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Longer term functional outcomes and everyday listening performance for young children through to young adults using bilateral implants

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Abstract

Objectives: Firstly, to document a broad range of functional outcomes of bilateral implantation for young children through young adults at a postoperative point at which stable outcomes could be expected. Secondly, to evaluate the relationship between functional outcomes and age at bilateral implantation and time between implants.

Design: A study-specific questionnaire was administered to parents in an interview 3.5 years or more after sequential (n=50) or simultaneous (n=7) implants were received by their child. Median age at bilateral implantation was 4.1 years (range 0.7-19.8) and time between implants was 2.7 years (range 0.0-16.7).

Results: Based on parent report, 72% of the sequentially implanted children and young adults found it easy/only "a bit difficult" to adapt to the second implant, and were "happily wearing both implants together most of the time" by 6 months or less; 26% had not adapted, with both implants not worn most of the time or worn as a parental requirement. Seventy-two percent of sequentially implanted children and young adults had a positive attitude towards the second implant, including nine whose early postoperative attitude was negative or neutral. The majority of children and young adults preferred bilateral implants (70%) and used the two full-time (72%), whilst around half demonstrated similar performance with each implant alone. The proportion of non- or very minimal users of the second implant was just 9%. Eighty-eight percent of parents reported superior performance with bilateral versus a unilateral implant (n=40), or that only bilateral implants were worn (n=10) so performance could not be compared. The most commonly identified areas of superiority were localization, less need for repetition, and increased responsiveness. In balancing risks and costs with benefits, most parents (86%) considered the second implant worthwhile. Regarding the relationship between outcomes and demographic

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factors, the group achieving similar performance with each implant alone was younger at bilateral implantation and had less time between implants, and the group bilaterally implanted before 3.5 years of age (who also had less than two years between implants) had a higher proportion of positive outcomes on all functional outcome measures.

Conclusion: Overall, the results indicate primarily positive functional outcomes for children and young adults receiving bilateral implants at all ages, including when the delay between implants is long. The results are important for evidence-based pre-operative counseling, which helps families to make informed decisions and develop appropriate expectations. The results are also important for the development of clinical management practices which support and encourage the minority of recipients who have difficulty adapting to bilateral implants and/or achieving full-time use.

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Introduction

Many studies reported in the literature have used objective testing to demonstrate the additional perceptual benefit provided by bilateral cochlear implants (BiCIs) over a unilateral implant (UniCI) (see, for example, Beijen et al. 2007; Galvin et al. 2008, 2010; Litovsky, 2011; Johnston et al. 2009; Van Deun et al. 2010; Wolfe et al. 2007). While such studies provide vital information, they involve a self-selecting sample of participants, viz: those who are willing and able to complete such testing. Willingness to participate is likely to be influenced by the child's attitude towards, and listening ability with, a second implant (CI-2). Studies which seek information regarding real-life experiences with BiCIs are likely to include a greater range of children, and will provide information which is complementary to that obtained via objective assessments. There are five such studies in the literature, reporting information about four groups of 15 to 36 CI users, almost all of whom were sequentially implanted. Three groups included

only children under 12 years and information was collected from their parents (Fitzpatrick et al. 2011; Scherf et al. 2009a,b; Sparreboom et al. 2012) whilst the fourth included only adolescents, from whom self-reports were collected (Redfern & McKinley 2011). A sixth study involving younger children was significantly more limited in focus, but did include significantly more simultaneously implanted children (Galvin & Hughes 2012).

When BiCIs are received sequentially, there is an immediate challenge to adapt to using two CIs after a period of time using one CI, or one CI plus a hearing aid (Galvin et al. 2009). For the simultaneously implanted child, the change from no CIs to two CIs is arguably more similar to the change from no CIs to one CI. The relatively small amount of information in the literature regarding adaptation to BiCIs indicates that it is easy for the majority however the process varies across individuals and is not always straightforward. Although not a main focus, three of the reports referred to above included general comments about adaptation, such as: "a few" of 15 children bilaterally implanted under 11 years of age had early difficulties adapting (Fitzpatrick 2011); some children implanted between 6 and 12 years "took a long time to get used to CI-2" (Scherf et al. 2009b.); and some individuals implanted after 10 years of age took longer than others to adjust to CI-2 (Redfern & McKinley 2011). A fourth study focused specifically on hours of device usage as a measure of adaptation (Galvin & Hughes 2012). At two months postoperative, one child simultaneously and eight children sequentially implanted by 3.5 years of age were having some difficulty adapting, i.e., full-time device usage had not been achieved. At 12 months, this group was reduced to just four sequentially implanted children. The study was limited by the age range of the children, the single quantitative measure of adaptation, and the short length of follow up. There is a clear need for more information about children and young adults' adaptation to BiCIs and who is likely to experience more difficulty. Such information will help parents to make decisions about the timing of bilateral implantation, and to formulate

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appropriate expectations for the postoperative period. It will also help clinicians to tailor postoperative management practices to individual needs.

Beyond the adaptation phase, stabilized rates of device usage are of interest as a functional outcome measure of bilateral implantation. A high rate of BiCI usage over the long term maximizes the child's opportunity to develop binaural listening skills, and also allows the child to take consistent advantage of the benefits of bilateral sound input. Previous work has established a link between implant use and outcomes for unilateral implants, with the daily amount of device use being the strongest predictor of speech recognition performance (Wie et al. 2007). For children bilaterally implanted under 12 years of age, there has been variation in the reported proportion demonstrating very high levels of usage in the medium and longer term. For one group, around 90% of children were full-time users of BiCIs at 18 months and at 36 months post bilateral implantation (Scherf et al. 2009a,b). For a demographically similar group, only 63% of children were described as "always wearing CI-2" at the 24-month point (Sparreboom et al. 2012). The discrepancy may have been due to differences in the definitions of the categories of usage (Sparreboom et al. (2012) reported that an additional 24% of children were "sometimes not wearing CI-2"), and/or to differences in the preoperative counseling and selection procedures across countries. For an adolescent group, 64% were described as "consistent" users at between 1 month and 8 years post bilateral implantation (Redfern & McKinley 2011). At the other end of the usage spectrum is the proportion of non-users. The proportion amongst children bilaterally implanted before 12 years was reported as 0% at 24 months post bilateral implantation (Sparreboom et al. 2012) and 8.6% at 36 months (Scherf et al. 2009b). A comparable rate of 4% was reported for adolescents, although this study included participants with minimal experience (Redfern & McKinley 2011).

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The functional outcomes studies in the literature have mainly focused on performance with BiCIs. Few studies have provided information about children's attitude towards or performance with CI-2 alone, both of which are likely to influence BiCI usage and the degree of bilateral benefit. Negative experiences reported by parents at 18 months post bilateral implantation included 10% of children "disliking having to wear CI-2" (Scherf et al. 2009a). On the positive side, "more than half" of 15 parents reported that their child had no preferred CI and that listening performance was equivalent with each (Fitzpatrick et al. 2011).

It can reasonably be assumed that bilateral benefit would be the most important functional outcome of bilateral implantation for most families. In the assessment of bilateral benefit in the studies in the literature, interviewees were asked to report on changes which had occurred since bilateral implantation, including in some specific areas. Across the studies, reference was made to change in many areas however the discussion below will be limited to those areas for which the authors specified the proportion of children demonstrating change.

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Localization is a key area of expected benefit from BiCIs (Fitzpatrick et al. 2011; Sparreboom et al. 2012). In one study, a fairly high proportion (76%) of children bilaterally implanted between 2 and 8 years of age demonstrated improved localization in the first 24 months postoperative (Sparreboom et al. 2012). Results were variable for slightly older (implanted before 12 years) and less experienced (average 18 months) groups, with 55% improving (Scherf et al. 2009a) versus the "majority" of 15 children being unable to localize (Fitzpatrick et al. 2011). Interestingly, despite their older age at bilateral implantation, 68% of adolescents reported improved localization (Redfern & McKinley 2011). Thus, with the exception of Fitzpatrick et al.'s very small group, improved localization occurred in everyday life for half to three-quarters of the children across studies.

Speech understanding per se was only specifically considered in two studies. The proportion of children demonstrating improvement was very high, being 80% for "speech understanding in quiet and noise" by 18 months (Scherf et al. 2009a) and 83% for "listening to speech without lipreading" by 24 months (Sparreboom et al. 2012). More often, authors summarized broader communication or listening changes, including: "more comfortable listening" (85%) and "easier communication" (55%) (Scherf et al. 2009a), "improved communication with others" (83%) (Sparreboom et al. 2012), and "better hearing and less difficulty hearing" (43%) (Fitzpatrick et al. 2011).

