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Perceptual benefit and functional outcomes for children using sequential bilateral cochlear implants

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Abstract

Objective: To evaluate the additional perceptual benefit provided to children through the use of two cochlear implants in comparison to one after 6 to 12 months experience with sequential bilateral implants.

Design: A second cochlear implant was received by 11 children. The principal selection criteria were being aged 5 to 15 years with a bilateral profound hearing loss, and being a consistent user of a first implant with a commitment to use of a second implant. Horizontal localisation was assessed using pink noise bursts presented from a 180^0 8-loudspeaker array. Speech perception was assessed using a four-alternative forced-choice spondee test, with speech presented from in front and adaptive background noise presented from 90^0 to the left or right. Both tests were completed in the first implant alone and bilateral conditions. A questionnaire measured the pre- to post-operative change in the parent's ratings of the child's performance in specific listening situations. Items were related to speech perception, spatial hearing, or quality of hearing. Regular parental reports of device use, attitude and performance were collected. Most subjects were assessed at 6 months post-operatively, with two assessed at 12 months.

Results: The eleven subjects demonstrated a great range of outcomes. For one subject only anecdotal data was collected. Speech perception testing indicated that when noise was presented ipsilateral to the first implant 8 out of 10 subjects showed a benefit in the bilateral condition. None of the 9 subjects tested showed a benefit when noise was contralateral to the first implant. Generally, there was no benefit to localisation in the bilateral condition. For eight subjects, post-operative performance ratings were generally higher than pre-operative ratings, particularly in the spatial hearing section. Anecdotal reports indicated that most subjects had a negative attitude towards, and gained limited experience with, the second implant alone. The subjects developed a range of speech perception skills, from detection to conversation level. Regarding the use of bilateral implants, attitudes were more positive and device use was consistent for eight subjects, and six parents reported some evidence of improved performance in daily life

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Conclusions: Children over 4 years may gain significant additional benefit from a second implant, including improved speech perception in some noise contexts and functional advantages in daily life. There is, however, no evidence to suggest that binaural listening skills, including localisation, will develop, at least in the first 6 months. Furthermore, some children who may be committed users of a first implant, cannot adapt to, or benefit from, a second implant. Although the factors influencing benefit cannot be clearly identified, limited pre-operative auditory experience with the second ear, a delay of years between implants, relatively advanced age, and lack of second-implant-alone experience do not preclude benefit. Continued evaluation of these and additional subjects will clarify the factors that do contribute to benefit. Such information will be vital in helping families of implanted children to make an informed decision regarding a second implant.

Keywords: bilateral cochlear implants, children, localisation, speech perception

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Introduction

Worldwide, tens of thousands of children with a profound hearing impairment now access sound through a cochlear implant. Recently, there has been increasing interest in the potential for children to gain additional benefit from bilateral cochlear implants. This interest stems, in part, from research with hearing aid users and normally-hearing subjects, which has demonstrated that listening with two ears is clearly superior to listening with one (Dillon, 2001). In addition, studies have also demonstrated benefits from listening with two ears for children using an implant plus a hearing aid (Ching, Psarros, Hill et al., 2001; Dettman, D'Costa, Dowell et al., 2004), and for adults using bilateral implants (see, for example, Müller, Schön, & Helms, 2002; Tyler et al., 2002; van Hoesel & Tyler, 2003; van Hoesel, 2004).

Although it is clear that adults may benefit from a second implant, significant differences between the populations make it imperative that the benefit to children is directly assessed. Gaining maximum advantage from listening with two ears requires good listening skills with each ear and binaural listening skills. True binaural listening requires the brain to combine and/or compare the information arriving at each ear and to utilise such cues as interaural timing and level differences (see, for example, Bronkhorst & Plomp, 1989; Wightman & Kistler, 1992; Dillon, 2001). The majority of adults will have experienced many years of near-normal bilateral sound input, developing listening skills with each ear as well as binaural listening skills. In contrast, the limited sound input resulting from a congenital or early onset hearing loss will negatively affect the development of the auditory system. Even with one implant, a degree of auditory deprivation will continue, with limited or no sound input on the non-implanted side, and limited or no binaural sound input, so that the developing auditory system may be further compromised. It is therefore unknown to what degree children will be able to access all of the "two-ear advantages" even if provided with two implants.

For people with normal hearing, the practical advantages of listening with two ears include improved localization, increased audibility, and improved speech perception, particularly in noise. Localization is the skill of locating the source of a sound in the

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environment, and is generally poor with just one ear. Even with sound input to two ears, localization may still be poor, as binaural listening skills are required to evaluate the relevant interaural timing and level differences. Localization is important for convenience (e.g. knowing where someone is calling from), for safety (e.g. in traffic), and for communication in groups (to quickly direct attention to the speaker). The ability to localize also helps the listener to spatially organise the auditory environment, thus making it easier to attend to a specific signal whilst ignoring others, and increasing awareness of changes in the auditory environment.

The second advantage of listening with two ears is binaural loudness summation, which occurs when a perceived increase in loudness results from combining the signals arriving at the two ears. The third advantage, speech perception improvement, is greatest in background noise due mainly to binaural unmasking and the headshadow effect. Binaural unmasking occurs when the noise and speech sources are separated, so that different signals arrive at each ear. The brain compares the two signals, and uses the waveform of the noise at the ear with the poorer signal-to-noise ratio to partially cancel out the noise received at the contralateral ear (Moore, 1989). The headshadow effect is also beneficial when noise and speech are separated. Advantage is taken of the physical barrier of the head to "shadow" one ear from the noise source to improve the signal-to-noise ratio. Localization, binaural loudness summation, and binaural unmasking require true binaural hearing, whilst the headshadow effect is available where adequate listening skills exist for each ear.

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There are also qualitative benefits from listening with two ears. Hearing aid users have described sound as fuller, more spacious and clearer (Erdman & Sedge, 1981; Balfour & Hawkins, 1992). Adult bilateral implant users have reported that sounds are more natural, clearer, richer, and fuller (Müller et al., 2002) or fuller and of better quality (van Hoesel, Reference Note 4).

Some initial research has evaluated the use of bilateral cochlear implants by children, with generally positive results being reported on objective and/or subjective criteria (Vermeire, Brokx, Van de Heyning et al., 2003; Kühn-Inacker, Shehata-Dieler, Müller et al., 2004; Litovsky, Johnstone, Parkinson et al., 2004; Peters, Litovsky, Lake et al., 2004;

Baumgartner, Jappel, Arnolder et al., Reference Note 1; Litovsky, Johnstone, Godar et al., 2006). Given the great variation in performance amongst implanted children, the small and/or heterogeneous subject groups, and the minimal amount of objective data reported so far, there is a clear need for continued research to evaluate bilateral implant use by children.

