

Outcomes That Matter for Children With Severe Multiple Disabilities who use Cochlear Implants: The First Step in an Instrument Development Process

Des Retombées qui Comptent Pour des Enfants Ayant des Handicaps Multiples et qui Utilisent des Implants Cochléaires : La Première Étape dans le Processus de Développement d'un Instrument

KEY WORDS

COCHLEAR IMPLANT

MULTIPLE DISABILITY

COMPLEX NEEDS

OUTCOMES

BENEFIT

PARENT REPORT

INSTRUMENT DEVELOPMENT

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Abstract

The goal of the current study was to begin an instrument development process for a tool that would capture outcomes of cochlear implantation for children with severe multiple disabilities that parents report matter most to themselves and their children. Participants comprised mothers of children who had profound hearing impairments and additional severe disabilities, and who had received cochlear implants within the last 10 years. Participants completed one to four interviews with a structured interview question set designed to capture participants' perceptions and observations of their child's communication, listening, behaviour, social interactions, and device management post-cochlear implantation.

Reported gains following cochlear implantation included increased: (a) awareness of sound in general, (b) receptive and expressive communication, (c) connectedness within the family and broader community, and (d) quality of life. Themes related to 'listening', 'expressive communication', and 'family systems' were the most frequently reported by participants; however, when asked to rank themes with respect to their importance to themselves and their child, 'child affect', 'connectedness/inclusion', and 'receptive communication' were ranked the highest. These results will form the basis for instrument development.

Abrégé

Le but de cette étude était d'amorcer le processus de développement d'un outil sensibles aux retombées de la pose d'un implant cochléaire chez des enfants ayant des handicaps multiples sévères, qui, au dire des parents, ont le plus d'importance pour eux et leurs enfants. Les participants étaient des mères d'enfants ayant une surdité profonde et d'autres handicaps sévères, et qui avaient reçu un implant cochléaire depuis moins de dix ans. Les mères ont participé à des entrevues (entre une et quatre) avec une série de questions structurées conçues pour recueillir leurs perceptions et observations de leur enfant concernant sa communication, son écoute, son comportement, ses interactions sociales et sa gestion de l'appareil après la pose de l'implant cochléaire.

Les améliorations rapportées après l'implantation cochléaire comprenaient une augmentation de (a) la conscience des sons en général, (b) de la communication au plan réceptif et expressif, (c) du lien au sein de la famille et avec la communauté, et (d) de la qualité de vie. Des thèmes reliés à « l'écoute », à « la communication au plan expressif » et aux « systèmes familiaux » ont été le plus souvent notés par les participants; toutefois, quand on leur demandait d'évaluer les thèmes par ordre d'importance pour eux et leurs enfants, ce sont « l'affect de l'enfant », « la connectivité/inclusion » et « la communication au plan réceptif » qui ont été classés les plus hauts. Ces résultats formeront les fondements pour le développement de l'instrument.

Cochlear implantation (CI) has proven to be an important treatment option for children with severe-to-profound hearing loss. However, 30-40% of children with severe-to-profound hearing loss have additional disabilities, many of which are also severe in nature, and CI has been inconsistently available to these children (Edwards, 2007; Johnson & Wiley, 2009). A number of factors have limited CI as a treatment option for this population including: (a) lower expectations in listening and spoken language outcomes, (b) increased surgical and medical risks, (c) possible outcome disappointments for families, (d) challenges programming the device, and (e) lack of adequate assessment and tools for monitoring progress to aid clinical decision-making (Berrettini et al., 2008; Johnson & Wiley, 2009; Schramm, Fitzpatrick, & Seguin, 2002; Trimble et al., 2008; Wiley, Meinzen-Derr, & Choo, 2008)

The goal of the present study was to undertake an instrument development process to assist in reliably documenting outcomes for children with severe multiple disabilities who use cochlear implants. Our goal evolved in response to a clinical need identified by the authors who provide services to children with severe multiple disabilities and their families following CI. Parents described positive changes in their children following CI yet the improvements were either too subtle to be captured or not captured at all by the standardized assessment instruments currently used to monitor CI effectiveness.

