (2005)
Event rate per 100 (Event rate in Pt DA)	17.24 (17)	45.95 (46)

Speech Recognition in Noise

Option	Unilateral Cochlear Implant	Bilateral Cochlear Implants
N Studies (N Participants)	2 (16)	2 (16)
Populations	Children 4-15 years Sequential BICIs	
Type of Study	Retrospective Cohort Study (1) Case Series (1)	
Event Rate	0/16	3/16
Case Definition	Performance of the AdSpon task at a Signal to Noise Ratio of -11.5 based on Rance et al., (2007)	
Study Time Frame	Range of 6 to 13 months experience with BICI	
Study Rating	Bronze	Bronze
Source	Galvin, Mok, & Dowell, (2007) Galvin, Mok, Dowell, & Briggs, (2007)	Galvin, Mok, & Dowell, (2007) Galvin, Mok, Dowell, & Briggs, (2007)
Event rate per 100 (Event rate in Pt DA)	0 (0)	18.0 (18)

Receptive Vocabulary

The systematic review did not identify any group studies comparing language development in children with bilateral CIs. Based on what is known about binaural hearing in individuals with normal hearing and hearing loss (Mencher & Davis, 2006) there is no expectation that language will improve in children with bilateral cochlear implants. Rates of language development that have been published for children with unilateral CIs are used here.

Option	Unilateral Cochlear Implant	Bilateral Cochlear Implants
N Studies	1	0
(N Participants)	(76)	(0)
Populations	Children	
Type of Study	Cross-sectional Study (1)	
Event Rate	51/76	
Case Definition	Receptive vocabulary (PPVT) at normal levels for age	
Study Time Frame	4-6 years post CI	
Study Rating	Bronze	Bronze
Source	Nicholas & Geers, (2008)	Mencher & Davis (2006)
Event rate per 100 (Event rate in Pt DA)	67.1 (67)	Expected to be the same as unilateral CI rate based on expert opinion and binaural hearing literature

Complications Data

Option	Unilateral Cochlear Implant	Bilateral Cochlear Implants
N Studies	12	0
(N Participants)	(2053 & 2161)	(0)
Populations	See Technical Background Document for Facts and Numbers behind the Bilateral CI Decision Aid	
Type of Study	Prospective Cohort (1) Retrospective Cohort (12)	
Event Rate	Minor Complications 169/2053 Major Complications 70/2161	
Case Definition	See Technical Background Document for Facts and Numbers behind the Bilateral CI Decision Aid	
Study Time Frame	See Technical Background Document for Facts and Numbers behind the Bilateral CI Decision Aid	
Study Rating	Bronze	Bronze
Source	Stratigouleas, Perry, King, et al. (2006) Cunningham, Slattery & Luxford (2004) Migrov, Muchnik, Kaplan-Neeman et al. (2006) Gysin, Papsin, Daya & Nedzelski (2000) Arnoldner, Baumgartner, Gstoettner et al. (2005) James & Papsin (2004) Black, Bailey, Albert, et al. (2007) Kandogan, Levent & Gurol (2005) Bhatia, Gibbin, Nikolopoulos et al. (2004) Postelmans, Cleffken & Stokroos (2006) Venail, Sicard, Piron et al. (2007) Yu, Hegarty, Gantz et al. (2001)	
Event rate per 100 (Event rate in Pt DA)	Minor Complications 8.23 Major Complications 2.23 Combined 10.46 (10)	Expected to be the same as unilateral CI rate based on expert opinion

Permanent Facial Palsy

Option	Unilateral Cochlear Implant	Bilateral Cochlear Implants
N Studies (N Participants)	9 (2450)	
Populations	See Technical Background Document for Facts and Numbers behind the Bilateral CI Decision Aid	
Type of Study	Prospective Cohort (1) Retrospective Cohort (8)	
Event Rate	3/2450	
Case Definition	Facial nerve weakness or paralysis occurring following CI surgery	
Study Time Frame	See Technical Background Document for Facts and Numbers behind the Bilateral CI Decision Aid	
Study Rating	Bronze	Bronze
Source	Fayad, Wanna, Micheletto et al. (2003) Venail, Sicard, Piron et al. (2007) Stratigouleas, Perry, King, et al. (2006) Migrov, Muchnik, Kaplan-Neeman et al. (2006) Gysin, Papsin, Daya & Nedzelski (2000) Arnoldner, Baumgartner, Gstoettner et al. (2005) Black, Bailey, Albert, et al. (2007) Kandogan, Levent & Gurol (2005) Bhatia, Gibbin, Nikolopoulos et al. (2004)	
Event rate per 1000 (Event rate in Pt DA)	1.22 (1/1000)	Expected to be the same as unilateral CI rate based on expert opinion

Meningitis

Option	Unilateral Cochlear Implant	Bilateral Cochlear Implants
N Studies (N Participants)	2 (13441 person years)	
Populations	See Technical Background Document for Facts and Numbers behind the Bilateral CI Decision Aid	
Type of Study	Retrospective Cohort (2) / nested case control study (1)	
Event Rate	26/13441 person years	
Case Definition	Bacterial Meningitis	
Study Time Frame	1 year	
Study Rating	Silver	Bronze
Source	Summerfield, Cirstea, Roberts, et al. (2004) Reefhuis, Honein, Whitney, et al. (2003)	
Event rate per 1000 (Event rate in Pt DA)	1.93 (2)	Expected to be the same as unilateral CI rate based on expert opinion