The aim of the present study is to evaluate sequential bilateral implant use by children by determining whether greater perceptual benefits are provided through use of two cochlear implants in comparison to one after limited experience (6 months for most subjects, 12 months for two subjects). A longer-term aim of the ongoing research project is to relate the degree of benefit to factors such as device experience, age, pre-operative hearing aid use, and time between the two implants. Outcomes from this process will allow evidence-based recommendations to be made to the families of future individual candidates.

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Materials & Methods

Subjects

Recruiting potential candidates: The research project was publicized to families and teachers of 5 to 15 year old unilaterally implanted children attending the Royal Victorian Eye and Ear Hospital/University of Melbourne Cochlear Implant Clinic (Implant Clinic). Children were accepted as potential candidates if they were full-time implant users with no evidence or reports of compromised functioning of that implant (e.g. <15 functioning electrodes or significantly poorer than expected speech perception skills). There also had to have been no reports of cognitive difficulties or developmental delay from teachers or other professionals. The audiological criterion was a profound hearing loss in the unimplanted ear (i.e. pure-tone average $\geq$ 90dB SPL. 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 criterion was that the child, as well as the family, was considered to be committed to the use of a second implant and likely to be able and willing to complete the research assessment battery. In the time period September 2003 to September 2005, 14 families requested that their children be

considered for a second implant. Three children did not fulfill the selection criteria, having respectively significant bilateral Mondini malformation with limited active electrodes in the first implant, developmental delay and cerebral palsy, and cognitive delay and learning difficulties. The remaining 11 children went on to complete the assessments to confirm their candidacy, and subsequently received a second implant. All children bilaterally implanted at the Melbourne Clinic in this time period (i.e., the eleven discussed here plus three other children aged less than 4 years) participated in the research project.

Confirming audiological candidacy: As per the Implant Clinic's protocol for a first implant, the confirmation of audiological candidacy was based on the predicted post-operative improvement in perceptual ability of the non-implanted ear alone. The standard protocol called for unaided and aided hearing threshold testing, and aided speech perception testing. Where a recent or successful aid fitting had not been made, hearing aid fitting or refitting and a 3-month aid trial were to occur prior to aided testing. Aid fitting was conducted by the responsible Government body, Australia Hearing, according to their protocols. Audiological evaluation established all 11 children as clear implant candidates, with no borderline cases.

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Subject demographics: Based on the process described above, 11 children aged 4 years or older received a second cochlear implant in the period September 2003 to September 2005. Hearing loss and hearing aid use details are presented in Table 1. Subject numbering includes younger children not discussed here and is therefore not consecutive. All subjects had a bilateral profound loss. A congenital onset of hearing loss was assumed for the majority of subjects, whilst S7 and S14 had meningitis at 11 months and 21 months respectively. Though all were fitted with hearing aids prior to receiving their first implant, the child removing the aids or recurrent otitis media resulted in six children not using their aids consistently. None of the children were voluntary, consistent users of a contralateral hearing aid with their first implant. At the time they presented for pre-operative assessment for a second implant, three subjects were already fitted with an aid, though S2 and S6 only wore the aid sometimes and S5 wore it only at school. A further three subjects (S4, S9 and S12) were re-fitted with an aid to complete a 3-month hearing

Table 1: Hearing loss and hearing aid demographics for the eleven subjects who received a second implant.

Subj	Etiology/Age at onset profound loss	3FA[1] CI-1 ear	Consistent aid use pre CI-1	3FA CI-2 ear	Period & consistency of contralateral aid use pre CI-2
S1	unkn/congenital?[2]	112[3]	no-child removed	115[4]	not fitted
S2	unkn/congenital?[5]	115[3]	no–child removed	103[4]	12mo/occasional
S3	KIDD synd/congen	NR[6]	no-recurrent OM	115 at 500Hz	not fitted
S4	unkn/congenital?[7]	102[8]	yes-bilateral	108[4]	3mo/full time
S5	unkn/congenital?[9]	115[3]	no–recurrent OM	118[4]	12mo/school only
S6	Duane's Syndr/16mo?[10]	118[3]	yes-bilateral	118[4]	3mo/occasional
S7	meningitis/11mo	NR[3]	no-child removed	115[8]	not fitted
S8	unkn/congenital	117[11]	yes-R only (L atresia)	NR[12]	not fitted
S9	unkn/congenital	120[3]	yes-bilateral	110[8]	3mo/kinder' only
S12	unkn/congenital?[13]	110[3]	yes-bilateral	115 at 500Hz[4]	3mo/full time
S14	meningitis/1yr9mo	NR	no-child removed	NR[8]	not fitted

[1] Average of unaided thresholds at 500Hz, 1kHz, and 2kHz. [2] Assumed congenital; diagnosed at 11 mo. [3] Obtained via Steady State Evoked Response (SSEP) testing. [4] Obtained via standard audiometry. [5] Assumed congenital; loss suspected at 6 mo, diagnosed at 9 mo. [6] Obtained via Auditory Brainstem Response (ABR) testing using click stimulus at 100dBHL. [7] Assumed congenital; loss suspected at 12 mo, diagnosed at 18 mo. [8] Obtained via play audiometry. [9] Assumed congenital; diagnosed at 8 mo. [10] Progressive, profound loss diagnosed at 16 mo. [11] Test procedure not specified. [12] Bone conduction thresholds obtained via play audiometry. [13] Assumed congenital; diagnosed at 11 mo.

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aid trial prior to pre-operative assessment for a second implant. The subjects wore the aid only as a requirement of the research, and S9 refused to wear the aid outside kindergarten. The remaining five subjects did not wear a contralateral aid. This was on the basis of contra-indicatory medical advice for S3 and canal atresia for S8. For S1, S7 and S14, the families did not wish their child to participate in an aid trial. Given that these three subjects removed the aids prior to their first implant, and unaided hearing testing demonstrated no auditory response above 500Hz at the audiometer limits, they were accepted as candidates for a second implant despite their failure to complete an aid trial.

Age at implantation, the basis of ear choice, and indicators of performance with the first implant are presented in Table 2. At the time of the first implant, the first implanted ear was considered slightly poorer in terms of hearing for S4 and S5, and slightly better in terms of the potential for a full electrode array insertion for S6, S7 and S8. For the remaining six children the ears were symmetrical. The mean age at first implant was 2 yr 1 mo (range: 1yr3mo–4yr3mo), and the mean at second implant was 8 yr 4 mo (range: 3yr11mo–14yr4mo). The mean time between implants was 6 yr 2 mo (range: 2yr8mo–10yr2mo). Details of the implant types, speech processors, and speech processing schemes used are provided in Table 3. All subjects used oral/aural communication, though S1 and S6 also used AusLan, to a minor and significant degree respectively.