Improvements in speech understanding, ease of listening, and general communication would be expected to result in psychosocial benefits for children. This was evident in the 84% of adolescents who reported increased confidence in group conversations following bilateral implantation (Redfern & McKinley 2011). Less impact was evident for children under 12 years, with only 30% of parents identifying "positive change in behavior (more open, happier, proud of CI-2)" (Scherf et al. 2009a) and 43% reporting benefits such as reduced frustration and improved peer relationships (Fitzpatrick et al. 2011). It is possible that psychosocial benefits are more easily identified by self-report rather than parent-report, and/or that some bilateral benefits had greater impact for adolescents due to the typical changes in social interactions with age.

As noted previously, in the studies in the literature interviewees were asked to identify changes that had occurred *since* bilateral implantation. With this approach, the change due to bilateral benefit could not be isolated from change due to other effects such as normal development and increased "hearing age". This would be the case particularly for children who were younger and/or had a longer postoperative period. Scherf et al. (2009b) highlighted this difficulty, and thus made a cross-study comparison of their results with those of unilaterally implanted children.

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Although such comparisons are complicated by methodological and participant differences, they suggested that, overall, the bilaterally implanted children appeared to make faster progress, develop higher level perceptual skills, and achieve more advanced abilities, such as more natural communication.

Irrespective of the outcome of bilateral implantation, there are associated risks and costs, financial and otherwise; this is particularly so for sequential implantation, which involves two operations and two adaptation periods. Only parents and older children are in a position to balance the risks and costs with the benefits, and to determine whether the experience of bilateral implantation was worthwhile for their family. In the few studies reporting such judgments, the results for children bilaterally implanted under 12 years have been consistently positive. All parents indicated "extreme satisfaction with the outcomes and their decision" (Fitzpatrick et al. 2011) or did not regret the decision to obtain CI-2 (Scherf et al. 2009b). In contrast, only 17 of 25 adolescents reported that it had been "worth the endeavor" (Redfern & McKinley 2011). This discrepancy may have been due to the degree of benefit gained and/or the value parents placed on indirect benefits, such as the value of CI-2 as a 'back-up' or the satisfaction of having provided their child with the best access to sound (Fitzpatrick et al. 2011). Parents may also have been more cognizant of the challenges their child would face as an adult in a hearing world, and therefore placed more value on any bilateral benefit gained.

For all outcomes of implantation it is clinically valuable to identify relationships with user characteristics. Two studies which considered such relationships examined only the outcome of BiCI usage rate. Weak-to-moderate significant negative relationships were found between the category of BiCI usage at the 12-month point and each of the factors of age at BiCIs and time between CIs for 47 children sequentially or simultaneously implanted by 3.5 years of age (Galvin

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& Hughes 2012). When simultaneously implanted children were excluded from the analyses, no significant relationships were found between BiCI usage rate and age at BiCIs (Galvin & Hughes 2012; Sparreboom et al. 2012), time between CIs (Galvin & Hughes 2012), or the balance of auditory/signed information provided to the children (Sparreboom et al. 2012). A moderate negative correlation was reported, however, between BiCI usage rate and the speech recognition score differential between CI-1 and CI-2 (Sparreboom et al. 2012). A third study considered the impact of age at BiCIs by comparing hearing performance at 36 months postoperative for children bilaterally implanted before versus after 6 years of age (Scherf et al. 2009b). Parent ratings were higher for the earlier-implanted group for positive aspects (e.g., directional hearing and hearing in complex situations) but not different for negative aspects (e.g., reliance on lipreading and difficulties in noise).

The five studies discussed above which reported on multiple functional outcomes have provided a significant amount of information regarding children receiving, primarily, sequential BiCIs (Fitzpatrick et al. 2011; Redfern & McKinley 2011; Scherf et al. 2009a,b; Sparreboom et al. 2012). This information is valuable for clinicians providing preoperative and postoperative care, and for parents and older children during preoperative decision-making and the postoperative phase. Nevertheless, there is a clear need for additional data in this area. Although all reported some functional outcomes, these studies varied in focus and breadth; for example, Fitzpatrick (2011) was primarily concerned with parental decision-making and expectations. The groups consisted of children implanted under 12 years or as adolescents, so that no study included a wide range of age at BiCIs. The range of time between CIs was also restricted, being less than one year for only three children across all studies. It was therefore difficult to examine the relationship between demographic factors and the reported outcomes, and the majority of the studies did not do so. The studies also included relatively small samples and mostly reported

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short-to-medium term outcomes. The authors of the one study to examine outcomes only for children who were 36 months postoperative emphasized the importance of long-term results. Their repeat examination of the same group at 18 and 36 months (Scherf et al. 2009a,b) indicated a change in the emphasis of parent reports from improved hearing and speech understanding to quality of life benefits and the development of more complex hearing skills.

The first aim of the current study was to document a broad range of functional outcomes for a large and heterogeneous group of bilateral implant recipients for whom a sufficient time had elapsed post implantation for there to be a reasonable expectation of stable outcomes. The second aim was to evaluate the relationship between the functional outcomes and the demographic factors of age at BiCIs and time between CIs.

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Materials and Method

Design

A study-specific questionnaire was administered in an interview with parents whose children had received BiCIs more than 3.5 years prior to interview.

Bilaterally implanted children and young adults

Figure 1 illustrates the formation of the group of 57 bilateral implant users whose parents were interviewed for the present study. Between September 2003 and September 2008, 80 children and young adults received BiCIs at the Royal Victorian Eye and Ear Hospital Cochlear Implant Clinic in Melbourne. In 2003, a broad bilateral implant research project was initiated, and the current study was a later part of that project. The selection criteria for the broader research project were: onset of hearing loss prior to adolescence, scheduled to receive sequential or simultaneous BiCIs, no significant developmental or cognitive delays reported by professionals working with the child, sufficient oral language skills to participate in testing, a parent with

sufficient English language skills to provide feedback on the child's progress, and a record of generally attending scheduled Clinic appointments. Given that their parents were interviewed for this study, the bilaterally implanted children and young adults will henceforth be referred to as "children", irrespective of their age.

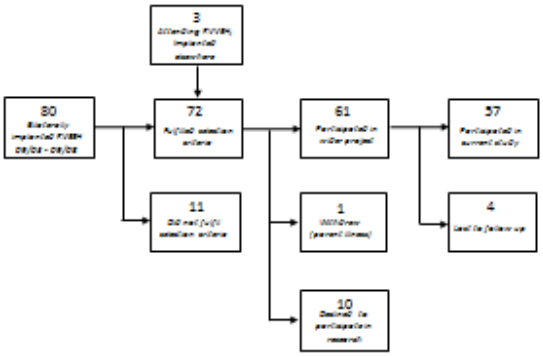


Figure 1: Flowchart of the formation of the group of children whose parents completed the questionnaire.

For 51 children, three-frequency pure tone averages indicated that their loss was bilateral profound loss before 26 months. The remaining six children had only a severe-profound loss in at least one ear at first implantation. A congenital onset was confirmed for 31 children and assumed for 18 children (based on etiology and early concern regarding auditory responses). For five children onset was unknown but diagnosis was between 8 and 19 months, and for the remaining three children meningitis was contracted between 9 and 21 months. Table 1 provides additional information regarding etiology, hearing aid use, and implant type.

