

Can adolescents and young adults with prelingual hearing loss benefit from a second, sequential cochlear implant?

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Abbreviations:

CI-1: first cochlear implant
CI-2: second cochlear implant
SSQ: Speech, Spatial, and Qualities of Hearing Scale
AdSpon: Adaptive Spondees in Noise Test
SNR: signal-to-noise ratio
BiCI: bilateral cochlear implants

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Abstract

The study aimed to determine if adolescents/young adults gained additional perceptual benefit from sequential bilateral cochlear implants within 12 months, and to document adaptation to the second implant. Assessments comprised a pediatric version of The Speech, Spatial and Qualities of Hearing Scale (SSQ), anecdotal reports of device use and daily listening, and the Adaptive Spondee Discrimination Test (AdSpon).

All nine participants achieved full-time use of, a preference for, and superior daily listening with, bilateral implants. Eight participants were comfortable using the second implant alone, and two achieved similar daily listening with either implant alone. SSQ ratings were higher post-operatively for the majority of participants. AdSpon performance was superior bilaterally for five participants with noise ipsilateral to the first implant, but not contralateral. Unilateral performance with either implant was similar for one participant.

A second implant may provide additional benefit up to 19 years of age, even with congenital hearing loss and >16 years between implants. Families and clinicians should understand the aspects of second-implant candidacy and post-operative use that are unique to adolescents/young adults.

Introduction

The provision of bilateral cochlear implants to children with profound hearing impairment of congenital or pre-lingual onset has become increasingly common. There are many reports in the scientific literature confirming that children may gain additional benefit from a second cochlear implant, received either simultaneously with the first implant or sequentially at a later date. Studies have demonstrated improved scores on speech perception testing in quiet and/or in noise (Kühn-Inacker et al. 2004; Galvin et al. 2007; Scherf et al. 2007; Wolfe et al. 2007), with the majority of the benefit in noise being attributed to the headshadow effect. Further studies have demonstrated improved performance on relatively simple sound source identification tasks. Improvement has been shown for the majority of young participants evaluated on left versus right tasks (Beijen et al. 2007; Galvin et al. 2008), and also on two-alternative, forced choice evaluations of minimum audible angle (Litovsky et al. 2006b). Reports of functional benefit in daily life, such as improved performance in complex listening situations, increased self-esteem, or longer attention span, have been based on feedback from parents and children (Kühn-Inacker et al. 2004; Bohnert et al. 2006; Galvin et al. 2007; Galvin et al. 2008). The aim of the present study was to determine if children with a profound, prelingual hearing loss who used a single cochlear implant could receive additional perceptual benefit from sequential bilateral implants if the second implant was received in adolescence or young adulthood.

Although the research so far has made it clear that there is significant potential benefit from bilateral cochlear implants, outcomes have varied amongst individuals, and some children have gained no benefit from a second cochlear implant. It is important that the factors contributing to

benefit or lack thereof are investigated. This will allow clinicians to develop suitable selection criteria and to provide children and families with appropriate pre-operative counselling. An understanding of such factors will also inform the development of post-operative management and habilitation approaches that will maximize an individual's benefit from bilateral implants. These issues are particularly important for children receiving sequential bilateral cochlear implants, who need to learn to use a second implant after what may be a significant period spent focused on the auditory input provided by the first implant.

Researchers have attempted to relate the degree of bilateral benefit or progress with the second implant to particular factors, despite the often small and/or very heterogeneous groups involved in the studies. As was the case with unilateral implants, the effect of age at implant has been of particular interest (Peters et al. 2007; Scherf et al. 2007; Wolfe et al. 2007), but there is also the new variable of time between implants (Manrique et al. 2007). The general conclusion from these studies is that children who are younger at the time of their second implant will gain more benefit, and that shorter duration between implants is also associated with greater benefit.

Although the effect of age at implant has been considered, the majority of studies so far have focused on the benefit provided to children who received their second implant before ten years of age, with few published studies in the literature involving adolescent or older participants. One study reported group results for three age groups, with Group III (aged 8 to 13 years) demonstrating a statistically significant group bilateral advantage on closed-set spondee discrimination in noise, but not on sentence perception in quiet (Peters 2007). A further study included four participants aged 10 to 14 years, and assessed localization acuity using a two-

alternative forced-choice minimum audible angle paradigm (Litovsky et al. 2006b). Three of the participants demonstrated superior performance in the bilateral compared to the unilateral condition, though only group statistics for the overall group (aged 3 to 14 years) were presented. One of these adolescents also participated in Litovsky et al. 2006a, and demonstrated superior performance in the bilateral condition on closed-set spondee discrimination testing in one noise condition, though not in the remaining two noise conditions or in quiet. Although these three studies included some adolescent participants, they did not establish if adolescents, as a group or as individuals, gained significant additional benefit from a second implant, because only group statistics and/or group results including young children were presented. No studies published in the literature report outcomes for those with early onset childhood hearing loss who received a second implant as a young adult.

It is important to have information regarding second implants that is specific to adolescents and young adults, given the large number of potential second implant candidates in this age range worldwide. This group is also very different to younger children because they are able to make their own decision regarding surgery (or contribute significantly to the decision), they are generally more responsible for their own device use, and they are more independent. It is this independence which means that they are more often communicating with people who do not make adaptations for their listening abilities in environments that are less controlled and less predictable than those of the school or home. Furthermore, as the adolescent moves towards adulthood, they will be in social, work and learning situations in which the failure to communicate successfully is more likely to have significant consequences.

Materials & Methods

Participants

The research project was publicized to clients and their families attending the Royal Victorian Eye and Ear Hospital/University of Melbourne Cochlear Implant Clinic (the Implant Clinic). Prior to July 2007, when eight of the nine participants in the present study were recruited, the Implant Clinic was offering second cochlear implants to older children only if they were willing and able to participate in research. This restriction was in place due to the lack of evidence in the literature regarding the likely benefit for this group. It was made clear to families that the provision of a second implant over the age of 10 years was considered experimental. Outcomes for the initial participants led to the removal of this restriction in July 2007, and the ninth participant (P42) joined the study in September 2007.