Crude Device Failure Rate

Option	Unilateral Cochlear Implant	Bilateral Cochlear Implants
N Studies (N Participants)	8 (14032)	0 (0)
Populations	See Technical Background Document for Facts and Numbers behind the Bilateral CI Decision Aid	
Type of Study	Retrospective Cohort (8)	
Event Rate	550/14032	
Case Definition	Hard failures of cochlear implants due to design errors and direct or indirect trauma to the CI site	
Study Time Frame	See Technical Background Document for Facts and Numbers behind the Bilateral CI Decision Aid	
Study Rating	Bronze	Bronze
Source	Stratigouleas, Perry, King, et al. (2006) Gysin, Papsin, Daya & Nedzelski (2000) Arnoldner, Baumgartner, Gstoettner et al. (2005) Kandogan, Levent & Gurol (2005) Battmer, O'Donoghue, & Lenarz (2007) Postelmans, Cleffken & Stokroos (2006) Weise, Muller-Deile, Brademann et al.(2005) Parisier, Chute, Popp & Suh (2001)	
Event rate per 100 (Event rate in Pt DA)	3.97 (4)	Expected to be the same as unilateral CI rate based on expert opinion

Five-Year Device Failure Rate

Option	Unilateral Cochlear Implant	Bilateral Cochlear Implants
N Studies (N Participants)	2 (464)	0 (0)
Populations	See Technical Background Document for Facts and Numbers behind the Bilateral CI Decision Aid	
Type of Study	Retrospective Cohort (2)	
Event Rate	31/464	
Case Definition	Hard failures of cochlear implants due to design errors and direct or indirect trauma to the CI site	
Study Time Frame	See Technical Background Document for Facts and Numbers behind the Bilateral CI Decision Aid	
Study Rating	Bronze	Bronze
Source	Venail, Sicard, Piron et al. (2007) Maurer, Marangos & Ziegler (2005)	
Event rate per 100 (Event rate in Pt DA)	6.68 (7)	Expected to be twice the rate as unilateral CI rate based on expert opinion

Glossary

Definitions of Study Ratings

Platinum: Evidence from a published systematic review that has at least two individual controlled trials each satisfying the following:

- Sample sizes of at least 50 per group – if these do not find a satisfactory significant difference, they are adequately powered for a 20% relative difference in the relevant outcome.
- Blinding of patients and assessors for outcomes.
- Handling of withdrawals >80% follow up (imputations based on methods such as Last Observation Carried Forward (LOCF) are acceptable).
- Concealment of treatment allocation.

Gold: Evidence from at least one randomized clinical trial meeting all of the following criteria for the major outcome(s) are reported:

- Sample sizes of at least 50 per group – if these do not find a statistically significant difference, they are adequately powered for a 20% relative difference in the relevant outcome.
- Blinding of patients and assessors for outcomes.
- Handling of withdrawals >80% follow up (imputations based on methods such as Last Observation Carried Forward (LOCF) are acceptable).
- Concealment of treatment allocation.

Silver: Evidence from a systematic review or randomized trial that does not meet the above criteria. Silver ranking would also include evidence from at lease one study of non-randomized cohorts that did and did not receive the therapy or evidence from at least one high quality case control study. A randomized trial with a 'head-to-head' comparison of agents would be considered silver level ranking unless a reference were provided to a comparison of one of the agents to placebo showing at least a 20% relative difference.

Bronze: Evidence from at least one high quality case series without controls (including simple before/after studies in which patients act as their own control) or from expert opinion based on clinical experience without reference to any of the foregoing (for example, argument from physiology, bench research or first principles).

References

Arnoldner C, Baumgartner WD, Gstoettner W, Hamzavi J (2005) Surgical considerations in cochlear implantation in children and adults: a review of 342 cases in Vienna. *Acta Oto-Laryngologica, 125*, 228-234.

Battmer RD, O'Donoghue GM, Lenarz T (2007) A multicenter study of device failure in European cochlear implant centers. *Ear and Hearing, 28*, 95S-99S.

Bhatia K, Gibbin KP, Nikolopoulos TP, O'Donoghue GM (2004) Surgical complications and their management in a series of 300 consecutive pediatric cochlear implantations. *Otology and Neurotology*, 25, 730–739.

Black IM, Bailey CM, Albert DM, Leighton SEJ, Hartley BEJ, Chatrath P, Patel N (2007) The Great Ormond Street Hospital paediatric cochlear implant programme 1992–2004 A review of surgical complications *Cochlear Implants International, 8,* 53–67.

Brown KD, Balkany TJ (2007) Benefits of bilateral cochlear implantation: a review, *Current Opinion in Otolaryngology, Head and Neck Surgery,* 15, 315-8.

Cunningham CD, Slattery WH, Luxford WM (2004). Postoperative infection in cochlear implant patients. *Otolaryngology - Head & Neck Surgery, 131*, 109-114.