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Post-operative Clinical Management

All subjects received standard post-operative audiological and medical care from the Implant Clinic. This included weekly mapping sessions in month one, fortnightly mapping sessions in month two, and then map adjustments as required. The second implant (CI-2) was mapped using a standard unilateral approach, and the processor was programmed with the same speech processing strategy as that used for the first implant (CI-1). Subject 1 and S2 required the application of a C-level modifier to decrease the loudness when listening in the bilateral condition; these subjects were provided with unilateral and bilateral maps for use as appropriate. It was not feasible to attempt to

Table 2: Age at implantation, first implanted ear, basis of ear choice, indicators of performance and age at testing with the first implant, and time between implants for the eleven subjects who received a second implant.

Subj	Age at CI-1 (yr;mo)	CI-1 ear /Basis of choice	Language/vocab age equivalent score[2]	Age at testing (yr;mo)	BKB[1] sentence score (CI-1 alone)	Age at CI-2 (yr;mo)	Time b/w implants (yr;mo)
S1	2;6	R/none	PPVT[3]: 3;10	7;6	70%[4]	9;8	7;1
S2	1;9	R/R-handed	PPVT: 8;6	8;6	96%[5]	11;3	9;6
S3	1;3	L/none	Reynell[6]: 3;3-3;6	3;5	78%[5]	3;11	2;8
S4	4;3	R/slightly poorer hearing	PPVT: 13;2	14;2	94%[3]	14;4	10;2
S5	2;7	R/slightly poorer hearing	PPVT: 5;8	7;9	94%[3]	8;3	5;8
S6	2;2	L/less malformed	PPVT: 3;7	7;1	68%[5]	9;4	7;3
S7	1;5	R/slightly clearer CT scan	CELF Pre[7]: 2;10	6;4	55%[5]	6;9	5;4
S8	1;6	R/non-atresic ear	PPVT: 3;10	5;2	88%[5]	5;10	3;6
S9	1;8	R/none	PPVT: 2;7	4;10	51%[5]	5;11	4;3
S12	1;9	L/slightly poorer hearing	PPVT: 4;8	6;10	86%[3]	10;2	8;4
S14	2;0	L/none	PPVT: 5;1	6;1	83%(CNCs[8])	6;3	4;3

[1] Bamford-Kowal-Bench open-set sentences; scored by % of key words correct. [2] Age at which a child with normal hearing would be expected to obtain the same score. [3] Peabody Picture Vocabulary Test. [4] Recorded presentation in quiet. [5] Live-voice presentation in quiet. [6] Reynell Developmental Language Scales. [7] Clinical Evaluation of Language Fundamentals - Preschool. [8] Phoneme score on the Consonant-Nucleus-Consonant (CNC) Word test.

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Table 3: Cochlear implant types, speech processors and speech processing scheme for the eleven subjects at the time the results reported here were collected.

	Implant type		Speech processor		Processing scheme
Subject	CI-1	CI-2	CI-1	CI-2	CI-1 & CI-2
S1	CI22[1]	CI24RCS[2]	ESPrit22[3]	ESPrit3G[4]	Speak[5]
S2	CI22	CI24RCA[6]	Spectra	ESPrit3G	Speak
S3	CI24RCS	CI24RCA	ESPrit3G	Sprint	ACE[7]
S4	CI22	CI24RCA	ESPrit22	ESPrit3G	Speak
S5	CI24M[8]	CI24RCA	ESPrit3G	ESPrit3G	Speak
S6	CI22	CI24RST	ESPrit22	ESPrit3G	Speak
S7	CI24M	CI24RCS	ESPrit3G	ESPrit3G	ACE
S8	CI24M	CI24RCA	ESPrit3G	ESPrit3G	ACE+ADRO[9]
S9	CI24M	CI24RCA	ESPrit3G	ESPrit3G	ACE
S12	CI22	CI24RCA	ESPrit3G	ESPrit3G	Speak
S14	CI24RCS	CI24RCA	ESPrit3G	Freedom	ACE

[1] CI22: straight array implant with 22-electrodes. [2] CI24RCS: curved array implant with 22-electrodes plus 2 extra-cochlear electrodes. [3] ESPrit22: ear-level speech processor for use with CI22 implant. [4] ESPrit3G: ear-level speech processor capable of higher-rate processing. [5] Speak: peak-picking filterbank speech processing strategy. [6] CI24RCA: soft-tip curved array implant with 22-electrodes plus 2 extra-cochlear electrodes. [7] ACE (Advanced Combination Encoder): filterbank strategy with variable high rate presentation. [8] CI24M: straight array implant with 22 electrodes plus 2 extra-cochlear electrodes. [9] ACE with ADRO: ACE combined with ADRO (Adaptive Dynamic Range Optimisation), an algorithm which works with the processing strategy to improve detection of soft sounds and speech, whilst maintaining a comfortable presentation level for loud sounds.

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"match" the implants or to trial alternative combinations of speech processing schemes because of the time limitations of the families and the clinicians, and, particularly, because of the limited ability of the children to provide accurate feedback on differences in sound, especially for a recent implant. The implant clinicians did not provide habilitation sessions to the subjects, with the exception of S3 who attended six 1-hour sessions that focused on listening with CI-2 alone.

Bilateral Implant Assessment Battery

The research assessment battery consisted of perceptual tests of speech perception in noise and localisation, and a questionnaire. Anecdotal reports of device use and daily performance were also collected in monthly phone interviews with the parents.

Speech Perception

Speech perception was assessed using AdSpon (Adaptive Spondee Discrimination Test); a four-alternative, forced-choice spondee discrimination test presented in background noise. The test is fully software controlled, with the auditory stimuli presented pictorially on a touchscreen alongside three foils. 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. Four test lists, each consisting of 10 sets of four spondees, were created. The spondees were pseudo-randomly selected with the following criteria also applied: each spondee occurred twice in each list, no spondee occurred in two consecutive sets, and the pairs hairbrush/toothbrush, eyebrow/eyelash, and blackbird/blackboard did not occur in one set. The lists were continuously presented in sequential order until the criteria for ceasing testing (outlined below) were fulfilled. The starting list was selected randomly. For each presentation of a set of four spondees, the test stimulus was randomly selected.

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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, James, & Martin, 2000).

Testing was conducted in a low-reverberation sound-proof booth. The subject was seated 1.15m from loudspeakers positioned at head height at 0^0 and at 90^0 to the right and left. Test sessions consisted of training and testing phases. In the training phase, two auditory presentations of each spondee were made in quiet with the relevant picture displayed. The tester confirmed that the subject could identify each picture. This initial training step was omitted if a second test session occurred on the same day. Next a practice test list of at least 10 stimuli was presented in noise. In the testing phase, sets of four pictures were presented with accompanying auditory presentation of the test stimuli until the criterion of the adaptive noise procedure was met. No feedback was provided and guessing was required. In each test phase subjects were assessed in two device conditions (CI-1 & BiCI (bilateral cochlear implant)) and two noise conditions (noise from 90^0 to the right or left). Speech was always presented from 0^0. When the device condition was changed, the subject was engaged in five minutes of conversation to allow adaptation to the new condition. Device and noise conditions were alternated across sessions and subjects.