The age criterion applied was the United Nations definition of adolescence, this being 10 years or over. The selection criteria relating to the first implant were full-time use of that implant and no evidence or reports of compromised functioning of that implant (examples of such evidence or reports would be a limited number (defined as <15) of functioning electrodes or significantly poorer than expected speech perception skills). There was no age-at-first-implant criterion. The audiological criteria were a profound hearing loss in the unimplanted ear (i.e. pure-tone average \geq 90dB SPL), and a congenital or pre-lingual onset of hearing loss. The medical criteria were that imaging results suggested that a full insertion of the array was likely and that there were no contraindications to surgery. The final selection criteria were that no cognitive difficulties or developmental delay had been reported by teachers or other professionals, and that the child was

Table 1: Hearing loss and contralateral hearing aid use information for the nine participants.

| Participant | Etiology/Age at onset of profound loss | Pre-operative 3FA(dB) ¹ | | Contralateral aid use between 1 st and 2 nd implant operations | |
|-------------|---|------------------------------------|--|--|--|
| | | CI-1 ² ear | CI-2 ³ ear | Consistent, long-term use | Comments on aid use |
| 2 | unkn/congenital? ⁴ | 115 ⁵ | 103 ⁶ | none | occasional for 12 mo pre CI-2 |
| 4 | unkn/congenital? ⁷ | 102 ⁸ | 108 ⁶ | none | full time 3 mo aid trial pre CI-2 |
| 12 | unkn/congenital? ⁹ | 110 ⁵ | 115 at 500Hz ⁶ | none | full time 3 mo aid trial pre CI-2 |
| 18 | unkn/congenital? ⁹ | 122 ⁶ | 118 ⁶ (was 97 until 3mo prior to op) | full time | discarded 3 mo pre CI-2 due to hearing change |
| 24 | genetic/congenital ¹⁰ | 120 ⁸ | 108 ⁶ | none | discarded 11 yr pre CI-2 |
| 25 | meningitis/9mo | 122 ⁸ | 120 ⁶ | none | -- |
| 26 | WVA ¹¹ /congenital | NR ^{6,12} | 110 ⁶ | none | discarded 2 yr 6 mo pre CI-2 |
| 28 | unkn/congenital? ¹³ | NR ¹⁴ | NR ⁶ | none | discarded 3 yr pre CI-2 |
| 42 | unkn/congenital | 107 ⁵ | 98 ⁶ | full time | -- |

¹ Pre-op 3FA: Average of unaided thresholds at 500Hz, 1kHz, and 2kHz obtained for the specified ear prior to the operation to implant that ear. ² CI-1: first cochlear implant. ³ CI-2: second cochlear implant. ⁴ Assumed congenital; loss suspected at 6 mo, diagnosed at 9 mo. ⁵ Obtained via Steady State Evoked Potential (SSEP) testing. ⁶ Obtained via standard audiometry. ⁷ Assumed congenital; loss suspected at 12 mo, diagnosed at 18 mo. ⁸ Obtained via play audiometry. ⁹ Assumed congenital; diagnosed at 11 mo. ¹⁰ Assumed congenital; diagnosed at 20 mo, loss suspected earlier. ¹¹ WVA: Wide vestibular aqueduct syndrome. ¹² NR: No auditory responses at audiometer limits. ¹³ Assumed congenital; diagnosed at 7mths. ¹⁴ Test method not specified.

likely to be able and willing to complete the research assessment battery. In the period October 2003 to September 2007, when recruitment to the present study was occurring, all of the children/young adults over the age of 10 years who requested to be considered for sequential bilateral implantation by the Implant Clinic fulfilled the above criteria and were recruited to the study; i.e., no candidate was rejected.

The hearing loss and contralateral hearing aid use details of the nine participants are presented in Table 1. As this group of adolescents and young adults is part of a larger cohort participating in the ongoing evaluation of bilateral implants, participant numbering is not consecutive. Prior to their first implant, all participants wore bilateral hearing aids, though usage was very poor for P2, who consistently removed the aids.

Table 2 presents information related to age at implant switch-on, ear choice, and performance with the first implant (CI-1). The age at first implant varied significantly from a minimum of 1yr 9mo to a maximum of 11yr 3mo, with a mean age of 4yr 6mo (SD: 3yr 1mo). The mean age at second implant (CI-2) was 14yr 5mo (SD: 3yr 6mo), with the mean time between implants therefore 9yr 11mo (SD: 3yr 2 mo). All participants used only oral/aural communication, were integrated into mainstream classrooms or attended a tertiary institution and, as indicated by the sentence perception scores in Table 2, were very successful users of their first implant. Details of the participants' implant types, speech processors, speech processing schemes and additional processing features (used by P42 only) are provided in Table 3. All participants used Nucleus CI22 and/or CI24 implants (Cochlear Ltd, Lane Cove, Sydney, Australia), and were assessed using the processor and standard program provided by their implant clinician. Six participants

Table 2: Age at implant switch-on, first implanted ear, basis of ear choice, indicators of performance and age at testing with the first implant, and time between implants for the nine participants.

| Participant | Age at CI-1 ¹ | CI-1 ear | Test scores using CI-1 alone | | | Age at CI-2 ⁵ | Time b/w |
|-------------|--------------------------|-----------------------------|------------------------------|--|------------------------|--------------------------|--------------|
| | switch-on (yr;mo) | /Basis for ear choice | Test age (yr;mo) | PPVT ² age equiv. score ³ / | BKB ⁴ score | switch-on (yr;mo) | CIIs (yr;mo) |
| 2 | 1;9 | R/R-handed | 8;6 | 8;6 | 96% ⁶ | 11;4 | 9;6 |
| 4 | 4;3 | R/slightly poorer hearing | 14;2 | 13;2 | 94% ⁷ | 14;5 | 10;2 |
| 12 | 1;10 | L/slightly poorer hearing | 6;10 | 4;8 | 86% ⁷ | 10;2 | 8;4 |
| 18 | 6;6 | L/poorer hearing | 11;10 | 9;1 | 98% ⁷ | 12;3 | 5;9 |
| 24 | 2;7 | L/slightly poorer hearing | 12;7 | 10;5 | 100% ⁷ | 19;3 | 16;8 |
| 25 | 5;10 | L/slightly poorer hearing | 10;1 | 6;10 | 96% ⁷ | 16;3 | 10;6 |
| 26 | 11;3 | R/slightly poorer hearing | 16;3 | 15;11 | 90% ⁷ | 19;9 | 8;6 |
| 28 | 2;1 | L/none (equivalent hearing) | 14;3 | not tested | 90% ⁷ | 14;6 | 12;5 |
| 42 | 4;0 | R/slightly poorer hearing | 9;1 | 11;8 | 78% ⁸ | 11;6 | 7;6 |

¹ CI-1: first cochlear implant. ² PPVT: Peabody Picture Vocabulary Test for receptive vocabulary. ³ Age equiv. score: Age at which a child with normal hearing would be expected to obtain the same score. ⁴ BKB: Bamford-Kowal-Bench open-set sentences; scored by % of key words correct. ⁵ CI-2: second cochlear implant. ⁶ Live-voice presentation in quiet. ⁷ Recorded presentation in quiet. ⁸ Recorded presentation at +5 signal-to-noise ratio.