Galvin KL, Mok M, Dowell RC (2007) Perceptual benefit and functional outcomes for children using sequential bilateral cochlear implants, *Ear and Hearing, 28*, 470-82.

Galvin KL, Mok M, Dowell RC, Briggs RJ (2007) 12-month post-operative results for older children using sequential bilateral implants, *Ear and Hearing, 28*, 19S-21S.

Nicholas JG & Geers AE (2008) Expected test scores for preschoolers with a cochlear implant who use spoken language. *American Journal of Speech-Language Pathology, 17*, 121-138.

Grieco-Calub TM, Litovsky RY & Werner LA (2008) Using the observer-based psychophysical procedure to assess localization acuity in toddlers who use bilateral cochlear implants, *Otology Neurootology, 29*, 235-9.

Gysin C, Papsin BC, Daya H, Nedzelski J (2000) Surgical outcome after paediatric cochlear implantation: Diminuation of complications with the evolution of new surgical techniques, *The Journal of Otolaryngology, 29,* 285-289.

Fayad JN, Wanna GB, Micheletto JN, Parisier SC (2003) Facial nerve paralysis following cochlear implant surgery. *Laryngoscope, 113,*1344–1346.

James AL, Papsin BC (2004) Cochlear implant surgery at 12 months of age or younger. *Laryngoscope, 114*, 2191-2195.

Johnston JC, Durieux-Smith A, Angus D, O'Connor A, Fitzpatrick E (unpublished) Bilateral paediatric cochlear implants: a critical review.

Kandogan T, Levent O, Gurol G (2005). Complications of paediatric cochlear implantation: experience in Izmir. *Journal of Laryngology & Otology, 119*, 606-610.

Litovsky RY, Johnstone PM, Godar S, Agrawal S, Parkinson, A et al. (2006) Bilateral cochlear implants in children: localization acuity measured with minimum audible angle, *Ear and Hearing, 27*, 43-59.

Litovsky RY, Johnstone PM, Godar, SP (2006) Benefits of bilateral cochlear implants and/or hearing aids in children, *International Journal of Audiology, 45*, S78-91.

Maurer J, Marangos N, Ziegler E (2005). Reliability of cochlear implants. *Otolaryngology—Head and Neck Surgery, 132*, 746–750.

Mencher, GT, Davis, A (2006) Bilateral or unilateral amplification: is there a difference? A brief tutorial, *International Journal of Audiology, 45,* S3-S11.

Moog JS, Geers AE (2003) Epilogue: Major findings, conclusions and implications for deaf education. *Ear and Hearing, 24*, 121S-125S.

Migirov L, Yakirevitch A, Kronenberg J (2006) Surgical and medical complications following cochlear implantation: comparison of two surgical approaches. *ORL, 68,* 213-219.

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 International, 7*, 194-201.

Parisier SC, Chute PM, Popp AL, Suh GD (2001) Outcome analysis of cochlear implant reimplantation in children. *Laryngoscope, 111*, 26-32.

Postelmans JTF, Cleffken B, Stokroos RJ (2007) Post-operative complications of cochlear implantation in adults and children: Five years' experience in Maastricht. *The Journal of Laryngology and Otology, 121*, 318-323.

Rance G, Barker E, Mok M, Dowell R, Rincon A, Garratt R. (2007) Speech Perception in Noise for Children with Auditory Neuropathy/Dys-Synchrony Type Hearing Loss, *Ear and Hearing, 28,* 351-360.

Reefhuis J, Honein MA, Whitney CG, Chamany S, Mann EA, Biernath KR, Broder K (2003) Risk of bacterial meningitis in children with cochlear implants. *The New England Journal of Medicine, 349*, 433-443.

Senn P, Kompis M, Vischer M, Haeusler R (2005) Minimum audible angle, just noticeable interaural differences and speech intelligibility with bilateral cochlear implants using clinical speech processors, *Audiology and Neurootology, 10*, 342-52.

Stratigouleas ED, Perry BP, King SM, Syms CA (2006) Complication rate of minimally invasive cochlear implantation. *Otolaryngology - Head & Neck Surgery, 135*, 383-386.

Summerfield AQ, Cirstea SE, Roberts KL, Barton GR, Graham JM, O'Donoghue GM (2004) Incidence of meningitis and of death from all causes among users of cochlear implants in the United Kingdom. *Journal of Public Health, 27*, 55-61.

Venail F, Sicard M, Piron JP, Artiered F, Uziel A, Mondain M (2007) Cochlear implantation safety and reliability in paediatric cases. *Audiological Medicine, 5*, 210-217.

Weise JB, Muller-Deile J, Brademann G, Meyer JE, Ambrosch P, Maune S (2005) Impact to the head increases cochlear implant reimplantation rate in children. *Auris Nasus Larynx, 32,* 339-343.

Yu KCY, Hegarty JL, Gantz BJ, Lalwani AK (2001) Conservative management of infections in cochlear implant recipients. *Otolaryngology-Head and Neck Surgery, 125*, 66-70.