Background

Cochlear implantation has become a common and widely accepted treatment option for children with profound hearing loss (Filipo, Bosco, Mancini & Ballantyne, 2004). Advancements in CI technology and success in rehabilitation have led to opening CI to additional groups including infants, and children with disabilities in addition to profound hearing loss. However, there is currently no widespread consensus in the literature or among CI centres regarding the implantation of children with multiple disabilities (Berrettini et al., 2008; Johnson & Wiley, 2009). For children where profound hearing loss is the sole disability, CI decisions are based on expected benefits to audition, speech, and language acquisition; however, what constitutes a benefit or how it should be quantified remains questionable for children with multiple disabilities. Compared to children with profound hearing loss as a sole disability, children with multiple disabilities achieve lower scores on measures of audition, speech perception, speech intelligibility, and language acquisition following CI (Berrettini et al, 2008; Johnson & Wiley, 2009; Waltzman, Scachuns, & Cohen,

2000). Notwithstanding poor scores on standardized assessment instruments, improved quality of life, increased connectedness and social interactions along with greater interest in the environment have been reported for children with multiple disabilities following CI (e.g., Berrettini et al, 2008; Filipo et al, 2004; Waltzman et al, 2000; Wiley, Jahnke, Meinzen-Derr, & Choo, 2005) and were reported as both relevant and significant to families (Hermannova, Phillips, O'Donoghue, & Ramsden, ND; Johnson & Wiley, 2009).

For example, analyzing post-CI outcomes for children with and without additional disabilities Waltzman et al. (2000) found that auditory skill development was not as immediate or favorable for the children with multiple disabilities. However, anecdotal observations revealed positive benefits, such as increases in social interaction and connectedness to the environment. Waltzman and colleagues stated

“the ability to provide greater access to the surrounding environment to a child otherwise deprived of that opportunity, should, in its own right, be considered as an achievement despite the lack of immediate gratification obtained from excellent results on test measures” (p. 334).

These authors suggested that although it is currently challenging to measure improvements in auditory and communication skills, social interaction, and connectedness for children with multiple disabilities, such changes are noticeable and reported with regularity. Similarly, Filipo et al. (2004), Donaldson, Heavener, and Zwolan (2004), and Wiley et al. (2005) all report positive benefits of CI for children with multiple disabilities including improved quality of life, speech perception, communication skills, and self sufficiency, along with greater awareness of environmental sounds, increased attentiveness and interest in their environment.

The accumulated evidence, although promising, does not yet provide clinicians consistent procedures to support CI candidacy decision-making or for measuring outcomes for children with multiple disabilities. Additionally, studies that have attempted to analyze post-CI outcomes for this population include children with a wide array of additional disabilities (e.g., cerebral palsy, learning disability, autism, cognitive delay, oral motor difficulties), and ranging from mild to severe diagnoses, making it difficult to draw conclusions about outcomes. Thus, in the present study we focus exclusively on post-CI outcomes for children with severe additional disabilities. For the purpose of this study, this meant that hearing loss was not the most significant influence on the child's development.

Establishing and quantifying the benefits of CI for this population is a critical issue due to the reported unsuitability of standardized instruments to capture the changes observed for these children. Moreover, the few standardized measures that do evaluate benefits in daily life (Berrittini et al., 2008; Waltzman et al., 2000) have limitations for this population. *The Glasgow Children's Benefit Inventory* (Kubba, Swan, & Gatehouse, 2004) aimed at capturing general quality of life benefits specific to children following an operation or medical intervention, lacks specificity in relation to the contribution of access to sound that participants described for their children with multiple disabilities and for themselves. While the *Champion's Evaluation Profiles* (Herrmannova, et al., n.d.) a recently developed tool for documenting both process and outcomes for pediatric cochlear implant users with additional disabilities, is valuable as a guide in the process of evaluating children with multiple disabilities for CI candidacy, it has not been subjected to reliability studies and thus, does not lend itself to measurement at present.

Parent identified benefits due to CI reported in previous research and observed in our own clinical practice form an important source of evidence related to both CI candidacy and outcome benefits for children with severe multiple disabilities (Herrmannova, et al., n.d.; Johnson & Wiley, 2009; Ritter & Peters, 2003). Measures need to be developed that reliably capture the benefits that parents witness, experience, and value on a daily basis (National Roundtable Steering Committee, 2011).

Method

Participants

Participants comprised mothers of seven children who had (a) profound hearing impairments, (b) received cochlear implants within the last 10 years, and (c) additional severe disabilities. Participants came from diverse social backgrounds, their first language was English, four had completed high school and three had post-secondary education. One participant resided in rural Alberta and six participants resided in the metropolitan area of Edmonton, Alberta, a large Western Canadian city. Edmonton has a tertiary rehabilitation facility that provides comprehensive multidisciplinary assessment and treatment services to families of children with hearing impairment, including determination for candidacy and CI follow-up.