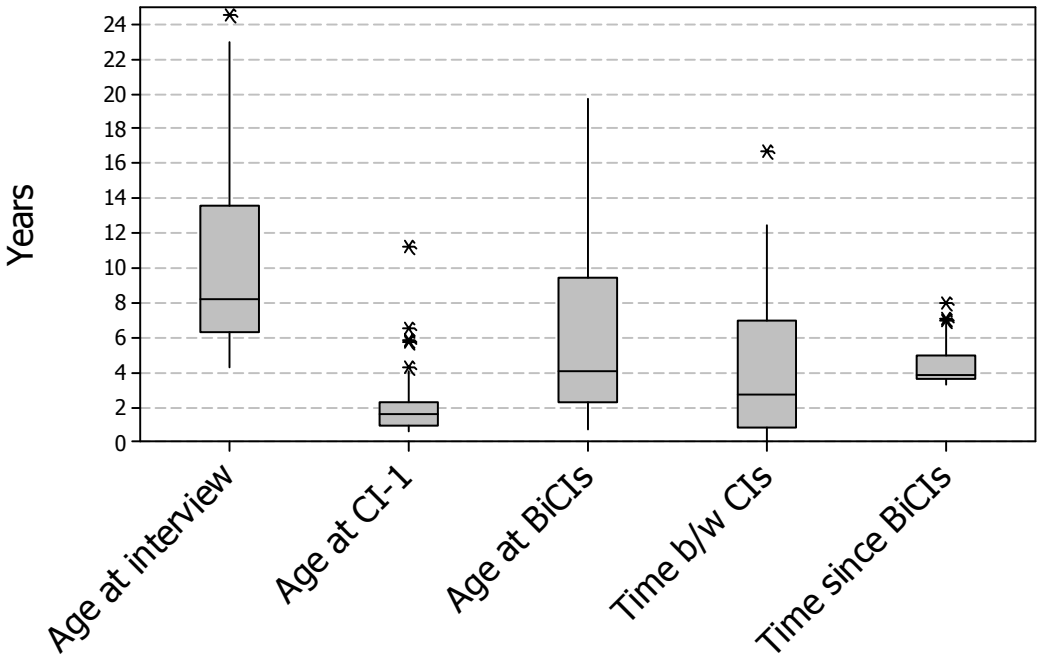


Figure 2: Age at interview, age at CI-1, age at BiCIs, time between CIs, and the time since bilateral implantation for the 57 children. The box represents the interquartile range, the line indicates the median, the whiskers indicate the minimum and maximum values, and the asterisks indicate outliers.

Figure 2 presents the median and range for age at interview, age at CI-1, age at BiCIs, time between CIs, and the time since bilateral implantation for the 57 children. Calculations were based on the date of switch-on of the relevant implant(s). The five children who are outliers for age at first implant were born prior to 1994 when early implantation was not routine. Age at BiCIs varied, being at 3.5 years or younger for 46% of children and at 7.5 years or older for 30% of children. The time between CIs was less than 12 months for 33% of children, including the seven simultaneously implanted children, but at least two years for 57% of children and at least 10 years for 9% of children. The children were at least 3.5 years post bilateral implantation, with three exceptions; three parents who were difficult to contact were interviewed earlier (3.3, 3.4 and 3.4 years respectively) when the opportunity for interview arose.

The Eye and Ear Hospital Implant Clinic provided the families with pre- and postoperative clinical care related to implant candidacy, surgery, and device maintenance and programming. Up to eight early learning-to-listen therapy sessions were available, although the target group was children receiving a first implant and very few sequentially implanted children attended such sessions post bilateral implantation. Habilitation/rehabilitation services were primarily accessed through external providers, so that post bilateral implantation rehabilitation varied enormously. Implant clinicians encouraged sequentially implanted children to use CI-2 alone, however anecdotal feedback indicated only a minority of children, primarily older adolescents and young adults, did so.

Study-specific questionnaire

The questionnaire (attached as Appendix A; Supplemental Digital Content 1) consisted of four sections. In section 1, Device Use, the parent was asked to identify the situations in which their child used one or no CIs in order to prompt them to think in detail about their child's pattern of device. The parent then selected a category of BiCI usage. The maximum category was >90%, as it is typical for CI(s) not to be worn for short periods of time each day, such as during bathing or immediately on rising in the morning. When BiCI usage was <90%, the parent was asked to identify which, if any, implant was worn when BiCIs were not used. In section 2, Benefit and Performance, the questions focused on the comparison of performance with CI-1 versus CI-2 and the comparison of performance with BiCIs versus a specified UniCI (termed "bilateral benefit"). It is important to note that the parent was asked to compare current performance in the different device conditions, and not asked to make comparisons with performance prior to bilateral implantation. With this approach, differences in performance due to development or increased hearing experience were excluded; the disadvantage of this approach

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Table 1: Demographic information relating to etiology, diagnosis of Auditory Neuropathy (AN), pre-implant use of hearing aids, and implant types for the 57 children, and use of a contralateral hearing with CI-1 for the 50 sequentially implanted children.

Etiology *(AN diagnosis)*	Unknown: n = 28 (AN: n = 3)	Genetic: n = 17[a]	Syndrome WVA[b]: n = 4 KID[c]: n = 2 Waardenburg's: n=1	Infection CMV: n = 2 Meningitis: n = 3	
Use of hearing aids pre-implantation	Consistent: n = 28[f]	Half-time or less[d]: n = 25	Short duration[e] (≤ 1mth): n = 4		
Use of contralateral hearing aid	Consistent between CI-1 and CI-2 operations n = 8	Partial (<30%) between CI-1 & CI-2 operations n = 8	Initially consistent; rejected 7mths-11yrs prior to CI-2 n = 6	At school only during 4-12 mth hearing aid trial prior to CI-2 n = 3	Refused to wear or continually removed aid n= 25
Nucleus[g] *implant type: CI-2/CI-1*	Freedom[h]/Freedom n = 25	Freedom/CI24[h] n = 19	Freedom/CI22[i] n = 2	CI24/CI24 n = 7	CI24/CI22 n = 4

[a] Connexin 26: n = 11; other genetic causes: n = 6. [b] Wide Vestibular Aqueduct syndrome. [c] Keratitis-Ichthyosis-Deafness syndrome. [d] Minimum ½ hour per week; most common report 2 to 3 hours daily. [e] After a maximum of one month the parents chose not to persist due to lack of evidence of benefit or the child was implanted. [f] Unilateral fitting due to atresia for one child. [g] Nucleus implants manufactured by Cochlear Limited, Macquarie University, Australia. [h] Implant with 22 electrodes and 2 extra-cochlear electrodes. [i] Implant with 22 electrodes.

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was that a high proportion of children would be relatively unpractised users of a UniCI at the time at which their performance was being observed. For this reason, the parent was first asked if they were able to make the comparison in question; if, for example, the child never wore a UniCI then the parent would not able to make either comparison. The parent was then asked if they could "see a difference in performance" between the device conditions and, then, what those differences were. It is important to note that neither the format of the question nor the interviewer indicated potential areas of difference or the likely direction of differences. The aim of this approach was to reduce parental bias in favor of the BiCIs in which the family had invested so heavily, and to limit the potential to be identifying parents' expectations of benefit in certain areas, rather than their observations of actual benefit.

In section 3, Adaptation and Attitude/Preference Over Time, the parent was asked to select the most suitable description of the ease with which their child adapted to BiCI use, including the time period before the child was "happily using both CIs together most of the time". The parent was also asked to indicate the child's attitude towards CI-2, as expressed or evident in their behavior, at different time points post bilateral implantation. The parent was also asked to indicate which implant(s) the child would have chosen to wear, of their own accord, at different time points.

In section 4, Final Questions, the parent was asked to take into account "the effort, time and risks in acquiring the second implant, and any extra benefit that was gained", and to then indicate whether obtaining the second implant had been worthwhile. The opportunity was also provided to detail any other relevant information.

The questionnaire was administered in a phone interview in which the interviewer sought the opinion of the parent as an observer and communication partner of their child; self-reports from

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older children were not included in this parent-reported data. As it was often difficult to contact parents by phone at a time at which they were available to complete the questionnaire, face-to-face interviews were conducted with five parents when their child was attending for other research assessments or for standard clinical services. A sixth parent with a hearing loss was also interviewed face-to-face, and a seventh parent was interviewed by email as the family was overseas.