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Localisation

Localisation testing was similar to that described for adults using bilateral implants (van Hoesel & Tyler, 2003; van Hoesel, 2004). The stimulus was a series of 4 pink noise bursts, each of 170msec duration with 10msec rise and fall times, with an inter-burst duration of 50msec. The presentation level was 66dBA, with a jitter of 8dB to limit the use of loudness cues. Testing was conducted in one of two low-reverberation sound-proof booths. The subject sat facing a 180^0 array of eight Bose 151 Environmental or Tannoy Reveal loudspeakers spaced 25.7^0 apart. The loudspeakers were positioned at

head height at a distance of 1.15m and were numbered one (at 90^0 on the left) through eight (at 90^0 on the right). Each test block consisted of training and testing phases. In the training phase, two "runs" were conducted in which a stimulus was presented from each loudspeaker in turn. In the testing phase, the presenting loudspeaker was randomly selected, with one stimulus presented from each loudspeaker before the next round of presentations. Nine presentations were made from each loudspeaker, with the first presentation from each loudspeaker discarded. No head movement was allowed, feedback was not provided, and guessing was required. In each of the two test sessions, subjects completed one test block in each of the CI-1 and BiCI conditions. When the device condition was changed, the subject was engaged in five minutes of conversation to allow adaptation to the new condition. Conditions were alternated across test sessions and subjects.

Questionnaire

The parents' subjective perception of the children's performance was evaluated via a questionnaire. The questionnaire was based on The Speech, Spatial, and Qualities of Hearing Scale (SSQ) designed for adults using bilateral hearing devices (Gatehouse & Noble, 2004; Noble & Gatehouse 2004). The first author modified the adult version to produce "The Speech, Spatial and Qualities of Hearing Scale for Parents of Children with Impaired Hearing" (and "The SSQ for Children with Impaired Hearing" for children aged 10+ years, which is not included here). Although appropriate tools exist for clinical purposes (such as P.E.A.C.H. and T.E.A.C.H. [1], and LifeUK IHR (version 2)[2]) there is no pediatric questionnaire that examines the relevant domains in sufficient detail for the present purpose. The three sections of the Scale examined speech perception (in quiet, on the telephone, in groups and/or in noisy or reverberant environments), spatial hearing (the location and direction of sounds), and quality of hearing (segregating and identifying sounds, the naturalness of sounds, and listening effort). A full list of questions is attached as Appendix A. For each type of situation described, the parent indicated their rating of the child's listening skill on a scale of 1 to 10 (refer to Appendix A). To maximize the

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[1] developed by T. Ching and colleagues; available from www.nal.gov.au
[2] adapted by D. Canning from the work of K. Anderson and J. Smaldino; available from www.hear2learn.com

accuracy of the ratings, a minimum of one week of observation was completed by the parent prior to the completion of each section in a face-to-face or phone interview. The SSQ was administered in the 6 weeks prior to receipt of the second implant and again at the post-operative assessment point, without reference to the pre-operative results.

Results

As S6 did not attend any assessment sessions, anecdotal reports were the only data collected. Of the remaining 10 subjects, a full set of results was collected for eight subjects. Subject 3 and S7 completed a partial objective assessment as, due to age and concentration span respectively; they required more time to complete the tests and were not available for additional sessions. The 10 subjects' age and amount of experience with each device at the time the post-operative questionnaire and perceptual tests were administered are presented in Table 4. Eight subjects had approximately 6 months experience with their second implant, whilst S3 and S7 had 13 months experience.

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

Subject	Age (yr;mo)	Time since start-up (yr;mo) *CI-1*	*CI-2*
S1	10;4	7;9	0;8
S2	11;9	10;0	0;6
S3	5;0	3;9	1;1
S4	15;0	10;9	0;7
S5	8;10	6;3	0;7
S6	9;11	7;9	0;6
S7	7;11	6;6	1;1
S8	6;5	4;10	0;6
S9	6;6	4;10	0;6
S12	10;8	8;10	0;6
S14	6;10	4;10	0;7

Speech Perception: AdSpon

Nine subjects were tested in the ipsilateral and contralateral noise condition, whilst S3 was tested in the ipsilateral condition only. The number of test lists completed varied as a result of the limited availability of the subjects.

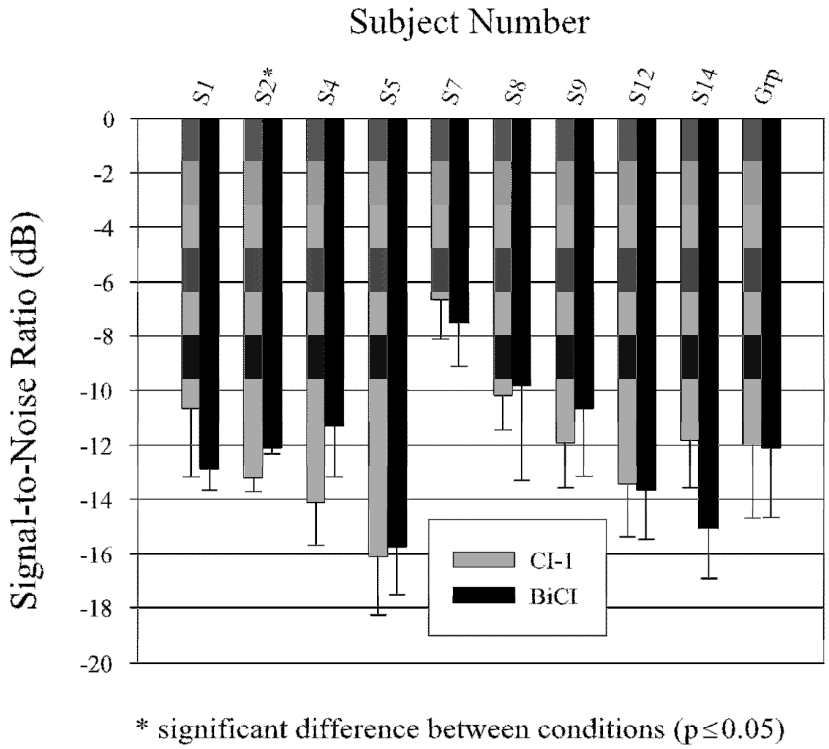


Figure 1: Mean (S1, S2, S4: n=3; S5, S7, S8, S9, S12, S14: n=4) SNR at which each of the nine subjects and the group achieved the criterion level of performance on the AdSpon test in the CI-1 and BICI conditions when noise was presented contralateral to CI-1. Error bars represent +/- 1 standard deviation.