Participants' children were all diagnosed with severe multiple disabilities and their hearing loss was a secondary diagnosis in terms of its influence on their overall functioning. Cochlear implantation for participants' children occurred between 2000 – 2009. This group of children represented approximately 80% of children with severe multiple disabilities with cochlear implants in northern Alberta at the time the study was conducted. Children were involved in community placements and were followed by a variety of assessment and follow-up clinics for their medical and developmental needs. In addition, the second author followed all seven children for communication habilitation support related to CI function. Table 1

Table 1. Characteristics of Participants' Children

| Participant | Child Age at Cochlear Implant ^a | Implant Type ^b | Additional Diagnoses |
|-------------|--|---------------------------|---|
| 1 | 1;8 | AB - Clarion | Autism |
| 2 | 2;4 | CA - Freedom | Severe neuromotor delay (unknown cause) |
| 3 | 1;6 | AB - 1.2 | Autism; Seizure disorder |
| 4 | 1;8 | CA - Freedom | Autism; Chromosome 10 deletion |
| 5 | 2;4 | AB - Hi Res 90K | Pervasive Developmental Disorder |
| 6 | 3;5 | AB - Hi Res 90K | Blind; Peroxisomal disorder, |
| 7 | 5;7 | CA - N-24 | Cerebral Palsy (quadriplegic); Pervasive Developmental Disorder |

Note. a = Age is expressed in years;months.

b = Cochlear Implant Company: AB -Advanced Bionics; CA-Cochlear Americas

describes relevant characteristics of the participants' children.

Recall the aim of our study was to begin development of an instrument that captured outcomes parents' consider significant related to CI for children who have severe multiple disabilities, which in turn influenced our methodological choice, development of the structured interview question protocol, and procedures. Our methodological approach was qualitative and informed by grounded theory (Lincoln & Guba, 1985) because a main tenant of the grounded theory approach supports the discovery and description of real concerns and perceptions of participants. Thus, it was well suited to our aim.

Procedure

Interview Protocol: A structured interview question set was developed based on previous research related to outcomes of CI coupled with the second author's extensive clinical experience working with families of children with hearing impairment and CI. Questions were specifically tailored to capture participants' perceptions and observations of their child's (1) communication, (2) listening, (3) behaviour, and (4) social interactions, post-cochlear implantation. Additionally, participants were asked about managing the device, and offered the opportunity to add observations that were not captured in the interview question set (see Appendix for structured interview questions).

Interview Procedure: Participants granted consent to complete the interview question set and completed one to four interviews (face-to-face or written). The second author conducted the face-to-face interviews as part of scheduled visits (home or clinic). The initial interview question sets were administered to participants 1, 2, 4, 6, and 7 following their child's implantation. For participants 3 and 5, whose children received their implants prior to the start of the study, initial interview question sets were administered upon enrolment in the study. Interviews two-through-four were conducted at convenience intervals, that is, with available participants, and at times the participants were accessible. Interviews were continued until data saturation was reached, resulting in a total of 18 interviews; 15 face-to-face and three written. Data saturation is the process by which sufficient data is obtained so that there is repetition of previously collected information, which allows for variation in participant responses to be accounted for and understood (Morse, 1994). Table 2 shows the number and interval of interviews completed for each participant.

During each interview the researcher took extensive field notes of the participants' responses and sought confirmation or clarification before concluding the interview that her notes accurately reflected participants' responses. A written summary of participants' responses was compiled after each interview (face-to-face and written) and participants

Table 2. Number and Intervals of Participant Interviews

| Participant | Total Number of Interviews | Interview 1 | Interview 2 | Interview 3 | Interview 4 |
|-------------|----------------------------|-------------|-------------|-------------|-------------|
| 1 | 2 | CA: 7;4a W | CA: 12;6 | | |
| 2 | 4 | CA: 1;6 | CA: 2;4 W | CA: 2;10 | CA: 3;5 |
| 3 | 1 | CA: 10;6 | | | |
| 4 | 3 | CA: 0;5 W | CA: 0;11 | CA: 0;17 | |
| 5 | 1 | CA: 0;2 | | | |
| 6 | 4 | CA: 0;3 | CA: 0;5 | CA: 0;16 | CA: 4;0 |
| 7 | 3 | CA: 0;7 | CA: 1;7 | CA: 2;11 | |

Note. CA = Child age at time participant completed interview
 a = Age is expressed in years;months
 W = Participant completed interview protocol in writing

were asked to review the summary to confirm accuracy and provide clarification if necessary.