Sound Localization Studies Identified in Systematic Review												
						Event Rate Unilateral CI			Event Rate Bilateral CI			
Authors	Definition	Study Design	Sample Size	Time Frame	Population Study Age Sequential/Simultaneous	#	base	CI Rate out of 100	#	base	BICI Rate out of 100	Study Rating
Greico-Calub, Litovsky & Werner (2008)	-MAA as measured by a 2-alternative-forced choice task -+ or - 20° is threshold of interest	Retrospective Cohort study	N=8 BICI N=8 normal hearing N=8 CI	5 to 20 months post BICI Mean= 8 months	Children 2-3 years Sequential BICIs	0	8	0	3	10	30	Silver
Litovsky, Johnstone, Godar, Agrawal, Parkinson, Peters & Lake (2006)	-MAA as measured by a 2-alternative-forced choice task -+ or - 20° is threshold of interest	Retrospective Cohort study	N=13 BICI N=6 CIHA	3 to 14 months post BICI Mean= 7 months	Children 3-16 years Sequential BICIs	2	13	15.38	8	13	61.54	Bronze
Litovsky, Johnstone & Godar (2006)	-MAA as measured by a 2-alternative-forced choice task -+ or - 20° is threshold of interest	Retrospective Cohort study	N=6 BICI N=10 CIHA	3 to 26 months post BICI Mean=14 months	Children 4-14 years Sequential BICIs	1	6	16.67	4	6	66.67	Bronze
Senn, Kompis, Vischer, & Haeusler (2005)	-MAA as measured by a 2-alternative-forced choice task -+ or - 20° is threshold of interest	Case-control study	N=2 BICI N=2 normal hearing	24 months post BICI	Children and adults 14 years Sequential BICIs	2	2	100	2	2	100	Bronze
Peters, Litovsky, Lake & Parkinson (2004)	-MAA as measured by a 2-alternative-forced choice task	Retrospective Cohort study	N=6 BICI	Not Reported	Children 8-13 years Sequential BICIs	0	6	0	0	6	0	Bronze
Total			N=35			5	35	14.29	17	37	45.95	Bronze

Speech Recognition in Noise Identified in Systematic Review

Authors	Definition	Study Design	Sample Size	Time Frame	Population Study Age Sequential/Simultaneous	Event Rate Unilateral CI		CI Rate out of 100	Event Rate Bilateral CI		BICI Rate out of 100	Study Rating
						#	base		#	base		
Galvin, Mok & Dowell (2007)	-Speech Recognition in noise (AdSpon) scores with ipsilateral presentation of noise at Signal to Noise Ratio of -11.5 dB SPL	Cohort Study	N=10	6 to 13 months post BICI Mean= 8 months	Children 4-15 years Sequential	0	10	0	1	10	20	Bronze
Galvin, Mok, Dowell & Briggs (2007)	-Speech Recognition in noise (AdSpon) scores with ipsilateral presentation of noise at Signal to Noise Ratio of -11.5 dB SPL	Case Series	N=6	Not Reported	Children 5-15 years Sequential	0	6	0	1	6	16.67	Bronze
Total			N=16			0	16	0	2	16	12.5	Bronze

Language Development

Authors	Definition	Study Design	Studies (Sample Size)	Time Frame	Population Study Age Sequential/Simultaneous	Event Rate Unilateral CI		CI Rate out of 100	Event Rate Bilateral CI		BICI Rate out of 100	Study Rating
						#	base		#	base		
Nicholas & Geers (2008)	-Receptive vocabulary (PPVT)	Cross-Sectional Study	1 (78)	4.5 years of age 3.5-1 year post CI	Children 4.5 years Unilateral CI Users	51	76	30.38				Bronze/ Silver
Total						51	76	67.1				

Minor Complications Not Requiring Hospitalization

Authors	Definition	Study Design	Sample Size	Time Frame	Population Study Age Sequential/Simultaneous	Event Rate Unilateral CI		CI Rate out of 100	Event Rate Bilateral CI		BICI Rate out of 100	Study Rating
						#	base		#	base		
Stratigouleas, Perry, King, & Syms (2006)	-Managed conservatively on an ambulatory basis -Flap infections, stitch abscess, transient tinnitus and dizziness, otitis externa	Retrospective cohort	N=176	Not reported Follow-up mean 289 days	Adults and Children 1 to 86 years (Mean 27yr) Unilateral CI	12	176	6.82				Bronze
Cunningham, Slattery & Luxford (2004)	- Postoperative infections including wound infections, complicated otitis media, or meningitis -Minor if requiring local wound care or oral antibiotics	Retrospective cohort	N=272 (children)	Implants between 1993 and 2002	Children 1 to 10 years (Mean 4 yr) Unilateral CI	6	272	2.21				Bronze
Migrov, Muchnik, Kaplan-Neeman & Kronenberg (2008)	-Minor complications defined as: wound infection, stitch granuloma	Retrospective cohort study	N=300	Implants between 1993 and 2005	Children 2 to 16 years Unilateral CI	53	300	17.67				Bronze
Gysin, Papsin, Daya & Nedzelski (2000)	- Minor complications: Flap cellulites, perilymph leakage	Retrospective cohort	N=100	Implants between 1990 and 1998	Children 1 to 17 years (Mean= 8 yr) Unilateral CI	2	100	2.00				Bronze
Arnoldner, Baumgartner, Gstoettner & Hamzavi (2005)	- Wound complications, keloid scar formation, cholesteatoma	Retrospective cohort	N=128	Implants between 1994 and 2003	Children 0.75 to 14 years (Mean= 5 yr) Unilateral CI	2	128	1.56				Bronze
James & Papsin (2004)	- surgical complications	Retrospective cohort	N=25	Implants between 2000 and 2003	Children 0.5 to 1 year Unilateral CI	0	25	0				Bronze