Figure 1 presents the mean AdSpon results for nine subjects and for the group when the speech was presented from in front and the noise was presented contralateral to CI-1. One-tailed, paired t-tests indicated no difference in the mean SNR between the CI-1 and BiCI conditions for S1, S4, ($t(2) \leq 1.93$, $p \geq 0.1$), S5, S7, S8, S9, S12 or S14 ($t(3) \leq 2.24$, $p \geq 0.06$) or for the group ($t(7) = 0.34$, $p = 0.74$). For S2, performance was superior (i.e., a lower SNR) in the CI-1 condition (difference = 1.1dB, $t(2) = 5.02$, $p = 0.02$).

Figure 2 presents the mean AdSpon results for 10 subjects and the group when the speech was presented from in front and the noise was presented ipsilateral to CI-1. One-tailed,

paired t-tests indicated superior performance (i.e., a lower SNR) in the BiCI as compared with the CI-1 condition for S2, S4 (difference $\geq$1.3dB, $t(2) \geq 2.8$, $p \leq 0.05$), S3, S5, S7, S8, S12, S14 (difference $\geq$2.5dB, $t(3) \geq 2.52$, $p \leq 0.04$) and for the group (difference = 3.1dB, $t(9) = 5.24$, $p < 0.001$). There was no significant difference between the conditions for S1 ($t(2) = 0.00$, $p = 0.5$) or S9 ($t(3) = 1.03$, $p = 0.19$).

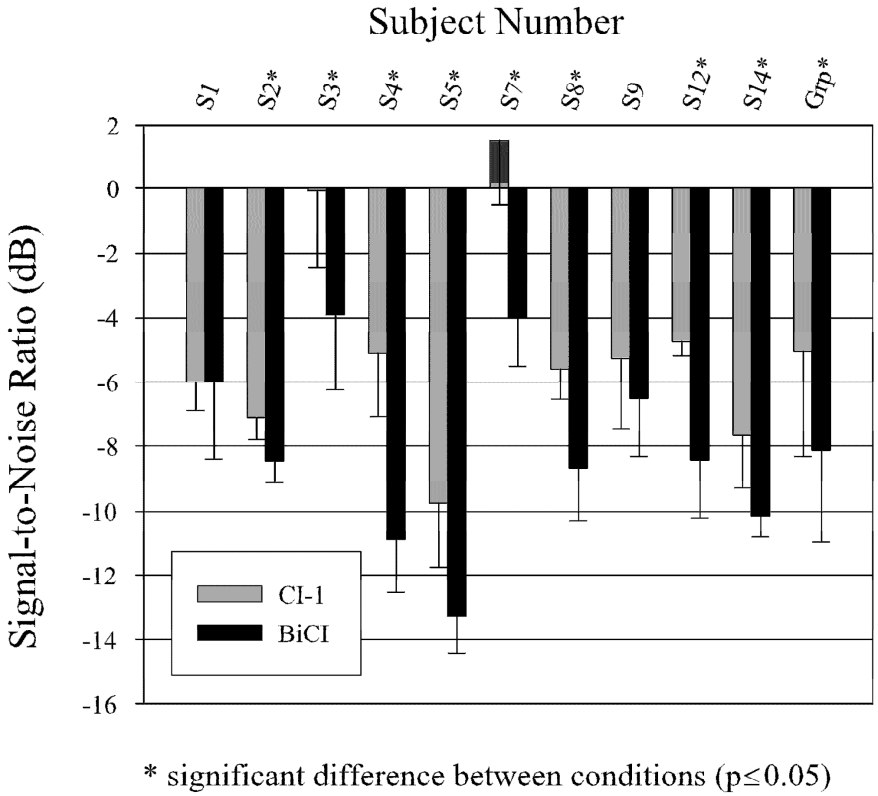


Figure 2: Mean (S1, S2, S4: n=3; S3, S5, S7, S8, S9, S12, S14: n=4) SNR at which each of the ten subjects and the group achieved the criterion level of performance on the AdSpon test in the CI-1 and BICI conditions when noise was presented ipsilateral to CI-1. Error bars represent +/- 1 standard deviation.

Localization

Eight subjects completed localization testing, with the exclusion of S3 and S7. Two test blocks were completed, for a total of 16 stimuli from each loudspeaker in each condition (S4 received just 12 stimuli from each loudspeaker in the BiCI condition due to a software error). Figure 3 presents the mean (n=128; n=96 for S4 in BiCI condition) root-mean-square (RMS) error in degrees in the CI-1 and BiCI conditions for each subject and

for the group. The RMS error approximates the average angle of error from the presenting loudspeaker. The mean is collapsed across presentations and loudspeakers. For each subject, the magnitude of the error made in response to each stimulus was ranked (to account for the non-normal distribution), and a two-way Analysis of Variance was conducted with condition and loudspeaker as factors. No significant condition effect was found for S1, S2, S5, S8, S12 or S14 ($F(1,255) \leq 0.92$, $p \geq 0.34$) or S4 ($F(1,223) = 0.51$, $p = 0.47$). A significant condition effect was found for S9 ($F(1,255) = 4.5$, $p = 0.035$), with the Holm-Sidak pairwise multiple comparison procedure indicating superior performance in the BiCI condition. No significant condition by loudspeaker interaction effect was found for S1, S5, S8, S12 or S14 ($F(7,255) \leq 1.4$, $p \geq 0.20$) or S4: $F(7,223) = 1.6$, $p = 0.13$). A significant interaction effect was found for S5 and S9 ($F(7,255) \geq 2.1$, $p \leq 0.044$). The Holm-Sidak pairwise multiple comparison procedure indicated that performance was significantly superior in the BiCI condition for S5 on loudspeaker 3 and for S9 on loudspeakers 1 and 6.

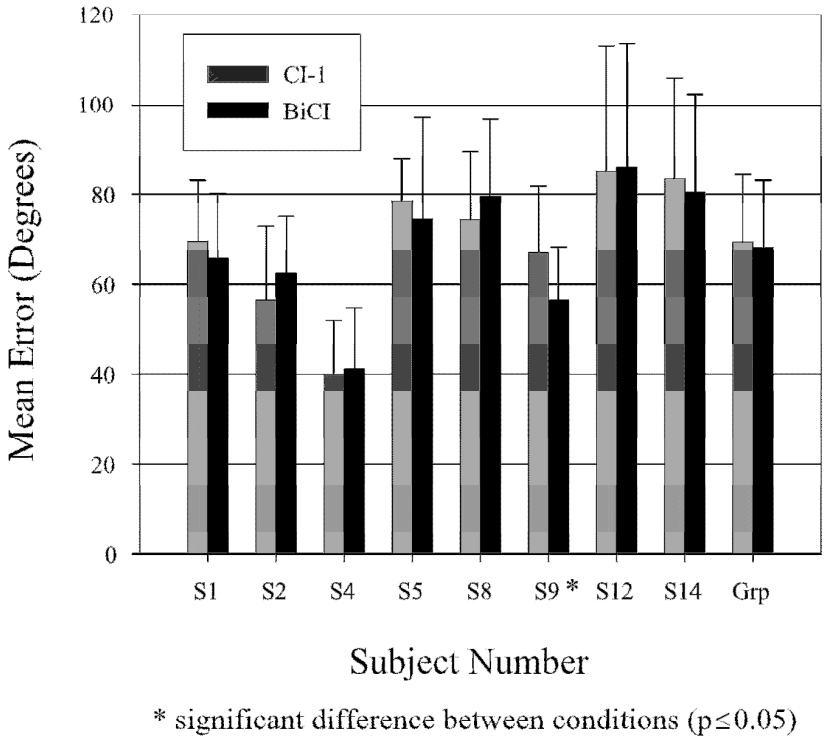


Figure 3: Mean RMS error in degrees in the CI-1 and BiCI conditions for each of the eight subjects and the group. Results are collapsed across 8 loudspeakers and 16

presentations (n=128; n=96 for S4 in BiCI condition). Error bars represent +/- 1 standard deviation.