Following the content analysis approach of Lincoln and Guba (1985) and Neuendorf (2002), as data was collected, classification themes were developed whereby the content of participants' responses were grouped into categories of similar responses, after which major themes were derived from the categories, and illustrative participant response examples extracted. Participants were asked to confirm that both the categories and themes accurately represented their responses. All participants were in agreement with the final set of categories and themes derived from their responses. Once agreement was achieved for major themes, the number of responses participants provided within each theme was tallied. Finally, participants were asked to rank the importance of each theme for themselves and their child.

Quality and Credibility

In qualitative research, the equivalents of the quantitative research concepts of validity and reliability are quality and credibility. "The credibility of qualitative enquiry is especially dependant on the credibility of the researcher because the researcher is the instrument of data collection and the center of the analytic process" (Patton, 1980, p. 461). The second author has in excess of 35 years experience working with families of young children with hearing loss along with a clinical and academic background related to instrument development and program evaluation (Ritter, 1997).

Quality and credibility of the data and interpretation was further enhanced by triangulation of sources (Patton, 1980) which involves data collection from multiple sources as a means of cross-checking regularities in the data, and overcoming intrinsic biases (Patton & Westby, 1992). Multiple data sources within the present study included seven participants' whose children varied in disabilities and age at the time of interviews as well as field notes summarizing observations and discussion with participants (see Tables 1 and 2).

Credibility of our findings was further enhanced via member checks throughout the study. Member checks, also referred to as respondent validation, and informant feedback, is a technique used to improve accuracy, quality, credibility, transferability, and validity of a study (Schwartz-Shea, 2006). Member checking was completed during all phases of the study, during and after interviews, and following classification and interpretation of participant responses, thus allowing participants multiple opportunities to critically analyze our interpretations. The use of

member checks throughout an entire study serves to decrease the incidence of data inaccuracy and incorrect interpretation of the data (Schwartz-Shea, 2006).

Results

Our goal was to gain information to support the development of an instrument that captured outcomes of CI for children with severe multiple disabilities that participants reported were substantive to their child, themselves, and their families. To accomplish this goal we first conducted a content analysis (e.g., Lincoln & Guba, 1985; Neuendorf, 2002) of the repeated structured interview question sets to identify the themes in participants' observations and comments. Seven distinct themes were identified: (a) listening (non-linguistic), (b) expressive communication, (c) family systems outcomes, (d) receptive communication, (e) connectedness/inclusion (f) child affect, and (g) challenges. Themes, response categories that characterized individual themes, and illustrative participant exemplars are displayed in Table 3. In the next section we describe each of the themes and provide examples of participant observations and comments related to the particular theme.

Themes

Listening (non-linguistic). A total of 40 participant responses related to the theme, listening to non-linguistic sounds, that is, environmental sounds and tone of voice. Participants described children reacting to environmental sounds after CI activation, such as laughing when sounds were heard or attempting to locate the source of a sound. The importance of access to sound for such purposes is shown in the comment by Participant 6 *"It's [sound] his WHOLE world, because he's totally blind and immobile. He can't compensate through vision or movement, he can't entertain himself, sound IS his entertainment."* Another participant spoke of the importance of their child's newly acquired ability to orient to sound, *"Our daughter will turn to look at me when I make one of her favorite sounds. It is the only time she orients toward me and it literally thrills me when she does it. Our daughter is not terribly relational, so to get that interaction from her is very meaningful to me. When she looks at me, even it is really only to see the sound, so to speak, she looks into my face and into my eyes and I get a chance to love her with my eyes. I use her ears, to get to her eyes, to get to her heart."* Participants also indicated that CI gave children access to sound as a means of enjoyment, be it music or other sounds heard within the child's environment, including the child's own voice.

Expressive Communication. Within this theme participant responses illustrated children's use of voice, sign, pictures, words and gestures. Similar to Listening,

participants provided a total of 40 responses related to this theme. Participant 5's comment, "He now knows for sure that his voice has power. He has differentiated tone of voice for a long time. But now it seems more deliberate. It's easier now to tell what his mood is from another room," reveals not only that the child was using his voice more deliberately as a means of communication but that

the parent was understanding the communicative intent of the child's vocalizations. Participants also commented that CI resulted in their child actively monitoring their vocal loudness and imitating some sounds upon request. Further, participants felt that they could make more demands for expressive communication from their child following CI.