Black, Bailey, Albert, Leighton, Hartley, Chatrath & Patel (2007)	- wound infections, otorrhoea, scar tissue	Retrospective cohort	N=242	Implants between 1992 and 2004	Children 1 to 17 years (Mean=5 yr) Unilateral CI	44	242	18.94				Bronze
Kandogan, Levent & Gurol (2005)	- minor complications that settled with conservative treatment, local care or medication	Retrospective cohort	N=227	Implants between 1999 and 2004	Children 1 to 16 years Unilateral CI	15	227	6.6				Bronze
Bhatia, Gibbin, Nikolopoulos & O'Donoghue (2004)	- swelling, hemotoma, infection settled with conservative treatment	Prospective cohort	N=300	Follow-up time 0.1-14 yrs (mean 4 years)	Children 1 to 16 years Unilateral CI	46	300	15.33				Bronze
Postelmans, Cleffken & Stokroos (2008)	- wound infection, haematoma	Retrospective cohort	N=32	Implants between 2000 and 2005	Children 1 to 11 years (mean=4 yr) Unilateral CI	2	32	6.25				Bronze
Venail, Sicard, Piron et al. (2007)	- Skin flap problems, haematoma, infection	Retrospective cohort	N=272	Implants between 1989 and 2005	Children .9 to 15 years (mean=5 yr) Unilateral CI	7	272	2.57				Bronze
Total						189	2053	9.21				Bronze

Complications Requiring Hospitalization or Additional Surgery

Authors	Definition	Study Design	Sample Size	Time Frame	Population Study Age Sequential/Simultaneous	Event Rate Unilateral CI		CI Rate out of 100	Event Rate Bilateral CI		BICI Rate out of 100	Study Rating
						#	base		#	base		
Stratigouleas, Perry, King, & Syms (2006)	-Required hospitalization and doesn't fall into other category -Mastoiditis, wound infection, migration	Retrospective cohort	N=176	Not reported Follow-up mean 289 days	Adults and Children 1 to 86 years (Mean 27) Unilateral CI	3	176	1.7				Bronze
Yu, Hegarty, Gantz & Lalwani (2001)	-Post Operative Infections requiring antibiotics, or drainage, hospitalization	Retrospective cohort	N=108	Implants between 1991 and 2000	Adults and Children Not Reported Unilateral CI	4	108	3.70				Bronze
Cunningham, Slattery & Luxford (2004)	- Postoperative infections including wound infections, complicated otitis media or mastoiditis -Major if requiring hospitalization, explantation, IV antibiotics	Retrospective cohort	N=272 (children)	Implants between 1993 and 2002	Children 1 to 10 years (Mean 4 yr) Unilateral CI	10	272	3.68				Bronze
Migrov, Muchnik, Kaplan-Neeman & Kronenberg (2008)	-Complications defined as: mastoiditis requiring IV antibiotics	Retrospective cohort study	N=300	Implants between 1993 and 2005	Children 2 to 16 years Unilateral CI	23	300	7.67				Bronze
Gysin, Papsin, Daya & Nedzelski (2000)	- Major complications; flap breakdown, perilymph gusher, cholesteatoma	Retrospective cohort	N=100	Implants between 1990 and 1998	Children 1 to 17 years (Mean= 8 yr) Unilateral CI	4	100	4				Bronze
James & Papsin (2004)	- surgical complications	Retrospective cohort	N=25	Implants between 2000 and 2003	Children 0.5 to 1 year Unilateral CI	0	25	0				Bronze

Black, Bailey, Albert, Leighton, Hartley, Chatrath & Patel (2007)	- mastoiditis, flap breakdown, perilymph gusher, cholesteatoma, failed insertion, electrode migration.	Retrospective cohort	N=221	Implants between 1992 and 2004	Children 1 to 17 years (Mean=5 yr) Unilateral CI	13	221	5.37				Bronze
Kandogan, Levent & Gurol (2005)	- CSF gusher	Retrospective cohort	N=227	Implants between 1998 and 2004	Children 1 to 16 years Unilateral CI	5	227	2.20				Bronze
Bhatia, Gibbin, Nikolopoulos & O'Donoghue (2004)	- Complications requiring surgical intervention or a permanent resulting in disability. Including cholesteatoma, electrode exposure	Prospective cohort	N=300	Follow-up time 0.1-14 yrs (mean 4 years)	Children 1 to 16 years Unilateral CI	7	300	2.33				Bronze
Arnoldner, Baumgartner, Gstoettner & Hamzavi (2005)	- major complications	Retrospective cohort	N=128 (children)	Implants between 1994 and 2003	Children 0.75 to 14 years (Mean= 5 yr) Unilateral CI	0	128	0				Bronze
Postelmans, Cleffken & Stokroos (2008)	- abscess formation near site	Retrospective cohort	N=32	Implants between 2000 and 2005	Children 1 to 11 years (mean=4 yr) Unilateral CI	1	32	3.1				Bronze
Venail, Sicard, Piron et al. (2007)	- electrode shifting, receiver displacement.	Retrospective cohort	N=272	Implants between 1989 and 2005	Children .9 to 15 years (mean=5 yr) Unilateral CI	3	272	1.10				Bronze
Total						73	2161	3.38				Bronze