Questionnaire: The Speech, Spatial, and Qualities of Hearing Scale

The rating on each item of the SSQ provided at the 6-month post-operative assessment point (or 12 months post-operatively for S3 and S7) was compared with that provided pre-operatively (in the month prior to the second implant operation). Pre-operative ratings were in the CI-1 alone condition for most subjects, and the cochlear implant plus hearing aid condition for S4 and S12, who were the only full-time aid users. The speech perception items examining conversation in ideal listening conditions (item 2) and on the telephone (item 9) were not included in the analysis because bilateral sound input was not expected to improve performance in these situations.

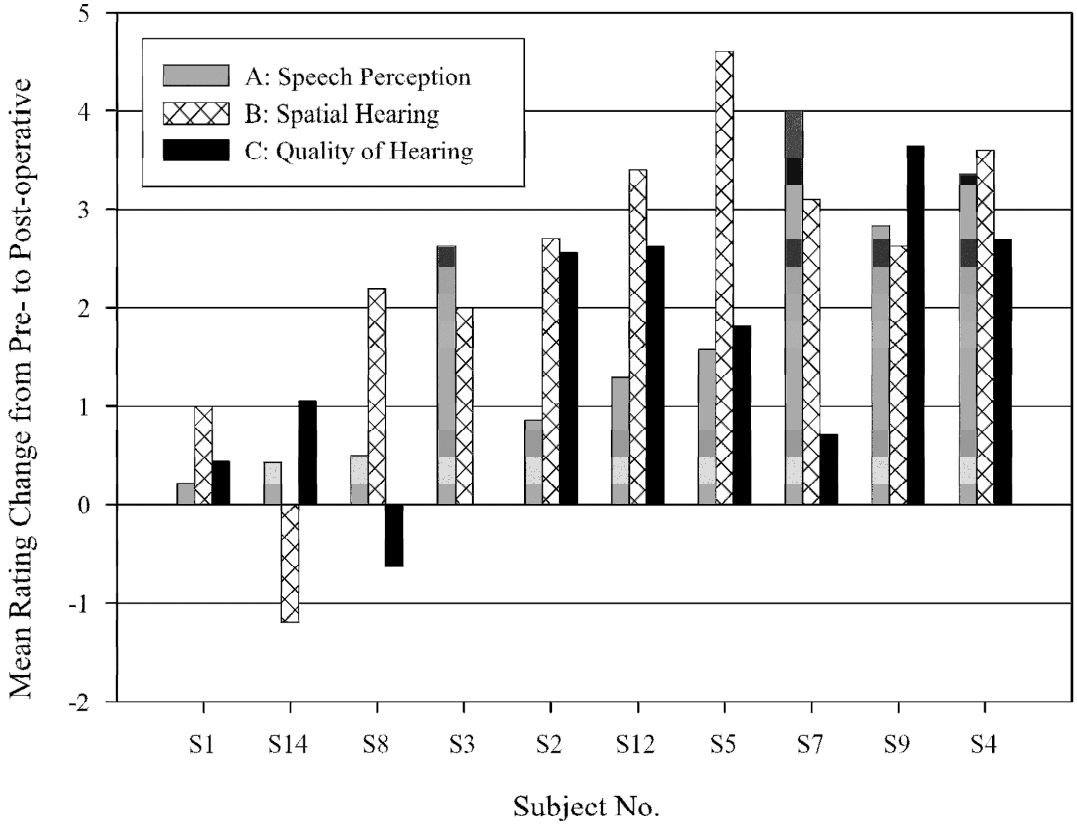


Figure 4: Mean change in performance rating for each subject for each section of the SSQ Scale. Change is the difference in ratings provided by the parent post-operatively versus pre-operatively. Ratings are collapsed across items within Section A: Speech

Perception (n=7; S3 & S9: n=6), Section B: Spatial Hearing (n=5; S1, S3 & S9: n=4), and Section C: Quality of Hearing (n=8; S3, S7, & S9: n=7).

Figure 4 presents the mean pre-operative to post-operative rating change for each subject in each of Section A: Speech Perception (n=7), Section B: Spatial Hearing (n=5), and Section C: Quality of Hearing (n=8). Where the parent was unable to provide a rating the item was excluded from the subject's results. This occurred for one item in each section for S3 and S9, and for one item in the spatial hearing and quality of hearing sections respectively for S1 and S7. In Figure 4, subjects are loosely ordered in increasing degree of rating change. For S1 and S14 there was no pattern of improved ratings in the pre-operative to post-operative comparison. For the remaining 8 subjects there was at least some increase in post-operative ratings. Ceiling effects limited the potential increase in ratings for S8 and S3, particularly for the quality of hearing section where the mean pre-operative rating was 7.9 and 9.3 respectively. Where ceiling effects did not apply ratings increased. For example, the mean increase for the spatial hearing section was around 2 points for each subject, and the mean increase on items relating to "group conversation without visual cues" was 5 and 2.3 respectively for S3 and S8. For the remaining six subjects (S2, S4, S5, S7, S9, and S12), ratings increased post-operatively as compared with pre-operatively on nearly all items. In the spatial hearing section there was a mean increase of more than 2.5 points for all six subjects, with a maximum of 4.6 points for S5. In the speech perception section there was a mean increase of more than 2.5 points for S4, S7 and S9, with a maximum of 4.0 points for S7. In the quality of hearing section there was a mean increase of more than 2.5 points for S2, S4, S9 and S12, with a maximum of 3.6 points for S9.

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Anecdotal Reports of Device Use and Daily Performance

Table 5 summarises the following factors relevant to use of the second implant alone in the first 6 months following the start-up of the device: the child's attitude, period of daily use, direct rehabilitation received, and listening skill level achieved. Also summarised are the following factors relevant to use of bilateral implants in the same time period: the child's attitude, consistency of use, and reported improvement in listening skills compared with CI-1 alone. The information is based on reports from parents (mostly) and teachers.