Table 3. Description of Themes, Response Categories, and Participant Exemplars

| Theme Definitions | Response Categories | Participant Exemplars |
|---|---|---|
| <p>Listening (non-linguistic): Child reacts appropriately to some environmental sounds or voice (not spoken words); enjoys music</p> | <ul style="list-style-type: none"> • Awareness/responsiveness to environmental sounds • Child responds to voice from another room • Child loves/appreciates music • Child laughs at funny sounds • Child reliably communicates when CI signal is bad or off • Child searches for environmental or voiced sound • Child removing CI headpiece is a good indicator of lack of engagement • Child independently replaces headpiece | <ul style="list-style-type: none"> • <i>When he lost his hearing nothing was funny anymore. Now we have fun during meals.</i> • <i>Taking his head piece off himself is a good way to both gauge quality of sound and engagement. It is important communication.</i> |
| <p>Expressive Communication: Child is able to express self with voice, signs, pictures, words or gestures</p> | <ul style="list-style-type: none"> • Meaningful communication regardless of modality (behaviour/gesture, sign, oral) • Child makes non-verbal efforts to engage others • Child's vocal volume moderated to socially acceptable level • Increase in amount and variety of vocalizations post-CI • Child uses inflection with meaning • Child imitates vocal patterns/melody • Child imitates C-V combinations • Expressive communication is inconsistent | <ul style="list-style-type: none"> • <i>We now ask her to ask for something, "Do you want the ball? Say bah." We wait her out a couple of minutes. Asking her a few times to say, "Bah" if she wants the ball. She purses her lips together and emphatically says, "Bah". My son plays this game with her now.</i> |
| <p>Family Systems Outcomes: Family interactions are easier because child has access to sound</p> | <ul style="list-style-type: none"> • Family feels more 'normal' to parent • Less effort for parent and siblings to include child in family interactions • CI technology easier to manage than hearing aids • Ability to interact through sounds brings parent joy • Family has more fun with child • Parents now feel that they have done everything possible to support child's optimal function | <ul style="list-style-type: none"> • <i>One of the best things is how far away he can hear. Now I can be washing dishes at the sink and be talking to him and he understands. That is a freedom no money can buy. He is a high needs kid; being able to engage and support him from a distance is HUGE.</i> • <i>You used to have to work so hard just to get her to look at you. Now it is effortless on our part, she's on her own steam.</i> |

| | | |
|--|--|--|
| <p>Receptive Communication: Child is able to understand at least some of what you say</p> | <ul style="list-style-type: none"> • Child turns consistently to name • Child continues to progress in auditory comprehension • Child responds with appropriate actions to familiar songs • Child follows some routine verbal directions • Child demonstrates some open set auditory comprehension • Comprehension is inconsistent | <ul style="list-style-type: none"> • <i>He objects when you are talking to another adult about him as if he wasn't there.</i> • <i>I mentioned the S-LP's name in the midst of a conversation and he sat up and clapped. It seemed like a return to the time when he understood, and could demonstrate that he did. We were trying to decide at that point if we should send him back to school after a long illness. He had one day back, saw his S-LP and I was telling his father about his day, and he perked right up and clapped. We decided to send him back to school.</i> • <i>"Our daughter will turn to look at me when I make one of her favourite sounds. It is the only time she orients toward me and it literally thrills me when she does it. Our daughter is not terribly relational, so to get that interaction from her is very meaningful to me. When she looks at me, even it is really only to see the sound, so to speak, she looks into my face and into my eyes and I get a chance to love her with my eyes. I use her ears, to get to her eyes, to get to her heart.</i> |
| <p>Connectedness/ Inclusion: Child is better able to connect and be included within the family or community because of access to sound with CI.</p> | <ul style="list-style-type: none"> • Child's participation and overall inclusion in family activities/interactions; tolerance of new experience, social interaction • Others interact more with child simply because they know the child can hear • Child more able to tolerate new experiences • Child's ability to interact with others enhanced • Child more present, more engaged • Broadened social sphere, not restricted to signers • Child more connected to family and peers | <ul style="list-style-type: none"> • <i>Before her implant it was extremely hard to first capture her attention and then secondly, hold her attention. Action could be happening all over the house and she would be sitting facing the wall completely missing everything and not taking part. Very shortly after her implant we were noticing her looking for where sounds were coming from, watching people's lips and faces more and paying attention.</i> • <i>She may not be a star in society's standard, but as her mother I see her as having woken up.</i> |
| <p>Child Affect: Child is happier because of access to sound with CI</p> | <ul style="list-style-type: none"> • Reduction in self stimulating behaviour • Less effort for child to participate in family and community interactions • Better quality attention • Child happier overall | <ul style="list-style-type: none"> • <i>The family gets more joy in interacting with her, life is more meaningful for her and for us. It's a shame when a dollar figure is put on it.</i> • <i>He seems to be immensely happy to be socially involved in others' lives. It was too hard to get him to focus, and hold focus (before the implant). Sound has definitely changed this.</i> |
| <p>Challenges: Difficulty managing equipment or in keeping device on child</p> | <ul style="list-style-type: none"> • Child removes headpiece to get attention or to protest • Child can't wear CI when doing some favorite things, e.g. roughhouse or trampoline play | <ul style="list-style-type: none"> • <i>The headpiece often falls out in the car.</i> • <i>The headpiece falls off during rough house play.</i> • <i>[Managing the device] isn't too big a problem.</i> |