Results

Device Use, Benefit, and Performance at the Time of Interview

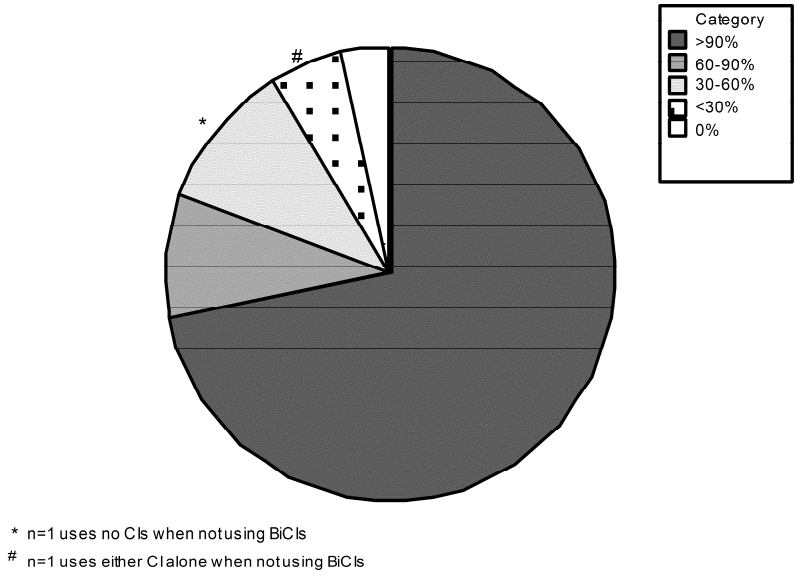


Figure 3: Proportion of the 57 children who, at the time of interview, used BiCIs more than 90% of the time, 60 to 90% of the time, 30 to 60% of the time, less than 30% of the time, or not at all (0%). Except where indicated, children with <90% usage used CI-1 alone when not using BiCIs.

Figure 3 indicates the proportion of children in each category of BiCI usage. Of the 57 children, 46 used BiCIs the majority of the time (i.e., more than 60%), with 41 of these using them full-time. For the 16 children not using BiCIs full-time, the reason and the alternative device used are

presented in Table 2. Five children used BiCIs less than 30% of the time, with two of these being non-users.

Table 2: Device preference and reason for less than full-time use for the 16 children who did not use BiCIs full time.

Number of children	Device preference	Reason for less than full-time use
12	CI-1	n = 10: prefers the sound of CI-1 alone; dislikes CI-2 n = 2: none provided
1	either CI alone	does not tolerate BiCIs in noisy environments
2	BiCIs	n = 1: does not tolerate BiCIs in very noisy environments n = 1: chooses to use no CIs for a period at the start and end of each day
1	CI-1 (some situations)	chooses to use CI-1 at home and BiCIs in difficult listening situations

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Fourteen parents were unable to compare performance with BiCIs versus a UniCI. This was because their sequentially implanted child made minimal or no use of BiCIs (n=4) or because their child only ever used BiCIs (n=10). The latter reason could be considered a positive outcome in terms of bilateral implantation. Forty-three parents, including four parents of simultaneously implanted children, were able to make the comparison of their child's current performance when using a UniCI versus their current performance with BiCIs; UniCI use most commonly occurred upon rising in the morning, prior to bedtime, when a replacement battery was not available, or when one speech processor was being repaired. The comparison UniCI was

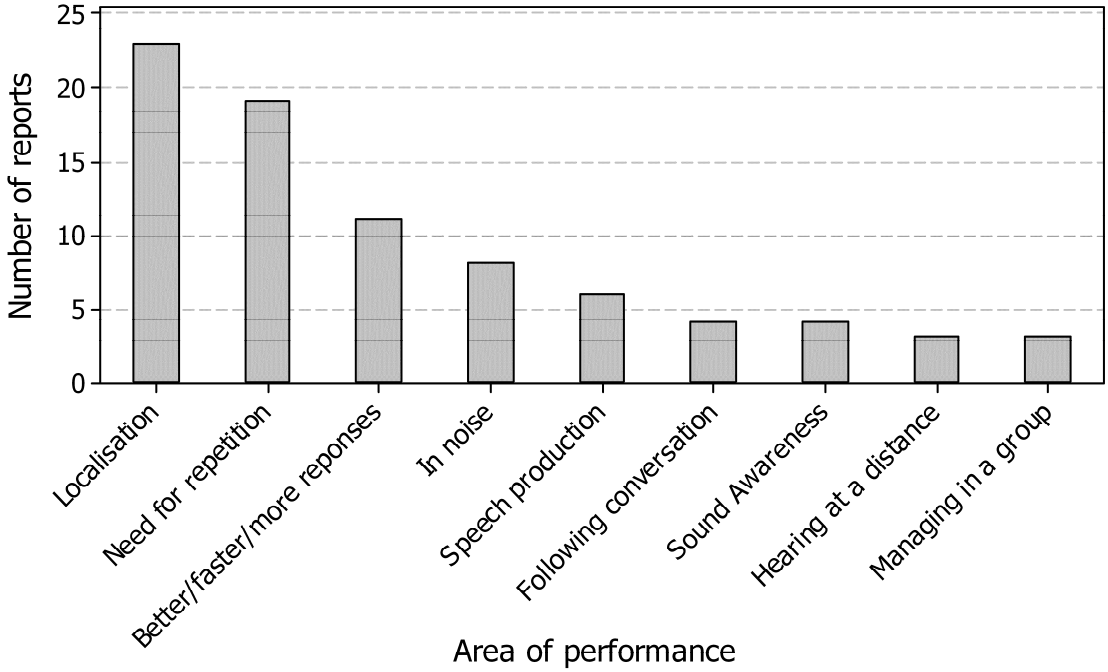


Figure 4: The specific areas in which superior performance was most often reported by the 40 out of 43 parents who reported superior performance with BiCIs versus a UniCI, and the number of parent reports in each area.

specified as CI-1 by 24 parents and either CI by 19 parents (who perceived performance as equivalent with each CI). The majority (93%) of the 43 parents reported that there was a difference in performance, and that performance was superior with BiCIs. Some parents reported that their child was "hearing more" or "hearing better", whilst others indicated specific area(s) in which performance was superior. The most commonly indicated areas of superior performance, and the number of reports in each area, are presented in Figure 4. Parents also identified psychosocial benefits, including that the child did not have to concentrate as hard (n = 1), was more involved with their surroundings (n = 2), was more confident in social groups (n = 2), was less frustrated (n = 1), was more talkative (n = 1), was less fatigued (n = 1), and paid attention more quickly and for longer (n=1).

Ten parents felt unable to make the comparison of their child's performance with each CI alone. Nine parents because their sequentially (n = 7) or simultaneously (n = 2) implanted child only wore BiCIs, and one because their simultaneously implanted child (n = 1) only wore BiCIs or no CIs. Of the 47 parents able to make the comparison, 24 reported that CI-1 was the superior implant, 22 reported that the implants were equivalent (including for the four simultaneously implanted children), and one reported that CI-2 was the superior implant.

Adaptation, and Attitude and Device Preference Over Time

For the simultaneously implanted children, adaptation to BiCIs could not be separated from adaptation to implant use in general. Six of these children found it "easy" to adapt and were happily wearing both CIs together most of the time within 2 to 3 months. The seventh child was still not wearing no CIs after school of each day at the time of interview.

As shown in Figure 5, the majority (72%) of sequentially implanted children found it easy or only "a bit difficult" to adapt to CI-2, and were "happily wearing both CIs together most of the time" by 6 months or less. An additional participant took significantly longer (5 years) to achieve this milestone. The remaining 26% of sequentially implanted children found the process very difficult and, at the time of interview, they were either not wearing both CIs together most of the time or were doing so as a parental requirement.

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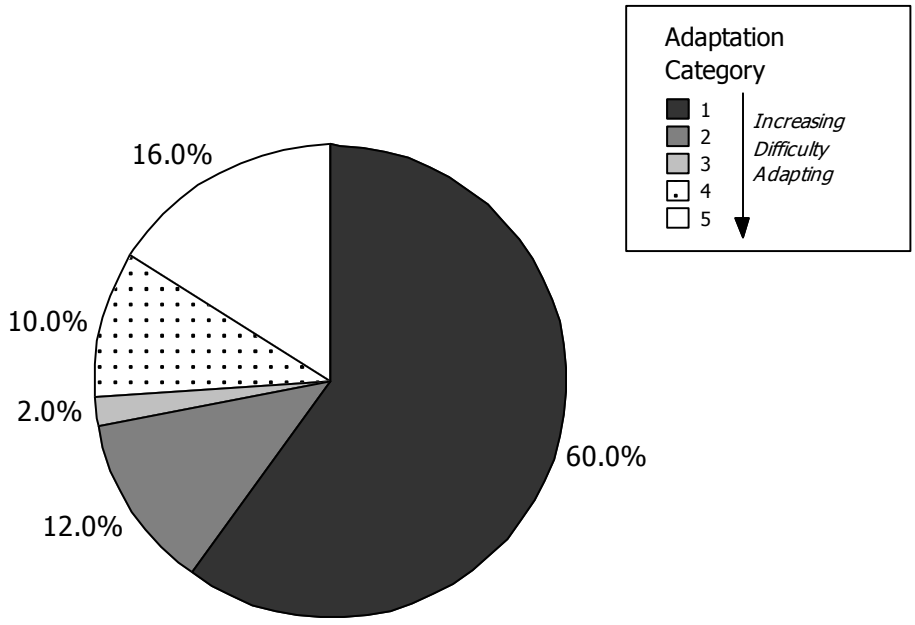


Figure 5: The proportion of sequentially implanted children (n = 50) classified into each of the adaptation categories. Category 1 adaptation was "fairly easy, with the child happily using both CIs together most of the time within 2-3 months"; category 2 was "a bit difficult, with the child happily using both CIs together most of the time within 3-6 months; category 3 was "very difficult, with the participant taking 5 years before happily wearing both CIs together most of the time"; category 4 was "very difficult, with the child only wearing both CIs together most of the time because he/she was required to by parents or teachers; category 5 was "still difficult, and the child was not wearing both CIs together most of the time".