Table 3: Cochlear implant types, speech processors and active features, and speech processing scheme for the nine participants at the time the results reported here were collected.

| Participant | Nucleus implant type | | Speech processor | | Processing scheme (+ any additional processing features) |
|-------------|----------------------|------------------------|-----------------------|-----------------------|--|
| | CI-1 ¹ | CI-2 ² | CI-1 | CI-2 | |
| 2 | CI22 ³ | CI24RCA ⁴ | Spectra ⁵ | ESPrIt3G ⁶ | Speak ⁷ |
| 4 | CI22 | CI24RCA | ESPrIt3G | ESPrIt3G | Speak |
| 12 | CI22 | CI24RCA | ESPrIt3G | ESPrIt3G | Speak |
| 18 | CI24M ⁸ | CI24RECA ⁹ | Freedom ¹⁰ | Freedom | ACE ¹¹ |
| 24 | CI22 | CI24RECA | ESPrIt3G | Freedom | CI-1:Speak CI-2: ACE |
| 25 | CI22M ³ | CI24RECA | ESPrIt3G | Freedom | CI-1:Speak CI-2: ACE |
| 26 | CI24M | CI24RECA | ESPrIt3G | Freedom | CI-1:Speak CI-2: ACE |
| 28 | CI24RST ⁸ | CI24REST ¹² | Freedom | Freedom | ACE |
| 42 | CI24M | CI24RECA | Freedom | Freedom | ACE (+ Whisper ¹³) |

¹ CI-1: first cochlear implant. ² CI-2: second cochlear implant. ³ CI22/CI22M: straight array implant with 22-electrodes. ⁴ CI24RCA: soft-tip curved array implant with 22-electrodes plus 2 extra-cochlear electrodes. ⁵ Spectra: body-worn speech processor for use with CI22 implant. ⁶ ESPrIt3G: ear-level speech processor capable of higher-rate processing. ⁷ Speak: peak-picking filterbank speech processing strategy. ⁸ CI24M/CI24RST: straight array implant with 22 electrodes plus 2 extra-cochlear electrodes. ⁹ CI24RECA: soft-tip curved array Freedom implant with 22-electrodes plus 2 extra-cochlear electrodes. ¹⁰ Freedom: ear-level speech processor which may be used with body-worn controller. ¹¹ ACE (Advanced Combination Encoder): filterbank strategy with variable high-rate presentation. ¹² CI24REST: straight array Freedom implant with 22-electrodes plus 2 extra-cochlear electrodes. ¹³ Whisper: a fast-acting compression circuit that increases the Instantaneous Input Dynamic Range

used the same strategy and, where relevant, the same additional processing features in each implant. The remaining three participants used the more advanced ACE strategy with CI-2 whilst retaining the Speak strategy for CI-1. This was due to the limitations of their first implant, which was a CI22, (P24 and P25) or personal preference (P26).

The participants received standard post-operative audiological and medical care from the Implant Clinic. This included mapping and device maintenance, but not habilitation or auditory training. Specific habilitation aimed at developing listening skills with CI-2 was received at school in fortnightly or weekly sessions by P4 and P28 respectively. Daily sessions were provided to P2, P12 and P42 (reduced to weekly after 5 months for P42).

Assessment Battery

The research assessment battery consisted of a speech perception in noise test, a questionnaire, and monthly interviews to collect anecdotal reports of device usage and performance.

Anecdotal Reports of Device Use and Daily Performance

Anecdotal reports of device use and daily performance with either device alone and with both together were collected in interviews at two weeks after switch-on, at one month after switch-on, and at monthly intervals thereafter. For the majority of the participants, this involved monthly telephone (or occasional face-to-face) interviews with the parent, while P26 was interviewed directly via email, and P24 via a combination of telephone and email. For P25, the parent was interviewed by phone and also provided information supplied by the participant. Irrespective of the interviewee or the method, the same questions were asked, and the interviewer took care to

ask for information in an unbiased manner. While general information was also collected, the interviews focused on whether or not particular milestones had been achieved. Milestones related to the use of the second implant alone were: if the adolescent/young adult was comfortable using the second implant alone, and if their daily listening performance using the second implant alone was similar to that using the first implant alone. Milestones related to the use of bilateral implants were: if the adolescent/young adult used the bilateral implants full time, if they were happy to use the bilateral implants, if they preferred to use the bilateral implants rather than either implant alone, and if their daily listening performance was superior when using the bilateral implants compared with one implant.