Family Systems. Twenty-six responses were classified within the Family Systems theme. Participants frequently stated that after their child's CI family interactions and family life in general felt more 'normal.' For example, *"We could not pleasantly go to a restaurant, people would look at her for her loudness and we felt that we were always bothering people. We often decided not to go places because of this. Almost immediately after her implant she toned down to normal volumes when her implant was on. This one factor alone to our family was worth her getting the implant."* Another participant stated *"One of the best things is how far away he can hear. Now I can be washing dishes at the sink and be talking to him and he understands. That is a freedom no money can buy. He is a high needs kid, being able to engage and support him from a distance is HUGE."* Additionally, participants described a reduction in the effort needed by family members to include the child with severe multiple disabilities in family interactions, along with feeling like they had done everything they could to help their child.

Receptive Communication. A total of 20 participant responses related to the theme of Receptive Communication. Responses exemplified children's understanding of verbal communication, even if inconsistent, orienting toward spoken sounds, and responding to their names or the names of familiar people. The following participant observation exemplifies the impact of CI on the receptive communication *"Our daughter seems to be getting the point faster with not as much need of repetition. On a funny note she also understands when we tell her that she is being silly, goofy, or a 'stinker.' She will do something silly to get our attention and then we comment verbally, saying, You're being silly! and she promptly signs 'stinker,' and starts to laugh."*

Connectedness/Inclusion. Participant comments described an increase in the children's interactions with both family and community, *"She's more present, more responsive, more connected to her environment. She's no longer alone in the room."* An intent on their child's part to connect with people in their environment was also noted. Participants also reported an increased inclusion of their child by others. Adults and children interacted more often with their child because they knew the child could now hear them *"Awareness of others has increased, others outside of her family, e.g. at church people will talk to her and she will smile. It takes less specialized effort to engage her so a wider circle of people can do it successfully."* Thus, a child's social sphere was broadening not only because others felt the child could hear them but also because now community members

needed no specialized skills to interact successfully with the child (i.e., sign language).

Child Affect. A total of 19 participant responses were classified within the Child Affect theme. Participants described children's increased happiness as being due to access to sound as shown in the statement, *"He seems to be immensely happy to be socially involved in others' lives. It was too hard to get him to focus, and hold focus [before the implant]. Sound has definitely changed this."* Changes also included a reduction in self-stimulating behaviours and improved attention.

Challenges. Responses related to Challenges included difficulties keeping the child from removing the device, particularly in the car and concerns about activities during which the device could not be worn. A total of six responses were reported by participants as Challenges. To overcome the child's initial resistance to the CI one participant reported *"At first she wanted nothing to do with sound and quickly learned to take it [the CI] off. We got a hat for her and my mom attached straps [helmet style] and we tied it under her chin."* Another participant kept her child's hair short in order to improve headpiece retention.

Importance of Themes

After themes were derived and collated from categories, participants were asked to rank each theme in terms of importance to themselves and their child. This step was necessary to determine which themes mattered most to the participants. Six out of seven participants provided rankings. One participant did not rank the themes because she felt that all of the themes were of equal importance.