The question regarding attitude was only applicable to sequentially implanted children. As shown in Figure 6, over half (62%) of the children had a generally positive attitude towards CI-2 in the first few months post bilateral implantation. The majority maintained this attitude over time, and an additional nine children also developed a positive attitude. In contrast, the proportion of children with a generally negative attitude decreased over time to just 12% at the time of interview.

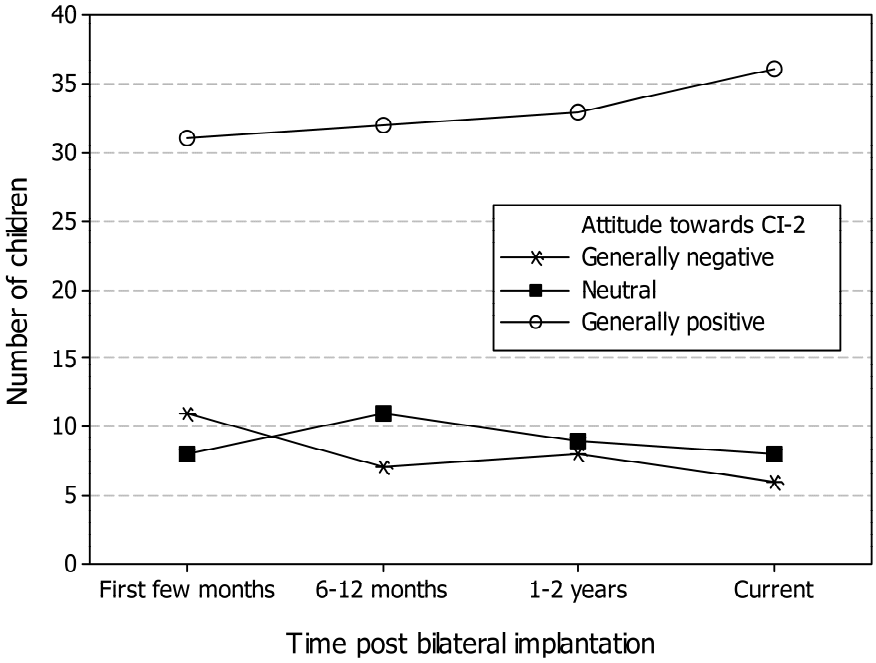


Figure 6: The number of sequentially implanted children (n = 50) who held a generally negative, neutral, or generally positive attitude towards their CI-2 at different time points post bilateral implantation.

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Figure 7 indicates the device preference in the first few months post bilateral implantation and the preference at the time of interview, as a function of time between CIs for the 57 children. Of the 47 children who expressed or demonstrated a preference in the first few months post bilateral implantation, 25 (53%) preferred BiCIs. By the time of interview, this proportion was 70%, with an additional seven children then old enough to express a preference for BiCIs and others having changed their preference from no CIs or a UniCI; for this latter group, the time taken to demonstrate a preference for BiCIs was 6 to 12 months (n = 6), 1 to 2 years (n= 2), or 2 to 5 years (n = 3). Preference change was not just in one direction. Of those initially preferring BiCIs, four children preferred a UniCI by just 6 to 12 months post bilateral implantation, and a fifth did so by 1 to 2 years. It is worth noting that, of the 47 children who expressed or demonstrated a preference in the first few months post bilateral implantation, 20 who preferred BiCIs and 11 who

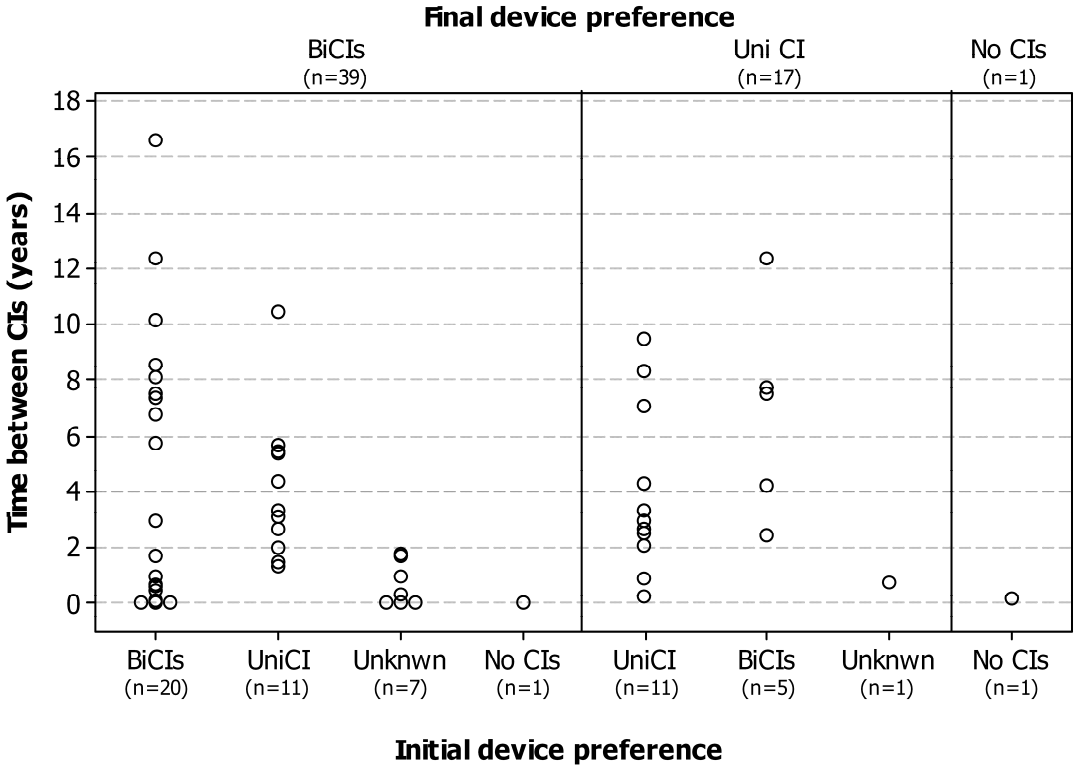


Figure 7: The initial and final device preference as a function of time between CIs (years) for the 57 children. Initial device preference was that demonstrated in the first few months post bilateral implantation, and final device preference was that demonstrated at the time of interview. Children who were too young to demonstrate a preference are categorized as unknown.

preferred a UniCI maintained this same preference through until the time of interview. There was a strong and significant correlation between device preference and attitude towards CI-2 at the time of interview ($r_s = 0.8$, $p < 0.0001$).

Worth of the Second Implant

By far the majority of parents considered that obtaining BiCIs had been worthwhile; this group included 86% of all parents, 84% of parents of sequentially implanted children, 100% of parents of simultaneously implanted children, and some parents of children in all categories of BiCI usage. Twenty-three parents provided no additional reasons for their opinion beyond the bilateral benefits previously identified. The most common additional reasons were related to the use of both ears or sound from both sides (n = 6), enhanced or better hearing overall (n = 6), the fact that

the child was "just like the other kids (with normal hearing)" when using two CIs (n = 7), and/or the value of a back-up device (n = 6). Six parents also referred to psychosocial benefits, such as a better social life, an increased sense of security, and a more confident, happier, and/or relaxed child. For the parent of a non-user, the reason was "they gave it a go" and hoped that CI-2 may be worn in the future.