Questionnaire

The participants' performance in everyday listening situations was evaluated using a questionnaire derived from "The Speech, Spatial and Qualities of Hearing Scale (SSQ)". This original questionnaire for adults was designed with some emphasis on binaural hearing functions and has been rigorously assessed (Gatehouse and Noble, 2004; Noble and Gatehouse, 2004). The first author of the present paper modified the adult SSQ to produce a version for parents of children with impaired hearing, a version for older children, and a version for teachers (copies of the Scales can be obtained by emailing the corresponding author). The first two versions have been used previously to evaluate bilateral implant use by children (Beijen 2007; Galvin et al. 2007), however no studies have yet evaluated the reliability or validity of these adapted versions of the SSQ. Regarding the parent or teacher versions, the authors acknowledge that there are potential difficulties in asking a third person to rate the listening performance of a child with a hearing loss. Ratings in this circumstance can only be based on observations. To maximize the accuracy of ratings, the third-person versions of the scale require an observation period in which

the teacher or parent observes the child in specified listening situations prior to providing their ratings. The authors acknowledge that providing accurate ratings would require an intimate knowledge of the child's listening abilities and typical patterns of performance, and that the rater had spent a significant amount of time with the child on a regular basis. Regarding the children's version, there are potential difficulties with the language and cognitive skills required to complete the scale. The chronological and language equivalence age at which it would be reasonable to administer the children's scale has not been established. A further important difficulty arises with the children's version if the interviewee has a congenital or early onset hearing loss. Even as adolescents or young adults such individuals may lack a full understanding of their disability, in that it is difficult to conceive of the listening skills of people with normal hearing if one has never experienced normal hearing. The authors' experience when administering the children's version of the SSQ was that the interviewees often struggled to rate their own performance. Further exploration of the use of the children's version is required. As a result, where possible in the present study, "The Speech, Spatial and Qualities of Hearing Scale for Parents of Children with Impaired Hearing" was administered. In all cases, the authors consider that the parents making the ratings were very knowledgeable regarding their child's listening skills and spent time with the child on a daily basis. Where participants did not spend sufficient time with their parents for an evaluation of their performance to be made "The Speech, Spatial and Qualities of Hearing Scale for Children with Impaired Hearing" was administered. This occurred for P25 and P26.

In each version of the Scale, there were three sections examining speech perception (in quiet, in groups and/or in noisy or reverberant environments), spatial hearing (the location, direction and

distance of sounds), and quality of hearing (segregating and identifying sounds and listening effort). In each item a type of listening situation was described and the parent or the participant rated their child's or their own performance in that situation. For example, "You are talking to your child in a room in which there are many other people talking. Can your child follow what you say?" Ratings were made on a representation of a ruler extending from 0 to 10; for the majority of items, the left-hand end was labelled "Not at all" and the right-hand end was labelled "Perfectly". There were also options to respond: "Do not know", "Not applicable", or "Would not hear it". To maximize the accuracy of their ratings, parents were asked to complete a minimum of one week of observation of their child in the specified types of listening situations prior to the completion of each of the three sections. The SSQ was administered via a face-to-face interview or, for some parents, via a phone interview. The interviews were conducted in the 6 weeks prior to receipt of the second implant and again after 12 months of bilateral implant use. At this latter point the interviewee was not able to consult their pre-operative responses.

Speech Perception

Speech perception was assessed using AdSpon (Adaptive Spondee Discrimination Test). AdSpon is a software-controlled, four-alternative, forced-choice spondee discrimination test presented in background noise. Twenty spondees served as stimuli and foils, these being: beanbag, birthday, blackbird, blackboard, dollhouse, eggshell, eyebrow, eyelash, football, goldfish, hairbrush, highchair, ice-cream, light switch, playground, rainbow, scarecrow, shoelace, teapot, and toothbrush. There were four test lists, each consisting of 10 sets of four spondees. In creating test lists 1 to 4, the spondees were pseudo-randomly selected with the following criteria also applied: each of the 20 spondees occurred twice in each list, no spondee occurred in two consecutive sets of four spondees, and the similar pairs of words (hairbrush/toothbrush,

eyebrow/eyelash, and blackbird/blackboard) did not occur in one set. The test procedure involved, for each set of four spondees, the presentation of four pictures on a touchscreen. The software randomly selected one of the four spondees for auditory presentation as the test stimulus. Beginning with a randomly-selected starting list, the lists were presented in sequential order until the criteria for ceasing testing (outlined below) were fulfilled.

Continuous speech-shaped broadband noise was used. An adaptive procedure was used to determine the signal-to-noise ratio (SNR) at which the criterion level of performance (79.4%) was achieved (Levitt 1971). In this process, the SNR was decreased following three consecutive correct responses and increased following any incorrect response. To maximise the efficiency of the test, the initial SNR change of 10dB was reduced to 5dB after two reversals, and to 2dB after a further four reversals; a “reversal” being a turn around in the direction of SNR change. Testing ceased after 12 reversals. The test output was the average SNR at the final six reversal points. The initial presentation level was 62dBA for spondees and 42dBA for noise. To reduce the SNR, the noise level was increased until a maximum of 62dBA was reached, from which point on SNR reductions were achieved by decreasing the spondee level so that a comfortable loudness level was maintained (Blamey et al. 2000).

Testing was conducted in a low-reverberation sound-proof booth. The participant was seated 1.15m from loudspeakers positioned at head height at 0° and at 90° to the right and left. Test sessions consisted of training and testing phases. The initial step in the training phase involved familiarising the participant with the test stimuli; two auditory presentations of each spondee were made in quiet with the pictorial representation of the relevant spondee displayed on the

touchscreen. The tester confirmed that the participant could identify each picture. The second step in the training phase involved familiarising the participant with the test procedure; a practice test list of at least 10 stimuli was presented in adaptive noise. In the testing phase, sets of four pictures were presented with accompanying auditory presentation of the randomly-selected test stimuli. The participant responded by selecting a picture on the touchscreen. This process continued until the criterion of the adaptive noise procedure was met. No feedback was provided and guessing was required. Testing occurred in three device conditions and two noise conditions (six conditions in total), with the order of conditions alternated across sessions and participants. The three device conditions were CI-1, CI-2 and bilateral cochlear implants (BiCI). When the device condition was changed, the participant was engaged in five minutes of conversation to allow adaptation to the new condition. The two noise conditions were speech from 0° with either ipsilateral or contralateral noise. In the ipsilateral condition, noise was presented from 90° on the side of the active implant (or the side of CI-1 in the case of the BiCI condition). In the contralateral condition, noise was presented from 90° on the opposite side to the active implant (or the opposite side to CI-1 in the case of the BiCI condition).