A rank of 1 indicated 'most important' and a rank of 7 indicated 'least important.' Rankings were summed and the modes were obtained for each theme. The six participants' rank ordering from most to least important amongst themes was as follows: (1) Child Affect, (2) Connectedness/Inclusion, (3) Receptive Communication, (4) Family Systems, (5) Listening, (6) Expressive Communication, and (7) Challenges. The total number of responses for each theme and rank ordering of themes by importance to participants are displayed in Table 4.

As can be seen in Table 4 the rank ordering of themes by importance to participants did not relate to the number of propositions participants described for each of the seven themes. For example, the two themes for which participants provided the highest number of responses, Listening and Expressive Communication, were ranked fifth and sixth respectively, while themes (Child Affect, Connectedness/Inclusion, Receptive Communication) with half the number of reported

Table 4. Total Number of Responses and Participant Ranking of Importance of Identified Themes

| Theme | Total number of participant responses | Participant ranking in order of importance |
|----------------------------|---------------------------------------|--|
| Child Affect | 19 | 1 |
| Connectedness/Inclusion | 19 | 2 |
| Receptive Communication | 20 | 3 |
| Family Systems | 26 | 4 |
| Listening (non-linguistic) | 40 | 5 |
| Expressive Communication | 40 | 6 |
| Challenges | 9 | 7 |

responses were ranked the three most important to participants.

Discussion

The benefits of CI for children with severe-to-profound hearing loss have been well documented. However, the lack of consensus in the literature and at CI centres regarding the benefits of CI for children with multiple disabilities has resulted in the inconsistent availability of this treatment option for this population. Further, the heterogeneity of this population and small numbers of children with similar disabilities limits what we currently understand about their outcomes (Wiley et al., 2005).

One of the major obstacles facing clinicians and implant teams is the challenge of measuring improvements for children with multiple disabilities following CI because these children are not only difficult to test with standard speech perception and language tests but as many researchers (e.g., Berrettini et al., 2008; Johnson & Wiley, 2009; Waltzman et al., 2000) attest, formal tests do not adequately capture gains within this population either because the changes are too subtle to be captured by these tests or the tests do not measure changes observed with these children at all. For example, the following observations *'my child is happier,' 'including my child is much easier than it was,' or 'my child's response to sound brings me joy'* are not captured yet considered foundational to parenting success, and therefore to parent well-being. These ideas require specific attention in the instrument development process particularly for children with multiple disabilities.

The current literature indicates that after implantation the majority of children with multiple disabilities do make progress in speech perception and communicative skills although at considerably lower rates compared to the progress achieved by children with hearing loss as a sole disability. A growing number of studies all report a variety of important benefits of CI for children with multiple disabilities resulting from greater access to the surrounding environment which include improved awareness to sounds, communication skills, attentiveness and interest in the environment, social interaction and connectedness, and quality of life (e.g., Berrettini et al., 2008; Donaldson et al., 2004; Filipo et al., 2004; Waltzman et al., 2000; Wiley et al., 2005). While these results have been consistently reported for children with an array of disabilities, severity of additional disabilities was not considered. In the present study participants' children all presented with severe multiple disabilities and the reported gains were strikingly similar to previous studies, that is, awareness to sound in general, receptive and expressive communication, connectedness within the family and broader community, and quality of life.

In our attempt to understand parents' perspective in interpreting outcome benefits of CI a unique contribution important to the instrument development process resulted from asking participants to rank the broad themes they identified in relation to their importance to themselves, and their child. If the number of times a theme was cited by participants were taken to be representative of its level of importance to participants, the interpretation of benefits as perceived by parents would have been in error. As

shown in Table 4 the number of times a theme was mentioned from most to least was as follows: listening, expressive communication, family systems, receptive communication, child affect, connectedness/inclusion and challenges. However, when these themes were ranked by importance to the participant the order was as follows: child affect, connectedness/inclusion, receptive communication, family systems, listening, expressive communication, and challenges.