Eight parents were unsure (n = 5) or considered that obtaining CI-2 had not been worthwhile (n = 3). The latter group reported their reasons as lack of evident benefit and limited usage. Some parents in the unsure group hoped that therapy received after CI-2 (prior to the child becoming a non-user of CI-2) may have benefitted skills with CI-1 (n = 1), or reported no benefit but hoped some benefit would be gained in the future (n = 2). The remaining parents reported that they were unsure if the bilateral benefit gained had been worthwhile because the child either did not like to wear the device (n = 1) or had suffered a postoperative infection (n = 1).

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Relationships Between Outcomes and Demographic Factors

The relationship between outcomes and the demographic factors of age at BiCIs and the time between CIs was examined. It is important to note that there was a very strong and significant association between these demographic factors ($r = 0.95$; $p < 0.000$). Spearman correlation coefficients were calculated for each demographic factor and each of the two outcome measures with more than three response categories. For the 57 children, there was no significant association between BiCI usage and age at BiCIs or time between CIs ($r \leq 0.19$; $p \geq 0.16$). There was a weak association between ease of adaptation and time between CIs ($r = 0.25$; $p = 0.058$) and age at BiCIs ($r = 0.29$; $p = 0.027$). Only the latter result was significant, and this relationship was not significant when the simultaneously implanted children were excluded ($r = 0.21$; $p = 0.14$).

In order to be able to employ a consistent analysis of relationships across all outcome measures, response options were collapsed to form a "positive outcome" and a "neutral/negative outcome" for each measure. Presented in Table 3 is the median age at BiCIs and the median time between CIs for the positive outcome group and the negative/neutral outcome group for each outcome measure; results are presented for the overall group and for the sequentially implanted children only. The median age at BiCIs was consistently younger, and the time between CIs was consistently less, for the positive outcome group, however Mann-Whitney tests indicated that nearly all differences were not significant ($p \geq 0.13$). The exception was for performance with CI-2 versus CI-1 alone; for this measure, the median age at BiCIs was around 5 years younger for the positive outcome group (overall group: $W = 337$, $p < 0.0001$; sequential-only group: $W = 697$, $p < 0.0001$) and the median time between CIs was around 4.6 years less (overall group: $W = 341$, $p < 0.0001$; sequential-only group: $W = 691$, $p = 0.0001$).

The relationship between outcomes and demographic factors was also considered in terms of outcomes for those receiving BiCIs at a younger versus older age. Electrophysiological studies involving children with unilateral implants have indicated a period of maximum neural plasticity before 3.5 years of age (Sharma et al. 2002). For the current group, the 25 children who received BiCIs before 3.5 years of age (mean = 2.0, SD = 0.9) had a time between CIs of 2.0 years or less (mean = 0.7, SD = 0.67), whilst the 32 who received BiCIs after 3.5 years (mean = 9.0, SD = 4.6) had a time between CIs of more than 2 years (mean = 6.4, SD = 3.5) Figure 8 shows the proportion of children in each of these groups with a positive versus a neutral/negative outcome for each of the outcome measures. The outcome measures and the relevant positive versus

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Table 3: Median age at BiCIs and median time between CIs (in years) for a "positive" outcome group and a "neutral/negative" outcome group on the various outcome measures when responses were collated for the sequentially and simultaneously implanted children and for the sequentially implanted children only. P-values indicate the result of Mann-Whitney tests comparing the age at BiCIs and the time between CIs for the two outcome groups; values significant at the .05 level are in bold.

		Median age at BiCIs (years)			*Median time between CIs (years)*		
Children	Outcome measure	Positive outcome	Neutral/ negative outcome	p-value	Positive outcome	Neutral/ negative outcome	p-value
Seq + sim	Adaptation[6] [a]	3.0	4.5	0.19	1.9	3.4	0.29
Seq only		4.6	5.2	0.50	2.8	3.8	0.46
Seq + sim	BiCI usage[7] [b]	3.1	5.8	0.14	1.8	3.2	0.25
Seq only		4.2	6.0	0.42	3.0	3.4	0.43
Seq + sim[1] [c]	Preference[8] [d]	3.1	5.7	0.16	1.8	3.4	0.13
Seq only[2] [e]		4.6	5.7	0.60	3.0	3.4	0.61
Seq only[3] [f]	Attitude to CI-2[9] [g]	4.0	5.8	0.31	2.8	3.8	0.27
Seq + sim[4] [h]	CI-2 vs CI-1[10] [i]	2.4	7.4	**<0.0001**	0.97	5.6	**<0.0001**
Seq only[5] [j]		2.5	7.4	**<0.0001**	1.0	5.6	**0.0001**

[a] Outcomes: adaptation fairly easy or a bit difficult, with child happily using both CIs together most of the time within 6 months versus difficult/very difficult/with ongoing difficulty being experienced and the child not wearing both CIs most of the time. [b] Outcomes: full-time use of BiCIs versus not full time. [c] N = 56; one sequentially implanted child excluded because the preference was for no CIs. [d] Outcomes: preference for using BiCIs versus preference for UniCI. [e] N = 49; one sequentially implanted child excluded because the preference was for no CIs. [f] Outcome measure relevant only to sequentially implanted children. [g] Outcomes: positive attitude towards CI-2 versus neutral or negative attitude. [h] N = 47; 10 parents could not compare CI-2 with CI-1 because their child always wore BiCIs (or no CIs, in one case). [i] Outcomes: performance with CI-2 at least as good as that with CI-1 versus CI-2 poorer. [j] N = 43; seven parents could not compare CI-2 with CI-1 because their child always wore BiCIs.

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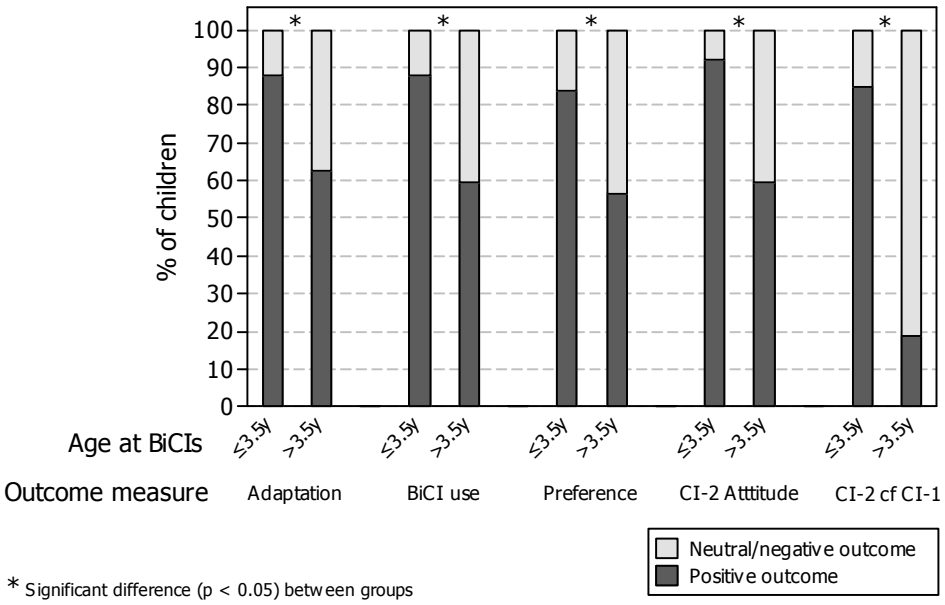


Figure 8: The percentage of children for whom a positive or neutral/negative outcome was reported on the specified outcome measure for children who received BiCIs before 3.5 years of age (and with 2 years or less between CIs; n = 25) or after 3.5 years (n = 32). The sample sizes are reduced to 20 and 27 respectively for the outcome measure of CI-2 cf CI-1 because 10 parents were unable to make the comparison.

neutral/negative outcomes were: ease of adaptation (fairly easy or a bit difficult with child happily using both CIs together most of the time within 6 months versus difficult, very difficult or still difficult), full-time BiCI usage (yes versus no), a preference for using BiCIs over a unilateral CI (yes versus no), attitude towards CI-2 (positive versus negative or neutral), and performance with CI-2 compared with CI-1 (at least as good as CI-1 versus poorer than CI-1). For each of these five outcome measures, Chi-square analysis indicated that the proportion of children who achieved a positive outcome was significantly higher in the group who received BiCIs before 3.5 years (and had 2 years or less between CIs) as compared with the group who received BiCIs after 3.5 years ($\chi^2 \geq 4.71$, $p \leq 0.03$). Separate analyses were conducted for the 50 sequentially implanted children on the outcome the measures of more relevance to this group,

i.e., ease of adaptation, attitude towards CI-2, and performance with CI-2 compared with CI-1. These analyses also indicated that the proportion of children who achieved a positive outcome was significantly higher in the group who received BiCIs before 3.5 years (and had 2 years or less between CIs) as compared with the group who received BiCIs after 3.5 years ($\chi \geq 4.98$, $p \leq 0.046$). It should be noted that the ten children whose parents were not able to compare performance with CI-2 versus CI-1 could not be included in the above analyses (n = 10; mean age at BiCIs 6.3 years (SD: 6.4); mean time between CIs 3 years (SD: 3.2)).