With regard to the use of CI-2 alone, most of the children had a negative attitude, or a neutral attitude at best (S4, S6 and S12). Only S8 had a neutral-to-positive attitude, and only this subject gained daily experience using CI-2 alone for an extended period. Ongoing, though short, rehabilitation sessions targeting CI-2 alone use were received by S2, S5, S12 and, less frequently, S8. The subjects demonstrated a range of perceptual skills when using their second implant alone. Superior skills were reported for S4, S7 and S8, who were able to converse without the use of vision, and for S3 who could converse if limited visual information was provided. The poorest skills were reported for S1, S6 and S14, who could only discriminate syllable patterns and/or detect sound. As S9 refused to use CI-2 alone it was not possible to determine the skill level achieved.

In contrast to their attitude towards the second implant alone, the majority of subjects had a positive attitude towards the use of bilateral implants and used the two devices consistently. The exceptions were S1, S6 and S14. Subject 1 and S6 were reportedly often switched the second implant off. Subject 14 had around 11 weeks of very irregular or non-use due to school holidays (6 weeks) and hardware failures (5 weeks). Regarding

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Table 5: Reported details of functional use of the second implant and bilateral implants in the 6 months following the start-up of the second device for each subject. Reports were made by parents (mostly) and teachers.

	Use of Second Implant Alone				Use of Bilateral Implants		
Subj	*Child's attitude*	*Period of daily use*	*Direct rehab*	*Perceptual skill level*	*Child's attitude*	*Consistent use*	*Reported improvement cf CI-1 alone*
S1	very negative	mth1-2:½hr/day	none	syllable pattern	negative -neutral	no[1]	none
S2	negative	school rehab sessions	20min/ school day	simple sentences	positive	yes	spatial awareness
S3	negative	mth5:45min/day	1hr /f'night (mth4-5)	conversation (with some vision)	positive	yes	localizing speaker general communication
S4	neutral	mth1-2:½hr/day	20min/ f'night	conversation	very positive	yes	communication in grps communication in noise
S5	negative	school rehab sessions	10min/ school day	simple sentences	positive	yes	communication in grps
S6	neutral	mth3:1hr/day	none	detection	negative -neutral	no[2]	none
S7	negative -neutral	none	none	conversation	very positive	yes	no opportunity to compare
S8	neutral-positive	mth1-2:½-1hr/day mth3-6:1-3hrs/day plus whole days	2 x 20min/ week	conversation	positive	yes	localizing sounds perception at a distance sound awareness
S9	negative	none	none	CI-2 never used alone	positive	yes	none
S12	neutral	mth2:½hr/day +school rehab sessions	10min/ school day	closed-set words	positive	yes	localizing speaker sound awareness
S14	negative	none	none	detection	neutral-positive	no[3]	none

[1] Both implants worn but teacher reports CI-2 often turned off (exact frequency unknown). [2] Worn consistently in mth 1-3; subsequently teacher reports CI-2 often turned off (exact frequency unknown). [3] Worn consistently in mth 1-2; little or no use in mth 3-6 due to speech processor faults and use of CI-1 alone in school holidays.

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the improvement in performance using BiCIs implants versus CI-1 alone, reports were clear that no improvement was evident for S1, S6, S9 and S14. Other parents (particularly of S7) commented that, because CI-1 was rarely or never worn alone, it was difficult to identify improvements and/or difficult to be certain that improvements were related to the use of two implants rather than normal development. Nevertheless, improvements were reported that related to spatial hearing and sound awareness (S2, S3, S8, and S12) and speech understanding in background noise and/or groups (S4 and S5).

Discussion

Outcomes varied widely amongst these 11 children who received a second implant over the age of 4 years. The following discussion of results focuses on the 10 subjects who were assessed. Subject 6 cannot be included as no assessment sessions were attended; however, it should be noted that subjective parent and audiologist reports indicated a poor outcome, with CI-2 worn inconsistently and no additional benefit provided over CI-1 alone.

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Objective speech perception testing indicated that the benefit gained depended on the noise condition. None of the nine subjects tested gained additional benefit in the BiCI condition when noise was presented contralateral to CI-1. Reports from parents, teachers and clinicians indicated that CI-1 remained the superior implant for all subjects, particularly in difficult listening conditions. With the superior CI-1 shadowed from the noise source, CI-2 made no measurable contribution to the perceptual task, suggesting that binaural unmasking was not occurring. One subject (S2) demonstrated a small decrement in performance in the BiCI condition. This result was not consistent with observed performance in daily life and is difficult to explain. In the second condition, with noise presented ipsilateral to CI-1, eight of the ten subjects tested gained additional benefit in the BiCI condition. This was due, at least in part, to the "headshadow effect"; with CI-2 shadowed from the noise source it was able to contribute to the perceptual task. However, the performance asymmetry between the implants, particularly for S2, S5, S12 and S14, would suggest that the subjects were utilizing information from both implants rather than relying only on the information from the shadowed CI-2. The lack of benefit

demonstrated by S1 and S9 suggests that CI-2 was so inferior that it did not contribute useful information despite being shadowed from the noise source.

The present speech perception results cannot be compared with those obtained in most other pediatric studies due to methodological differences, particularly with regard to the speech/noise presentation conditions. The exception is the study of Litovsky, Parkinson et al. (2004), which found a bilateral advantage for two out of three subjects when noise was presented ipsilateral to CI-1, and for no subjects when noise was presented contralateral to CI-1. A similar trend has been demonstrated in adult research, with a consistent and robust bilateral advantage with noise presented ipsilateral to CI-1, and variation across subjects and studies with noise presented contralateral to CI-1 (Müller et al., 2002; van Hoesel, Ramsden, & O'Driscoll, 2002; van Hoesel & Tyler, 2003).

On localisation testing, only S9 demonstrated a significant condition effect. Given the poor map used by this subject, and the fact that there was minimal difference between the conditions in the centre of the array and more towards the ends of the array, the result is likely due to the very significant loudness asymmetry between the ears. For the remaining seven subjects, the localisation results generally indicate no additional benefit in the BiCI condition. Previous research has demonstrated that adults benefit from a second implant when localizing sound in the horizontal plane (van Hoesel et al., 2002; van Hoesel & Tyler, 2003), with results suggesting that they are sensitive to interaural level cues (van Hoesel, Tong, Hollow et al., 1993; van Hoesel & Clark, 1997), but less sensitive to interaural timing cues (van Hoesel & Clark, 1995; Lawson et al., 1998; van Hoesel et al., 2002). The present results are consistent with those reported for a group of three children with 3 to 9 months experience (Litovsky, Parkinson et al., 2004; Peters, Litovsky, Lake et al., 2004). It is possible that the localisation performance of the present subjects may improve with further bilateral experience. Litovsky et al. (2006) measured the minimal audible angle thresholds of 13 sequentially implanted older children with 2 to 23 months bilateral experience. Of the nine children who were able to perform the task with sufficient skill, seven showed a bilateral benefit. In addition, although there was no significant difference in bilateral performance between the more and less experienced subjects, three subjects tested repeatedly demonstrated improved performance in the bilateral condition. Interestingly, all three also demonstrated improved performance in at

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least one unilateral condition. In contrast to the present results, four of Litovsky's subjects demonstrated a bilateral benefit with 8 months or less experience. Although there were testing differences (e.g., Litovsky measured minimal audible angle, used words as stimuli, and provided feedback), it also appears that there is significant inter-subject variation.