Facial Palsy or Facial Nerve Paralysis

Authors	Definition	Study Design	Sample Size	Time Frame	Population Study Age Sequential/Simultaneous	Event Rate Unilateral CI		CI Rate out of 100	Event Rate Bilateral CI		BICI Rate out of 100	Study Rating
						# temp (# perm)	base	# temp (# perm)	#	base		
Fayad, Wanna, Micheletto & Parisier (2003)	- Facial nerve weakness or paralysis occurring following CI surgery -Complication occurred between 18 hrs and 19 days post-implant	Retrospective cohort	N=705	Implants between 1980 and 2002	Adults and Children Not Reported Unilateral CI	5 (0)	705	0.71 (0)				Bronze
Stratigouleas, Perry, King, & Syms (2006)	-Facial nerve weakness or paralysis occurring following CI surgery	Retrospective cohort	N=176	Not reported Follow-up mean 289 days	Adults and Children 1 to 86 years (Mean 27) Unilateral CI	4 (1)	176	2.27 (0.57)				Bronze
Migrov, Muchnik, Kaplan-Neeman & Kronenberg (2006)	-Facial nerve paralysis	Retrospective cohort study	N=300	Implants between 1993 and 2005	Children 2 to 16 years Unilateral CI	2 (0)	300	0.66 (0)				Bronze
Gysin, Papsin, Daya & Nedzelski (2000)	- Facial weakness	Retrospective cohort	N=100	Implants between 1990 and 1998	Children 1 to 17 years (Mean= 6 yr) Unilateral CI	1 (0)	100	1 (0)				Bronze
Arnoldner, Baumgartner, Gstoettner & Hamzavi (2005)	- Transient facial palsy	Retrospective cohort	N=128 (children)	Implants between 1994 and 2003	Children 0.75 to 14 years (Mean= 5 yr) Unilateral CI	0 (0)	128	0 (0)				Bronze
Black, Bailey, Albert, Leighton, Hartley, Chatrath	- facial nerve palsy	Retrospective cohort	N=242	Implants between 1992 and 2004	Children 1 to 17 years (Mean= 5 yr)	3 (0)	242	1.24 (0)				Bronze

& Patel (2007)					Unilateral CI							
Kandogan, Levent & Gurol (2005)	- Facial Nerve Paresis	Retrospective cohort	N=227	Implants between 1998 and 2004	Children 1 to 16 years Unilateral CI	1 (0)	227	0.44 (0)				Bronze
Bhatia, Gibbin, Nikolopoulos & O'Donoghue (2004)	- Temporary facial weakness	Prospective cohort	N=300	Follow-up time 0.1-14 yrs (mean 4 years)	Children 1 to 16 years Unilateral CI	2 (0)	300	0.66 (0)				Bronze
Venail, Sicard, Piron et al. (2007)	- Facial Stimulation	Retrospective cohort	N=272	Implants between 1989 and 2005	Children 0.9 to 15 years (Mean= 5 yr) Unilateral CI	1 (2)	272	0.36 (0.74)				Bronze
Total						17 (3)	2460	0.69, [1] (0.12, [0.1])				Bronze

Bacterial Meningitis

Authors	Definition	Study Design	Sample Size	Time Frame	Population Study Age Sequential/Simultaneous	Event Rate Unilateral CI #	Event Rate Unilateral CI base	CI Rate out of 100	Event Rate Bilateral CI #	Event Rate Bilateral CI base	BICI Rate out of 100	Study Rating
Reefhuis, Honein, Whitney, Chamany, Mann, et al. (2003)	- Bacterial Meningitis	Retrospective Cohort and nested case control study	N=4262 (children)	Implants between 1997 and 2002	Children Less than 6 with CI Unilateral CI	26	4264 (10852 person years)	0.240/100 person years				Silver
Summerfield, Cirstea, Roberts, et al. (2004)	- Bacterial Meningitis	Retrospective cohort	N=1851	Implants between 1997 and 2002	Children 1 to 16 years Unilateral CI	0	1851 (2589 person years)	0				Bronze
Total						26	13441 person years	0.193				Bronze

Device Failure (with 5 year time frame)