Testing in the CI-2 versus CI-1 conditions evaluated the new second implant relative to the original implant, and determined the superior implant for comparison with the BiCI condition. Testing in the unilateral versus BiCI conditions evaluated the additional benefit provided by two implants relative to one (whichever had been shown to be the superior implant). Based on research with participants with normal hearing, there are a number of potential advantages to listening with two ears rather than one: these include binaural summation, binaural redundancy, the headshadow effect and binaural unmasking (Dillon, 2001). As it is difficult to isolate the

contribution of these individual advantages to overall benefit, research is yet to conclusively determine whether and to what degree these individual advantages are available to all or some children using bilateral cochlear implants. Theory suggests that individual children may access more of these advantages if they develop equivalent listening skills with either implant and/or have previous binaural listening experience. As noted, it is difficult to conclusively isolate the contribution of each individual advantage, and this study does not do so. Testing in the unilateral versus BiCI device conditions with ipsilateral noise primarily evaluated the size of the headshadow effect; that is, the benefit gained from the addition in the BiCI condition of the implant that is shadowed from the noise source. Testing in the unilateral versus BiCI device conditions with contralateral noise primarily evaluated the degree of binaural unmasking occurring; that is, the improvement in perception in the BiCI condition when the brain uses the signal at the ear with the poorer SNR (in this case, the contralateral ear) to effectively improve the SNR at the ear with the better SNR. Theoretically, there may have been other processes contributing to any improved perception in the BiCI relative to the unilateral condition.

Results

The participants' age and amount of experience with each device at the time the post-operative questionnaire and speech perception test were administered are presented in Table 4. All participants had approximately 12 months bilateral implant experience.

Table 4: Age and amount of experience with each implant at the time the post-operative speech perception and SSQ Scale results reported here were collected.

| Participant | Age | Time since start-up (yr;mo) | |
|-------------|---------|-----------------------------|-------------------|
| | (yr;mo) | CI-1 ¹ | CI-2 ² |
| 2 | 12;5 | 10;7 | 1;1 |
| 4 | 15;7 | 11;4 | 1;2 |
| 12 | 11;2 | 9;4 | 1;0 |
| 18 | 13;5 | 6;10 | 1;1 |
| 24 | 20;3 | 17;8 | 1;0 |
| 25 | 17;4 | 11;6 | 1;1 |
| 26 | 20;10 | 9;7 | 1;1 |
| 28 | 15;6 | 13;5 | 1;0 |
| 42 | 12;7 | 8;7 | 1;1 |

¹ CI-1: first cochlear implant. ² CI-2: second cochlear implant.

Anecdotal Reports of Device Use and Daily Performance

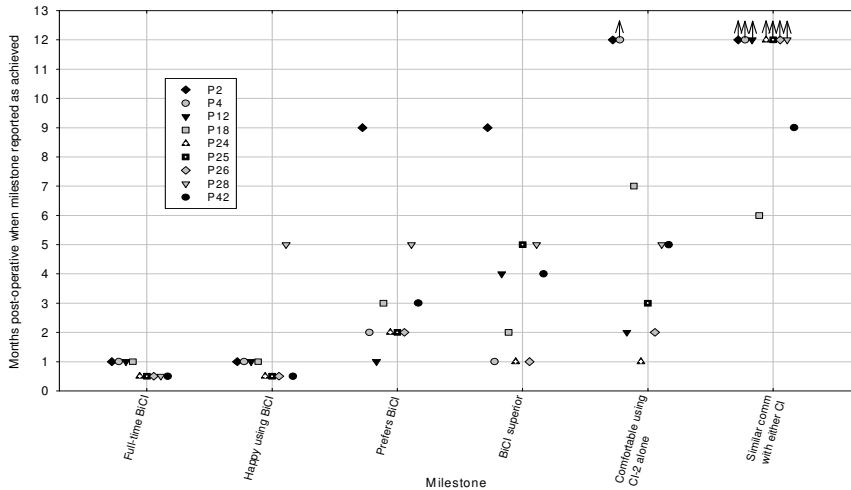


Figure 1: Post switch-on point (in months) at which specified milestones were reported as having been achieved by individual participants. Arrows indicate the milestone was not yet achieved.

Figure 1 presents the post switch-on point in months at which the interviewee (parent or adolescent/young adult) reported that specific milestones were achieved. Regarding the milestones related to the use of bilateral implants, within one month of the second implant being switched on all nine participants were using both implants full time, and eight were happy to do so, though P28 was not happy doing so until five months experience had been gained. Regarding the milestones related to performance with bilateral implants, a preference for using bilateral implants was reported for seven participants within three months of switch-on, and for P2 and P28 within 9 months. The demonstration of superior daily listening was reported for eight participants within 5 months but, again, was not reported for P28 until 9 months. Achievement of these two bilateral implant milestones was reported at the same post switch-on point (or within

one month of each other) for seven of the participants. In contrast, for P12 and P25, their preference for bilateral use was reported three months before superior listening with bilateral implants was reported. The achievement of milestones related to the use of CI-2 alone was not reported so soon. Being comfortable using CI-2 alone was achieved by most participants within 5 months, though P18 and P2 took 7 and 12 months respectively, and P4 had not achieved this milestone at the 12 month post switch-on point. The majority of participants did not achieve similar daily listening performance with CI-2 alone as with CI-1 alone, though this was reported for P18 and P42 at 6 and 9 months respectively. Table 5 presents details of usage of CI-2 alone and bilateral implants as reported by parents and young adults. The participants' attitude towards the use of CI-2 alone, and the perceptual level achieved and areas of improvement when using BiCIs are also included.

Questionnaire: The Speech, Spatial and Qualities of Hearing Scale

Participant 28 did not complete the SSQ, as he was implanted interstate and so was not a participant in the research study pre-operatively. For the remaining eight participants, pre-operative ratings were obtained in the six weeks prior to the operation in their everyday listening condition. This condition was CI-1 alone for five participants, and CI-1 plus hearing aid for P42, a long-term consistent user of a contralateral hearing aid, and for P4 and P12, who were using a hearing aid as part of a 3-month pre-operative aid trial. Post-operative ratings were obtained in the BiCI condition 12 months after the switch-on of CI-2. Results are from the child's (i.e., self-rating) version of the scale for P25 and P26 (who did not spend sufficient time with their parents for ratings to be made), and from the parent's version for all other participants. For both versions, the speech perception items evaluating conversation with one person in ideal listening

Table 5: Reported details of the use of the second implant and bilateral implants in the 12 months following the start-up of the second device for each participant. Reports were provided by parents and young adults.