Discussion

For those receiving sequential bilateral implants, adapting to the new sound input via both CIs is the first crucial step post-implantation. Fifty children in the current study had a delay between implants of 1.5 months to 16 years 8 months, with 33 having at least a two year delay. Prior to bilateral implantation, the majority received only unilateral auditory stimulation, whilst eight consistently used a contralateral hearing aid. In addition, many were no longer young when they received BiCIs, with 31 children being more than 3.5 years old and 17 being more than 7 years old. For these reasons, it is a very positive outcome that 72% of the sequentially implanted children found it easy or only "a bit difficult" to adapt to CI-2, and were "happily wearing both CIs together most of the time" within 6 months. It is valuable to consider the adaptation of the remaining 14 children in order to learn from their experiences. One child developed self-motivated BiCI use at around 5 years postoperative, following discussions with an older BiCI recipient, and a second child used BiCIs but chose a UniCI in easier listening situations. The remaining 12 children had more clearly not adapted; BiCis were either not used or were used as a parental requirement. The group was heterogeneous, being aged between 1 and 11 years at bilateral implantation (mean: 5.6; SD: 3.2) with from 0.2 to 9.5 years between CIs (mean: 4.0;

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SD: 2.9). A previous study, which included 18 children from the present group, found that a similar proportion of children (4 of 27) implanted at or before 3.5 years were not using their two implants full-time at 12 months postoperative, giving a consistent indication of early adaptation difficulties for a small proportion of younger children (Galvin & Hughes 2012). Clearly, children of all ages are in potential need of support and encouragement during the adaptation period following sequential implantation. Parents need to be informed that adaptation can be a long process for a minority of children, and that significant benefit can still be achieved in the longer term even when the initial adaptation period is difficult or protracted.

Consistent with the ease of adaptation for the majority of sequentially implanted children, 62% had a positive attitude towards CI-2 in the first few months postoperatively. A positive attitude could be expected to promote use of CI-2, which, in turn, would aid adaptation. A positive attitude may also be the result of pleasant and/or useful sound being perceived via the implant, and this percept may drive CI-2 use. Parent feedback suggested that both of these scenarios occurred. Many young children were comfortable with the sound percept via CI-2 from switch-on, were therefore happy to use both implants together, and quickly developed listening skills with CI-2; thus a cycle of positive reinforcement was initiated. Parents also reported that some children who were older (adolescents and young adults) were determined to develop their listening skills and to maximize their benefit from the BiCIs, including choosing to practise using CI-2 alone. Thus their internally generated positive attitude also initiated a positive cycle of use and skill improvement. Irrespective of the origin of their positive attitude, the majority of children maintained this attitude through to the time of interview. An additional five children moved from a neutral or negative to a positive attitude within 6 to 12 months; this relatively quick change of attitude suggests that the initial sound was not perceived as pleasant or useful, but that it became so fairly quickly. Interestingly, of the 39 children with a positive or neutral

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attitude, seven did not experience adaptation that was easy or only "a little bit difficult". On the other hand, of the 11 children with a negative attitude, adaptation was easy or only "a little bit difficult" for four of them. Clinicians and families need to be aware of these variations in co-occurring outcomes, and ensure that sufficient support is provided to the child during the period of adaptation.

Although there was a strong and significant correlation between device preference and attitude towards CI-2 at the time of interview, device preference is subtly different to attitude. All of the children with a neutral or negative attitude preferred to wear a UniCI, however four children with a positive attitude also preferred to wear a UniCI. Device preference was also correlated with BiCI usage ($r_s = 0.7$, $p < 0.0001$). Nevertheless, it is clear that preference must be considered in addition to usage in order to obtain a complete picture. For example, two children who sometimes chose to use no CIs or a UniCI still had an overall preference for BiCIs. Also, four children who were required by their parents to use BiCIs full-time actually preferred to use no CIs (n = 1) or a UniCI. Device preference is a vital outcome measure. If a child or young adult has a preference for using a UniCI, this should be an indicator for the clinician and family that additional intervention may be beneficial (for example, modification of the map(s) to make bilateral listening more comfortable) and/or that BiCI usage should be monitored.

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Naturally, device preference can change over time. The optimal outcome is for a child to prefer to use BiCIs. This was reported for the majority (39) of children in this study at the time of interview, however 11 of these children initially preferred a UniCI. It is important for families and children to know that, with persistence and support, a preference for BiCIs can develop over time. Recognition that a preference for a UniCI may not change, or that an initial preference for BiCIs may not be maintained, is also important to ensure that preoperative expectations are

realistic. The fact that an initial preference for BiCIs was not always maintained highlights the importance of ongoing clinical and parental support for children using sequential BiCIs. It is worthwhile to note that no simultaneously implanted children preferred to use a UniCI at any time.

Given the attitude and preference changes that occur over time, it is most valuable to measure device usage at a long-term postoperative point. It was a very positive outcome that 41 of the 57 children (72%) were using BiCIs full-time at a median time of 3.8 years post bilateral implantation. Interestingly, of the eight children who received CI-2 over the age of 12 years, seven used BiCIs full-time and the eighth wore BiCIs at school and in more difficult listening situations. It is likely that the primary decision-making responsibility taken by these older children was relevant to their high levels of motivation to use BiCIs. The result is in contrast to a previous finding that only 64% of adolescents used BiCIs full-time, although the minimal experience of some participants in this earlier study may have been relevant to this outcome (Redfern & McKinley 2011).

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For children in the present group who were bilaterally implanted before 12 years of age, the proportion using BiCIs full-time was 69% for all children, and 63% for the sequentially implanted children. For previous groups which were relatively similar in terms of mean age at CI-1 and CI-2, and time between CIs, the reported proportion of full-time users was similar for a group at 24 months postoperative (Sparreboom et al. 2012), but far higher for a group examined at both 18 months (90.1%) and 36 months (91%) (Scherf 2009a,b). The higher full-time usage rate may have been due to differences in preoperative counseling and selection procedures across countries, as these procedures determine which families request BiCIs and which families are offered BiCIs. The availability of resources to provide postoperative support to help children to

learn to listen with CI-2 and to encourage full-time BiCI may also influence the rate of full-time use.

For the 16 children not using BiCIs full-time, it is important to consider device preference and reasons for less than full-time use in order to gain a complete clinical picture. As indicated in Table 2, only 13 children who used BiCIs less than full-time had a consistent preference for using a UniCI. The three other children considered BiCIs advantageous in many situations, but chose a reduced level of stimulation some of the time by wearing just one or no CIs. An understanding of the reasons why the BiCIs are not used full-time may allow the provision of more targeted postoperative support.

The parents who were able to compare performance with BiCIs versus a UniCI spontaneously identified a range of areas in which superior performance was demonstrated with BiCIs. The most common area, localization, was identified by 53.5% of parents; this was followed by less need for repetition (44.2%), increased responsiveness (25.6%), and improved listening in noise (18.6%). Superior performance in all of these areas, but particularly the first three, would contribute to ease of communication and facilitate social interaction, thus benefitting family and peer relationships. It may also be indicative of an increased awareness of, and connectedness with, the auditory environment, thus allowing the listener to quickly identify and respond to changes therein.