Authors	Definition	Study Design	Sample Size	Time Frame	Population Study Age Sequential/Simultaneous	Event Rate Unilateral CI #	Event Rate Unilateral CI base	CI Rate out of 100	Event Rate Bilateral CI #	Event Rate Bilateral CI base	BICI Rate out of 100	Study Rating
Maurer, Marangos & Ziegler (2005)	-Hard failures of cochlear implants due to design errors and direct or indirect trauma to the CI site	Retrospective cohort	N=134 (children)	Implants between 1990 and 2001 Follow-up over 2 years	Children Not Reported Unilateral CI	10	134	7.5				Bronze
Venail, Sicard, Piron et al. (2007)	- Device Failure and soft failures	Retrospective cohort	N=272	Implants between 1989 and 2005	Children .9 to 15 years (mean=5 yr) Unilateral CI	17	272	6.25				Bronze
Total						31	464	6.68				Bronze

Appendix G: Questionnaires from Study 4

General Intake Questionnaire

Knowledge Questionnaire

Decisional Conflict Measure

Acceptability Measure

General Information Questionnaire.

I'm the child's: Mother_____ Father_____ Guardian _____

Parent's Age:_____

Parent's Education Level: (check highest level reached)

_____None _____ Elementary School

_____ Secondary School _____ High School Diploma

_____Trade, Technical or Vocational School _____ Some University

_____ Bachelor's or undergraduate degree _____ Master's Degree

_____ Doctorate _____ Other _____

Languages spoken in your home: (check all that apply)

_____ English _____ French _____ ASL _____ Other _____

Age of your child with hearing loss now: _____ Age at diagnosis: _____

Age at first implantation:_____

Degree of Child's Hearing Loss: Severe _____ Profound _____

Centre for Identification: _____

Centre of Intervention: _____

Type of Intervention chosen: _____

Other information that you feel is relevant:

_____

Knowledge Questionnaire

Find out how well this decision aid helped you learn the key facts.
Check ☑ the best answer.

	2nd Implant	Decline 2nd Implant	Both are equal	Don't know
1. Which option has the highest chance of **your child being able to locate sounds in the environment?**	☐	☐	☐	☐
2. Which option has the highest chance of **your child being able to understand speech in a noisy environment?**.	☐	☐	☐	☐
3. Which option has the highest chance of **meningitis?** . . .	☐	☐	☐	☐
4. Which option has the most scientific evidence on long term outcomes?	2nd implant ☐	1st implant ☐	☐	☐

Decisional Conflict Scale

1. Which treatment option do you prefer? Please check ☑ one.

☐ continue with bilateral cochlear implant evaluation for my child
☐ continue using my child's unilateral cochlear implant
☐ Unsure

2. What do you think about the options?

Decisional Conflict Scale ©O'Connor, 1993, revised 2005	**Strongly Agree** [0]	**Agree** [1]	**Neither Agree Or Disagree** [2]	**Disagree** [3]	**Strongly Disagree** [4]
a. I know which options are available to me.	☐	☐	☐	☐	☐
b. I know the benefits of each option.	☐	☐	☐	☐	☐
c. I know the risks and side effects of each option.	☐	☐	☐	☐	☐
d. I am clear about which benefits matter most to me.	☐	☐	☐	☐	☐
e. I am clear about which risks and side effects matter most.	☐	☐	☐	☐	☐
f. I am clear about which is more important to me (the benefits or the risks and side effects).	☐	☐	☐	☐	☐

3. How do you feel about making a decision?

Decisional Conflict Scale ©O'Connor, 1993, revised 2005	**Strongly Agree** [0]	**Agree** [1]	**Neither Agree Or Disagree** [2]	**Disagree** [3]	**Strongly Disagree** [4]
a. I have enough support from others to make a choice.	☐	☐	☐	☐	☐
b. I am choosing without pressure from others.	☐	☐	☐	☐	☐
c. I have enough advice to make a choice.	☐	☐	☐	☐	☐
d. I am clear about the best choice for my child.	☐	☐	☐	☐	☐
e. I feel sure about what to choose.	☐	☐	☐	☐	☐
f. This decision is easy for me to make.	☐	☐	☐	☐	☐
g. I feel I have made an informed choice.	☐	☐	☐	☐	☐
h. My decision shows what is important to me.	☐	☐	☐	☐	☐
i. I expect to stick with my decision.	☐	☐	☐	☐	☐
j. I am satisfied with my decision.	☐	☐	☐	☐	☐

Acceptability of the Decision Aid

My thoughts on the education package on cochlear implants in children

We would like to know what you think about the education package you have just reviewed.

1. Please rate each section, by circling 'poor', 'fair', 'good', or 'excellent' to show what you think about the way the information was presented on:

Understanding Cochlear Implants	poor	fair	good	excellent
Available Options	poor	fair	good	excellent
Criteria for Bilateral CIs	poor	fair	good	excellent
Evidence About Bilateral CIs	poor	fair	good	excellent
Risks with Bilateral CIs	poor	fair	good	excellent
Benefits with Bilateral CIs	poor	fair	good	excellent
Risks without Bilateral CIs	poor	fair	good	excellent
Benefits without Bilateral CIs	poor	fair	good	excellent

2. The length of presentation was (*check one*)

|__| too long
|__| too short
|__| just right

3. The amount of information was (*check one)*
|__| too much information
|__| too little information
|__| just right

4. I found the presentation (*check one*)

|__| slanted towards going ahead with bilateral cochlear implant surgery
|__| slanted towards taking staying with unilateral cochlear implant use
|__| balanced

5. Did you find this decision aid useful when you were making your decision about bilateral cochlear implants for your child?
|__| Yes
|__| No

Comments:

6. What did you think of the rest of the personal worksheet?
Did it make the decision
|__| easier, or
|__| more difficult?
Comments:

7. Do you think we included enough information to help a parent decide about bilateral cochlear implants?
|__| Yes
|__| No
Comments:

8. What did you like about the decision aid and worksheet?

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9. What suggestions do you have to improve the decision aid or worksheet?