| Participant | Participant Attitude | Use of second implant alone | | Perceptual level achieved | Use of bilateral implants Reported areas of improvement compared with 1 st implant alone |
|-------------|----------------------|---|--|---------------------------|---|
| | | Structured rehabilitation (post-op mths of occurrence) | Unstructured usage (post-op mths of occurrence) | | |
| 2 | negative | 20min/school day (1-12) | none | simple sentences | spatial awareness localizing speakers confidence |
| 4 | Neutral | 20min/f ^o night (1-6) | 0.5hr/day (1 – 2) | conversation | communication in noise & groups |
| 12 | Neutral | 15min/school day (1-12) | 1-3hrs/wk (1 – 3) | closed-set words | sound awareness localizing speakers |
| 18 | not necessary | none | none | conversation | spatial awareness localizing sounds communication in noise soft & distant sound perception confidence |
| 24 | not necessary | none | 0.5-1hrs/wk (1 – 1.5) | conversation | localizing speakers communication in groups sound clarity & quality |
| 25 | negative | none | 0.75hrs/day (1 – 1.5) | aid to lipreading | speech perception sound clarity & quality soft sound perception |

| | | | | | |
|----|----------|--|---------------------------|----------------------|---|
| 26 | Positive | none | 1.5 – 5 hrs/day (1 – 12) | aid to lipreading | spatial awareness speech perception self-monitoring speech soft sound perception |
| 28 | negative | ~0.5hr/wk (5 – 12) | 0.5 - 3 hrs/wk (4 - 12) | simple sentences | communication in noise & groups distant sound perception |
| 42 | negative | 2.5hrs/wk (1 – 2) 0.5-1.25hrs/wk (3 – 4) 0.5hrs/wk (5 – 8) | 1.5 – 2.75 hrs/wk (1 - 8) | conversation | sound awareness speech production clarity speech perception not orientating “good” ear |

Post print

conditions and on the telephone were not included in the analysis because bilateral sound input was not expected to improve performance in these situations. For the child's version, the two quality-of-hearing items which evaluated the change in perceived loudness when one device was turned off were not included because the pre-operative condition was usually unilateral.

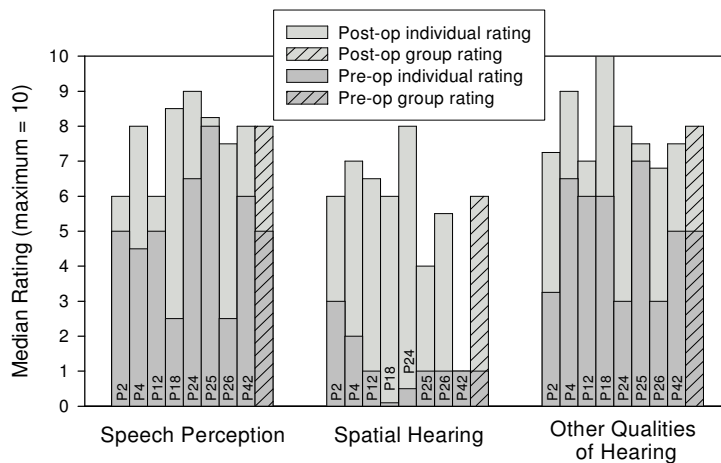


Figure 2: Median pre- and post-operative performance rating for individual participants and for the group for each section of the SSQ Scale. Ratings were collapsed across items within Section A: Speech Perception, Section B: Spatial Hearing, and Section C: Quality of Hearing. Ratings were self-ratings for P25 and P26; all other ratings were provided by a parent.

Figure 2 presents the median pre-operative and post-operative ratings for each participant and for the group in each of the three sections of the SSQ: Section A - Speech Perception, Section B – Spatial Hearing, and Section C – Other Qualities of Hearing. The number of questions in each section varied across versions (refer to Method). For those participants whose data was collected via the parent version, Figure 2 presents the median rating of 7 questions for Section A, 6 questions for Section B, and 8 questions for Section C. For those participants whose data was

collected via the child version, Figure 2 presents the median rating of 8 questions for Section A, 13 questions for Section B, and 10 questions for Section C. Where the interviewee was unable to provide a rating, the item was excluded from the results; this occurred for P2 (two questions in Section B), P12 (one question in Section B), P18 (two questions in Section C), and for P25 (two questions in Section B).

The median post-operative performance rating was higher than the median pre-operative rating for all three sections for the majority of participants and for the group; the change in the median rating being between 1 and 7.5 points on the 0-to-10 scale. The only participants to demonstrate essentially no change for some sections were P25 (for Speech Perception and Other Qualities of Hearing) and P42 (for Spatial Hearing). Participant 18 and P26 demonstrated a particularly strong pattern of improvement, with a change of more than 4 points for each section. Participant 24 demonstrated the single greatest improvement, with a change in median rating of 7.5 points for Spatial Hearing. In terms of the areas of greatest benefit, the most consistent pattern of improvement across participants, and the largest pre- to post-operative improvements, occurred for Spatial Hearing. All participants except P42 showed an improvement of at least three points for this section. For Speech Perception and Other Qualities of Hearing, the sizes of the increases in median rating were more variable across participants.

Speech Perception: AdSpon

All participants were tested in the CI-1 and BiCI conditions, and the majority were also tested in the CI-2 condition. Due to poor listening skills, P12 could not complete the test in the CI-2 condition. Due to limited availability, P2 and P4 completed only limited testing in the CI-2 condition. Two-tailed, paired t-tests ($\alpha = 0.05$) were used to determine if performance using

BiCIs was different to that using CI-1 alone, and to determine if performance using CI-1 was different to that using CI-2. Where the data failed the normality test, the Wilcoxon signed-rank test was used.