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Of the four most commonly reported areas of superior performance with BiCIs, only localization and listening in noise were also referred to in previous studies. The proportion of parents in the present study reporting superior localization was within the broad range reported previously. Most studies have identified improvement for more than half (55% to 76%) of BiCI users of all ages (Sparreboom et al. 2012; Scherf et al. 2009a; Redfern & McKinley 2011), although a less

experienced group mostly demonstrated poor localization (Fitzpatrick et al. 2011). This latter result emphasizes the importance of continuing to assess and report outcomes in the longer term. The proportion of parents in the present study reporting improved listening in noise (18.6%) was far lower than the 80% reported by Scherf et al. (2009a) as demonstrating "better speech understanding in quiet and noise". It is possible that parents have described similar types of improvements in different ways; for example, faster and more accurate responses could equally well be described as "better speech understanding". There are also clear methodological differences which are likely to have had an impact on the differences in the proportions of children demonstrating improvement and the reported areas of improvement. In the current study, parents were specifically asked if they were able to make a comparison of performance at the time of interview using a UniCI versus BiCIs, and an entirely open-ended question was then posed. Around 18% of parents could not make the comparison because their child only ever wore BiCIs, so that outcomes for these committed BiCI users were excluded from the assessment of bilateral benefit. In addition, the majority of the children had a current BiCI usage rate of more than 90%, so that parents had limited opportunities for observing and comparing performance in the two device conditions. The opportunities which did occur were more likely to involve less challenging listening situations in which additional benefit from BiCIs may have been minimal, given that anecdotal reports indicated that a UniCI was more often used in easier listening situations, such as watching TV at home. Some parents had a good opportunity to observe differences when their child was required to use only a UniCI due to a need for repair or maintenance. In contrast, in previous studies respondents were asked to identify changes in performance that had occurred since bilateral implantation; such changes may have been due to the use of BiCIs and/or due to development and auditory experience. Also, questions commonly referred to changes in performance in specified areas of listening. For these reasons, it is not

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surprising that the proportion of children in the current study identified as demonstrating superior performance with BiCIs in particular areas was lower than has been reported in previous studies.

In the present study, when comparing performance with BiCIs versus a UniCI, parents also identified a number of psychosocial benefits, such as increased confidence and decreased fatigue. Although each such difference was reported by only one to two parents, it is of significant interest that such benefits were spontaneously identified by parents when the focus of the questionnaire was listening performance. Psychosocial benefits have been reportedly previously, with 30% of parents reporting positive behavioural changes for young children (Scherf et al. 2009a), and 84% of adolescents reporting increased confidence in group conversations (Redfern & McKinley 2011). Again, with respondents specifically asked to identify behavioural or quality of life benefits, these higher rates may be due to the methodological differences across studies.

Of the 47 parents able to compare everyday listening performance with each CI alone, 49% reported that performance with CI-2 was at least equivalent to that with CI-1. This was a very positive result but was somewhat surprising, given that this group included children bilaterally implanted as late as 9.6 years of age, with a time between CIs of up to 8 years. A previous study also reported that half of their small group of 15 children demonstrated equivalent performance with each CI (Fitzpatrick et al. 2011). Achieving equivalent performance with both CIs has significant advantages, including being able to take advantage of the headshadow effect irrespective of the location of a noise source, not needing to orientate the better ear to the speaker, being able to converse equally well with a partner located to either side, and having an effective back-up device. In contrast, if there is a significant performance differential, the listener is likely to be receiving more useful auditory information from one implant than the other and/or focusing

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more attention on the signal from one implant, a situation which is unlikely to be conducive to the development of binaural listening skills.

As noted previously, only each family can judge if their unique experience of bilateral implantation was worthwhile. In the current study, 86% of parents overall and 84% of parents of sequentially implanted children considered bilateral implantation to have been worthwhile. The results for specific groups of sequentially implanted children are not consistent with those reported previously. For those receiving CI-2 before age 12 years, 81% of parents in the current study regarded CI-2 as worthwhile, compared with 100% of parents in previous studies expressing "no major disappointments or regrets" (Fitzpatrick et al. 2011) or indicating that they would "choose a second CI again" (Scherf et al. 2009b). The emphasis in the current study on balancing the benefits with the risks and costs is likely to have contributed to this difference across studies. Given that it has been reported elsewhere that older children may take longer to adapt and to learn to use CI-2 (Bohnert et al. 2006; Galvin et al. 2010), the expenditure of time, effort, and emotion is likely to be higher for adolescents and young adults. Nevertheless, for children receiving CI-2 after 10 years of age, 91% of parents in the current study considered CI-2 worthwhile. A lower rate of 68% was found previously for a less experienced group which provided self-reports (Redfern & McKinley 2011). As noted above, adolescents may value benefits, particularly indirect benefits, differently to parents. The outcome that a high proportion of parents of children of all ages considered that bilateral implantation was worthwhile will be of value to families who are considering BiCIs.

Another very valuable aid to helping families to make informed preoperative decisions is information about the relationship between outcomes with BiCIs and demographic characteristics. In the present study, support for the existence of a relationship was not consistent

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across analyses for the majority of the functional outcome measures and the factors of age at BiCIs and time between CIs. The exception was performance with CI-1 alone versus CI-2 alone. For all children, and for the sequentially implanted children only, the group achieving performance with CI-2 which was at least as good as that with CI-1 was significantly younger and had significantly less time between CIs. In addition, compared with older implanted children, the group bilaterally implanted before 3.5 years (and with 2 years or less between CIs) contained a significantly higher proportion of children who achieved performance with CI-2 which was at least as good as that with CI-1. Given the high correlation between them, it is not possible to identify if either of the demographic factors had more impact than the other. For the remaining functional outcome measures, results were inconsistent. There was no monotonic relationship between the multiple-category outcomes of adaptation to BiCIs or BiCI usage and the demographic factors of age at BiCIs and time between CIs. In addition, for the outcome measures of adaptation to BiCIs, BiCI usage, device preference, and attitude towards CI-2, the group with a positive outcome was not significantly different to the neutral/negative outcome group in terms of age at BiCIs or time between CIs. In contrast, when compared with older implanted children, the group bilaterally implanted before 3.5 years contained a significantly higher proportion of children (at least 84%) who achieved a positive outcome on the measures of adaptation to BiCIs, BiCI usage, device preference, and attitude towards CI-2. The group also had 2 years or less between implants. This finding is consistent with Sparreboom et al.'s (2012) clinical observation that children with less than 2 years between CIs had no preference for CI-1 over CI-2, whilst those with more than 2 years between CIs preferred CI-1. Unfortunately, no statistical analysis was conducted by the authors as the short delay group was too small.

In line with the inconsistent statistical evidence of a relationship between age at BiCIs or time between CIs and functional outcomes is the variation across individuals. One child received CI-2

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at 1 year 1.6 months of age, only 2.8 months after CI-1, yet 4-year postoperative outcomes included continuing difficulty adapting to CI-2, a usage rate of less than 30%, a negative attitude towards CI-2, and a preference for using CI-1. Similar poorer outcomes occurred for another child sequentially implanted at 3 years 6 months after a delay of only 2 years. In contrast, consistently positive functional outcomes, with the exception of equivalent performance between CI-1 and CI-2, were reported for an individual sequentially implanted at age 19 years 3 months after an extensive delay of almost 17 years between CIs. Furthermore, two older children sequentially implanted at around 8 to 9 years, after a delay of 7 to 8 years, achieved the unexpected outcome of equivalent performance with CI-2 and CI-1. Although influential, it is clear that age at BiCIs and/or time between implants cannot be used alone to make preoperative predictions of functional outcomes for individuals.

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In summary, at a median time of 3.8 years post bilateral implantation, three-quarters of the group examined used BiCIs full-time and the proportion of non-users or very minimal users was just 9%. Eighty-eight percent of parents reported performance was superior using BiCIs versus a UniCI (n = 40), or that their child only ever wore BiCIs (n = 10) so that an estimate of bilateral benefit could not be assessed. The most commonly identified areas of superior performance were localization, less need for repetition, and responsiveness. At the time of interview, the majority preferred BiCIs (70%), had a positive attitude to CI-2 (72%), and considered CI-2 worthwhile (86%), whilst around half demonstrated similar performance with CI-2 compared with CI-1. Overall, functional outcomes were primarily positive across all ages, even when the delay between CIs was long. Nevertheless, there was some evidence of a critical point between 3.5 and 4 years of age, with an increased likelihood of positive functional outcomes when BiCIs were received before this age and/or with less than two years between CIs. The results of this study are important for evidence-based preoperative counseling, which will help families to make

informed decisions and to develop appropriate postoperative expectations. The results are also important for the development of best-practice clinical management approaches which will support and encourage the minority of children and young adults who have difficulty adapting to bilateral implants and/or who do not easily achieve full-time device use.

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Supplemental Digital Content

Supplemental Digital Content 1. Text file which is a copy of Bilateral Cochlear Implants for Children and Young Adults Long-term Follow-up Questionnaire used in this study. pdf

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