Acceptability

Appendix H: Manuscript Submission and Decision Letters

Manuscript 1: First Page of Publication

Manuscript 2: Decision Letter: International Journal of Audiology

Manuscript 4: Submission Letter: International Journal of Audiology

An Assessment of Parents' Decision-Making Regarding Paediatric Cochlear Implants

Un examen du processus décisionnel des parents concernant l'implantation cochléaire pédiatrique

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Abstract

Parents of children with severe to profound hearing loss have to make a number of fundamental decisions for their children. These decisions include communication and amplification options. In particular, the parents must decide whether and when their child will receive cochlear implants, and whether these will be implanted unilaterally or bilaterally. The objective of this study was to describe the decision-making needs of parents making the cochlear implant decision for their children. Semi-structured interviews were conducted with eight parents and eight cochlear implant team members at a Canadian cochlear implant centre to document parental and clinician recollections and opinions of the decision-making process related to a unilateral or bilateral cochlear implantation. The results demonstrated that the decision to go ahead with a cochlear implantation was consistently based on the parents' preferences for spoken communication for their children. Parents reported satisfaction with the cochlear implant decision-making process. Two of eight parents felt that additional information on unilateral cochlear implantation risks and benefits should have been provided. Four of eight parents described how more information on the experiences of other families would have been helpful for their decision. Parental and clinical perceptions of the bilateral implantation decision were highly variable. All parents stated that additional information on bilateral cochlear implantation was needed. Based on the results of the interviews, it is concluded that there is a need for information and resources for bilateral cochlear implantation decision-making.

Abrégé

Les parents d'un enfant ayant une perte auditive de degré sévère à profond ont des décisions fondamentales à prendre pour leur enfant. Ces décisions comprennent des options de communication et d'amplification. Plus spécifiquement, ils doivent décider si leur enfant recevra un ou deux implants cochléaires et à quel moment. La présente étude visait à décrire les besoins des parents dans le processus décisionnel de l'implantation cochléaire pour leur enfant. Des entrevues semi-structurées ont été menées auprès de huit parents et de huit membres d'une équipe d'un centre canadien d'implantation cochléaire pour documenter ce dont se souviennent les parents et les cliniciens et leur avis concernant la décision menant à une implantation unilatérale ou bilatérale. Les résultats montrent que le fait de choisir l'implantation cochléaire était systématiquement fondé sur la préférence des parents pour la communication orale de leur enfant. Les parents ont dit être satisfaits du processus de décisions liées à l'implantation cochléaire. Deux des huit parents trouvent qu'ils auraient dû recevoir davantage d'information sur les avantages et les risques de l'implantation unilatérale. Quatre des huit parents ont dit qu'ils auraient trouvé utile d'avoir davantage d'information sur l'expérience d'autres familles avant de prendre leur décision. La perception des parents et des cliniciens concernant le choix de l'implantation bilatérale variait considérablement. Tous les parents ont précisé qu'ils

03-Dec-2008

Dear Ms. Johnston:

Thank you for sending your revised manuscript. Based on the revisions made, it is a pleasure to accept it in its current form for publication in the International Journal of Audiology.

Your manuscript is being sent to the publisher for final production. Page proofs will be sent to you during the production process. It is very important that you read your page proofs carefully and return them promptly so that your paper will be processed on schedule. Currently there is a 4-5 month delay in having accepted papers in an issue of the journal. However, the finished article will appear in electronic form and all readers will be notified through SARA that it is available shortly after you return your page proofs. The electronic posting represents a formal publication.

Thank you for your fine contribution. On behalf of the Editors of the International Journal of Audiology, we look forward to your continued contributions to the Journal.

Sincerely,

Ross J. Roeser, PhD
Editor-in-Chief
International Journal of Audiology

March 17, 2009

J. Cyne Johnston
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Dear Volta Review Editorial Staff,

Please find attached a manuscript entitled, "The development and piloting of a decision aid for parents considering sequential bilateral cochlear implantation for their child with hearing loss." I hope that you consider this manuscript for publication in the *Volta Review*.

This original research has not been previously published and is not under review elsewhere. Ethical approval for this research has been granted by both the University of Ottawa (Ethics file # H 08-08-15) and Children's Hospital of Eastern Ontario Research Ethics Boards (Protocol #08/57X).

Please contact me if you have any concerns or questions. I look forward to receiving a response from the journal regarding the possibility for publication.

Sincerely,

J. Cyne Johnston