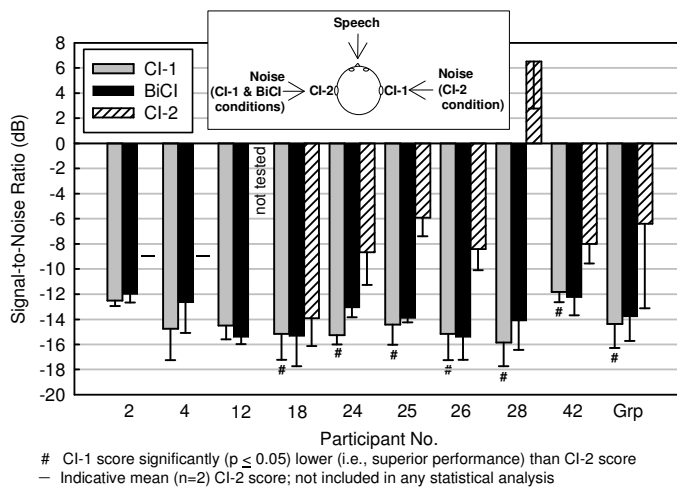


Figure 3: Mean (n = 4; P28, BiCI condition: n = 3) SNR at which the individual participants and the group achieved the criterion level of performance on the AdSpon test in the CI-1, CI-2 and BiCI conditions when noise was presented contralateral to the active implant in each unilateral condition and contralateral to CI-1 in the BiCI condition. Error bars represent one standard deviation.

Figure 3 presents the mean AdSpon results for the individual participants and for the group when the speech was presented from in front and the noise was presented contralateral to CI-1 (in the CI-1 and BiCI conditions) or contralateral to CI-2 (in the CI-2 condition). For the majority of the participants and conditions, the mean is the average result for four separate administrations of the AdSpon test. The exception was for P28 in the BiCI condition, where the mean is the average result for three administrations of the test. The limited CI-2 alone results for P2 and P4, being the

average of just two administrations of the test, are indicated in the figure for information purposes but were not included in the statistical analysis. Comparisons of performance using either device alone indicated superior performance (i.e., a significantly lower SNR) in the CI-1 compared with the CI-2 condition for all six participants tested (P18, P24, P25, P26, P28 and P42: mean difference = 8.2dB (SD = 7.4); $t(3) \geq -3.187$, $p \leq 0.050$) and for the group (mean difference = 8.2dB (SD = 7.4); $n = 24$, $W = 300.0$, $p < 0.001$). The limited scores for P2 and P4 suggest a similar result although no statistical analysis was completed. As performance with CI-1 was superior to that with CI-2, the former was used for comparison with the BiCI condition. Analysis indicated that performance in the BiCI condition was not different to that in the CI-1 condition for any of the nine participants (mean difference = 0.1dB (SD = 1.6); P25: $n = 4$, $W = 2.0$, $p = 0.87$; P28: $t(2) = 0.49$, $p = 0.65$; P2, P4, P12, P18, P24, P26, and P42: $t(3) \leq -3.15$, $p \geq 0.051$) or for the group (mean difference = 0.49dB (SD = 1.6), $t(34) = -1.76$, $p = 0.088$).

Figure 4 presents the mean AdSpon results for the individual participants and for the group when the speech was presented from in front and the noise was presented ipsilateral to CI-1 (CI-1 and BiCI conditions) or ipsilateral to CI-2 (CI-2 condition). For the individuals, the mean is the average result for four separate administrations of the AdSpon test. Comparisons of performance using either device alone indicated superior performance in the CI-1 compared with the CI-2 condition for five of the six participants tested (P24, P25, P26, P28, and P42: mean difference = -8.3dB (SD = 6.8), $t(3) \geq -4.46$, $p \leq 0.021$) and for the group (mean difference = 7.2dB (SD = 6.8), $n = 24$, $W = 294.0$, $p < 0.001$). There was no significant difference between scores in the two conditions for P18 (mean difference = -1.9dB, $t(3) = -1.17$, $p = 0.33$). Comparisons of performance in the CI-1 and BiCI conditions indicated superior performance in the BiCI condition for five participants (P4, P12, P18, P24, and P42: mean difference = 4.8dB (SD = 1.7),

$t(3) \geq 3.50$, $p \leq 0.040$) and for the group (mean difference = 3.7dB (SD = 2.7), $t(35) = 8.41$, $p < 0.001$). There was no significant difference between scores in the two conditions for the remaining four participants (mean difference = 2.4dB (SD = 1.1); P25: $n = 4$, $W = -10.0$, $p = 0.125$; P2, P26 and P28: $t(3) \leq 2.79$, $p \geq 0.068$).

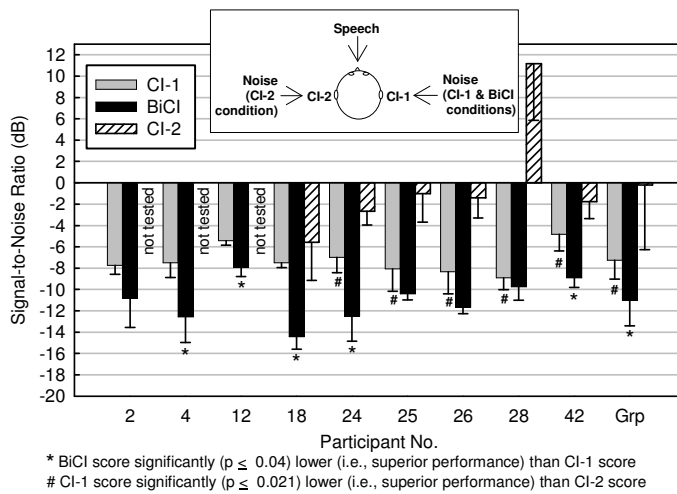


Figure 4: Mean ($n = 4$) SNR at which the individual participants and the group achieved the criterion level of performance on the AdSpon test in the CI-1, BiCI and CI-2 conditions when noise was presented ipsilateral to the active implant in each unilateral condition and ipsilateral to CI-1 in the BiCI condition. Error bars represent one standard deviation.

Discussion

Anecdotal reports indicated that these nine participants adapted to full-time bilateral implant use and were happy wearing two implants together, generally within a month of switch-on. Within five months the majority also demonstrated superior everyday listening skills with, and preferred, bilateral implants. This was somewhat surprising given that previous research has suggested a

decrease in benefit with increasing age at CI-2 or delay between implants (Manrique et al. 2007; Peters et al. 2007; Scherf et al. 2007; Wolfe et al. 2007); however no studies have provided evidence of an upper age limit beyond which no benefit is gained. Significantly more experience was required before the participants were comfortable using CI-2 alone, and only P18 and P42 achieved the milestone of similar everyday communication with either device. The overall progress of P2 and P28 also stands out somewhat, with slower achievement of most milestones.