This finding is key in informing the practice of cochlear implant clinicians when working with families of children with multiple disabilities and severe to profound hearing loss. Historically, cochlear implant clinicians and implant teams focus on receptive and expressive auditory oral communication outcome benefits and on the possible challenges the cochlear implant procedure may present with this population. The fact that 'expressive communication' and 'challenges' were ranked the least important by participants and that 'child affect' and 'connectedness/inclusiveness' were ranked as the two most important themes suggest that cochlear implant clinicians working with these families and children need to be aware that what is considered important regarding clinical outcomes is different from their traditional frames of reference. Furthermore, clinicians need to be cognizant that their communication interactions with parents and caregivers about benefits and challenges influence the scope of questions that are asked and answered as a consequence of the clinician-client relationship. Thus, it is no surprise that the highest number of participants' comments and observations related to receptive and expressive communication, given that clinicians ask questions about these topics and parents understand that this is what clinicians want to hear about. In order to provide the best services and opportunities for these children clinicians must be willing to broaden the scope of their discussions regarding benefits beyond those conventionally considered (i.e. receptive and expressive communication) to understand both the specific needs of this population and family perceptions of benefit. Children with severe multiple disabilities challenge a clinician's ability to predict the likely benefit of CI. We concur with Wiley et al. (2005), that observations reported by parents of children with severe multiple disabilities are essential in evaluating and monitoring the benefit of CI. Parents are able to observe capabilities in their child in a variety of settings that are less structured and more familiar for the child. Therefore more skills are likely to be observed than in a structured unfamiliar clinic environment (Wiley et al., 2005). Although parental observation may introduce some bias it has the potential to add important information

regarding child function in everyday situations (Berrettini et al., 2008). In fact, the 2011 National Roundtable Steering Committee on the health of families of children with disabilities concluded that it is necessary to embed the values and well being of caregivers into every aspect of service delivery for children with disabilities. The growing body of research makes clear the need for instruments that will allow the benefits observed and valued by parents and caregivers to be captured and become part of the candidacy and outcome benefit analysis for children with multiple disabilities.

Similar to formal tests, cost utility studies (e.g., Bergeron, 2003; Cheng et al., 2000; Francis, Koch, Wyatt, & Niparko, 2000) do not take into account the family's perspective and thus, do not adequately capture meaningful benefit for children with multiple disabilities, particularly for severely involved children.

Additionally, cost utility studies for children with disabilities focus on the cost to educate, yet many children with severe multiple disabilities will not participate in general education programming leading to questions regarding what cost utility might constitute for this population. Such questions remain unanswerable at this time; however, with the development and application of instruments that adequately capture meaningful changes reported by parents in the present and previous studies, these important issues may begin to be addressed.

Limitations

There are specific limitations that the reader needs to consider when evaluating the merits of the present study. The first relates to recall bias of the participants. While it is possible that participants overstate benefits accredited to CI, Cheng, et al (2000) point out that this may be substantially less for participant reports following CI. Participants and their children revisit the state of deafness when the processor is removed daily for bathing and sleeping, when the battery power is exhausted, and when equipment fails, thus, the benefits are likely well-understood and less prone to recall bias.

Secondly, this study is subject to the limitations of all qualitative research. The sample is small, and local. No assumptions regarding generalization can be made. However, issues with a small participant sample are inherent with this population whether using qualitative or quantitative methods due to the low numbers of children with multiple disabilities who use cochlear implants. Nonetheless, our results are strikingly similar to reports from studies conducted across North America, which lends support to the credibility of our findings. However, it is ultimately important for the reader to

judge the applicability of the research to their setting and client base.

Conclusions

With expanding CI criteria more children with multiple disabilities are receiving cochlear implants. It is crucial that parents' perspectives, observations, and values be included in our assessments of their children's abilities, and that we accept as legitimate such evidence in evaluating CI candidacy, progress, and success. Identifying CI outcomes that parents themselves describe as important provides the foundation for developing an outcome instrument that more closely reflects parental and family priorities. We believe that such instruments, combined with those currently used by clinicians, will result in a better informed process for determining CI candidacy, and provide a more complete profile of cochlear implant outcomes for children with severe multiple disabilities.

Finally, although the specific focus of this work was outcomes of cochlear implantation, the core issue that we were exploring was the contribution that access to sound makes to the lives of children with multiple disabilities and to their families. Because of this, it is possible that the instrument under development will have application beyond the cochlear implantation process.

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Appendix Structured Interview Questions

1. Please describe any differences you have seen in your child's behaviour that you believe are due to cochlear implantation.
2. Please describe any differences you see in your child's communication that you believe are due to cochlear implantation.
3. Please describe any differences in family interactions that you believe are due to cochlear implantation.
4. Please describe any differences in your child's social interactions that you believe are due to cochlear implantation.
5. Please describe any differences in your child's listening behavior since cochlear implantation.
6. Please describe any difficulties that you have had in managing your child's cochlear implant.
7. Is there anything you would like to add?