The SSQ results indicated that all participants' performance in everyday listening situations was perceived to have improved 12 months after receiving CI-2, particularly in the area of spatial hearing. There was some variation across participants, though this appeared to be unrelated to factors such as absolute level of pre-operative ratings. A particularly strong pattern of increased ratings was demonstrated by P18 and P26. Given that P18 was reported to have achieved similar everyday communication with either CI, increased ratings would be expected. Although P26 did not achieve this CI-2 alone milestone, reports indicated early achievement of milestones such as superior performance with, and preference for, bilateral implants. The more modest increases in ratings for P42 are somewhat at odds with this participant's achievement of the milestone of similar communication with either CI.

When considering the data from the anecdotal reports and the SSQ, the potential for bias in favour of bilateral implants must be acknowledged. The participants and their families invested a significant amount of time and emotional energy in the process of obtaining a second implant, and hoped that additional benefit would be gained. Some families expected additional benefit, though others stated they had no such expectation but were following through with any option available to their child. It is not possible to measure the effect of any such bias on the results,

only to consider the data from the anecdotal reports and the SSQ in tandem with the objectively measured results.

Similarly to the reported achievement of milestones, the speech perception results indicated that CI-1 generally remained the superior implant, irrespective of the noise source location. The exception was P18, who demonstrated no significant difference between the implants with ipsilateral noise; the power of the test may have been a factor in the failure to detect a difference. Although similar communication in daily life with either implant was reported for P18 and P42, the speech perception results indicated superior performance with CI-1, though the difference was minimal for P18. Participant 28's slower than average achievement of milestones is reflected in the CI-2 alone scores, which were particularly poor relative to CI-1 alone scores. In contrast, P12 did not demonstrate slower achievement of milestones, despite CI-2 alone listening skills that were too poor to complete the test.

With contralateral noise no participant demonstrated superior performance in the BiCI compared with the CI-1 condition. Given that CI-2 was next to the noise source and listening skills with CI-2 were inferior to those with CI-1, no additional benefit was expected when CI-2 was added. These results suggest that binaural unmasking was not occurring; whether unmasking would occur if performance was equivalent between the two implants cannot be determined. With ipsilateral noise CI-2 was shadowed from the noise source. Five of the nine participants demonstrated superior performance when CI-2 was added in the BiCI condition. This result was expected for P18, given the minimal difference in listening skills between CI-1 and CI-2; theory would propose that attention would switch in the BiCI condition to the shadowed CI-2, which had a better signal-to-noise ratio. For the remaining four participants CI-2 was inferior,

suggesting that the participants were not simply relying on the shadowed CI-2, but may have been gaining useful information from both implants. Of the four participants who did not demonstrate superior performance in the bilateral condition, two (P2 and P28) were also those who had demonstrated slower achievement of milestones.

Despite some inconsistencies in results across assessment methods, some clear conclusions can be drawn. All participants adapted to the use of the second implant and gained a benefit from the use of bilateral implants within 12 months. For some participants this was measured objectively on speech perception testing in noise, and for all participants it was perceived subjectively in daily listening situations. There is clear evidence that a second cochlear implant should be considered for all children, even those with a pre-lingual onset of hearing loss. Being older or many years post-CI-1 should not be barriers to candidacy for a second implant.

Obviously there is an as-yet-unknown hearing level below which a contralateral hearing aid would be a more suitable option. In the present participant group, only P42 had a three-frequency-average below 100dB and only P42 was wearing a hearing aid when a second implant was requested. Unfortunately this participant was not available to complete the research assessment battery pre-operatively so that the important comparison of bimodal versus bilateral benefit could not be made. The question of hearing level should be investigated, however the following points are important. Firstly, the pool of potential participants is limited. The majority of children who are cochlear implant candidates have minimal residual hearing in the second ear. The pool is further reduced as children with an implant may reject their hearing aid, even when their residual hearing may be judged “useful”. Secondly, age-related effects such as neural plasticity, unilateral auditory deprivation and device experience will influence the degree of

benefit gained from any device. The level of hearing below which a hearing aid may be the better option may therefore vary as a function of the child's age at the time the device(s) are fitted. Thirdly, it is important to consider performance using the hearing aid versus the second implant alone. Good open-set speech perception ability with the second device affords important benefits, including being able to converse with speakers located on either side, and being able to continue to function in everyday life when the original implant has flat batteries or hardware problems. These benefits were highly valued by the present participants and their families.

Although this study did not aim to determine the factors which influenced the benefit gained from a second implant, the detailed information collected allows some comments to be made. Considering pre-operative hearing thresholds and hearing aid use, the only two participants to achieve the milestone of similar daily communication using either implant alone were the only participants who were consistent, full-time users of a hearing aid prior to receiving CI-2, and had the lowest 3FA of the group (refer to Table 1). Regarding CI-2 alone habilitation, the majority of participants received limited or no habilitation, yet still demonstrated a bilateral benefit.

Regarding the attitude towards and consistency of CI-2 use, the two participants (P2 and P28) who were slower at achieving most milestones had some difficulty establishing consistent bilateral use, and queried the benefit of the second implant in some situations. Whether their attitude and pattern of device use influenced their performance with CI-2 or vice versa cannot be established. In contrast, P24 and P26 recognized and identified the difficulties of operating in the "hearing world" using just one implant, took responsibility for learning to use their CI-2, and expressed a determination to develop their listening skills. Their attitude and persistence may have had a positive impact on the benefit gained.

Collection of data from these and additional participants is continuing. It is important that long-term outcomes are reported as they may differ from early outcomes. It has already been observed that, in the 12 to 24 months following switch-on of CI-2, the issues with consistent device use and attitude reported here for P2 and P28 have resulted in less than full-time CI-2 use and specific situations in which CI-2 is not considered by the participant to be beneficial.

The results of this study indicate that adolescents and young adults up to the age of 19 years may gain additional benefit from a second cochlear implant, even if their hearing loss is congenital and it is more than 16 years since the receipt of their first implant. Families and clinicians should understand the aspects of second-implant candidacy and post-operative use that are unique to this group, in particular, responsibility for the decision to seek a second implant and self-motivation to learn to use the new device may be more relevant.

Declaration of Interest

The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

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