Parent questionnaire results indicate that, for eight subjects, parents perceived improved performance in daily life post-operatively. For S9, the SSQ results indicated improved performance in many listening situations, which is inconsistent with the lack of stimulation being provided by the device. This subject entered a transition-to-school program at the time the second implant was received, and this change to a more formal educational setting may have resulted in the significant improvements that were noted by the parents when providing the post-operative ratings. Although areas and degree of improvement varied across subjects, generally the most consistent improvements were shown for spatial hearing and communication in groups without visual cues. These are expected areas of benefit from listening with two ears. The authors acknowledge the potential reliability and validity issues inherent in the use of parent ratings, and the difficulty of separating the effect of treatment from that of normal development with repeat administrations of the scale over time. In considering the reliability and validity of the results, it is informative to note that there was minimal change in parental ratings over time for the situation of a one-on-one conversation in ideal conditions. This item was not included in the analysis of SSQ results as no post-operative improvement in performance was expected. The pre- to post-operative rating change was 0.9 points for seven subjects (with three subjects excluded due to ceiling effects).

Overall, there was reasonable consistency across the objective and subjective results, and the anecdotal information summarized in Table 5. Subject 1, S6, S9 and S14 varied from the pattern of benefit demonstrated by the other subjects assessed. For all four subjects, no benefit was reported for listening in daily life; for S1 and S9 no objective benefit to speech perception was measured; and for S1 and S14 there was no increase in performance ratings. Subject 1, S6 and S14 used CI-2 inconsistently, whilst S9's map is likely to have limited the auditory information provided by the device. The remaining 7 subjects showed benefit on objective and subjective assessments, which was consistent

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with the anecdotal reports of consistent bilateral device use, a neutral-to-positive or better attitude towards two implants and, generally, some additional benefit in daily life.

Due to the size and heterogeneous nature of the group, there is insufficient evidence to identify which factors contributed to the benefit gained from a second implant by individual subjects. Based on clinical experience with this group, the authors speculate that influential factors may include potential auditory capability as indicated by performance with CI-1 alone, consistency of device use, and the attitude and motivation of the child. Targeted rehabilitation may also be important, both for its effect on the development of listening skills and on the recognition by the child of the value of the input provided by CI-2.

Although the influential factors cannot be confirmed, the outcomes identify factors that are not necessarily barriers to success with bilateral implants. These are: limited or no previous consistent use of a hearing aid (as was the case for all of the present subjects), limited experience using the second implant alone (as was the case for all but S8), limited rehabilitation targeting use of the second implant (as was the case for S3, S4 and S7), and relatively advanced age at implantation and delay between implants (as was the case for S4). These findings are in contrast to the expectations of the authors based on unilateral experience, and are in contrast to other reports of bilateral research. With regard to second implant alone experience, (Kühn-Inacker et al., 2004) suggested daily experience was mandatory. With regard to specific rehabilitation, Kühn-Inacker et al. (2004) and Huarte, Cervera-Paz, Rama et al. (Reference Note 2) concluded that listening skills with a second implant would not develop without specific rehabilitation. With regard to age at implantation, (Peters et al., 2004) concluded that children under 8 years acquire listening skills in the second ear faster than older children, and Litovsky, Johnstone, Parkinson et al. (2004) suggested that there may be a critical period for maximizing benefit from a second implant, based on the absence of demonstrated benefit for their 12 year old subject. Clearly, success with a second implant is a complex interaction of factors that cannot be predicted based only on clinical and research experience with unilateral implants, combined with the pediatric bilateral studies conducted so far.

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Conclusions

The results of the present study indicate that children over the age of 4 years may gain significant additional benefit from a second implant, though there is great inter-subject variation. Those who do gain benefit demonstrate improved speech perception in some noise contexts and functional advantages in daily life. However, there is virtually no objective evidence of true binaural listening, including localisation skills, at least in the first 6 months. The factors influencing benefit cannot be clearly identified, though limited pre-operative auditory experience with the second ear, a delay of years between implants, relatively advanced age, and lack of second-implant-alone experience do not preclude benefit. There is a clear need for further research to increase understanding of the potential benefit of bilateral implantation for individual children. The present subjects will be re-evaluated at 12 months and 24 months post-operative to measure changes with further experience, with additional subjects also being recruited to this ongoing project.

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Appendix A: Questions and response format for the "Speech, Spatial and Qualities of Hearing Scale for Parents of Children with Impaired Hearing"

Questions

Section A: Speech Perception

1. You are talking with your child and there is a TV on in the same room. Without turning the TV down, can your child follow what you're saying?

2. You are talking with your child in a quiet, carpeted lounge-room. Can your child follow what you're saying?

3. Your child is in a group of about five people, sitting round a table. It is an otherwise quiet place. Your child can see everyone else in the group. Can your child follow the conversation?

4. Your child is in a group of about five people, sitting round a table. It is a noisy room, such as a busy restaurant or large family gathering at home. Your child can see everyone else in the group. Can your child follow the conversation?

5. You are talking with your child. There is a continuous background noise, such as a fan or running water. Can your child follow what you say?

6. Your child is in a group of about five people, sitting round a table. It is a noisy room, such as a busy restaurant or large family gathering at home. Your child cannot see everyone else in the group. Can your child follow the conversation?

7. You are talking to your child in a place where there are a lot of echoes, such as a school assembly hall or indoor swimming pool. Can your child follow what you say?

8. You are talking to your child in a room in which there are many other people talking. Can your child follow what you say?

9. Can your child easily have a conversation with a familiar person on the telephone?

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Section B: Spatial Hearing

1. Your child is outdoors in an unfamiliar place. A loud constant noise, such as from a lawnmower, aeroplane or power tool, can be heard. The source of the sound can't be seen. Can your child tell right away where the sound is coming from?
2. Your child is sitting around a table with several people. Your child cannot see everyone. Can your child tell where any person is as soon as they start speaking?
3. You and your child are in different rooms at home. It is quiet. If your child hears you call out their name, will he/she know where in the house you are?
4. Your child is outside. A dog barks loudly. Can your child tell immediately where it is, without having to look?
5. Your child is standing on the footpath of a busy street. Can your child hear right away which direction a bus or truck is coming from before they see it?

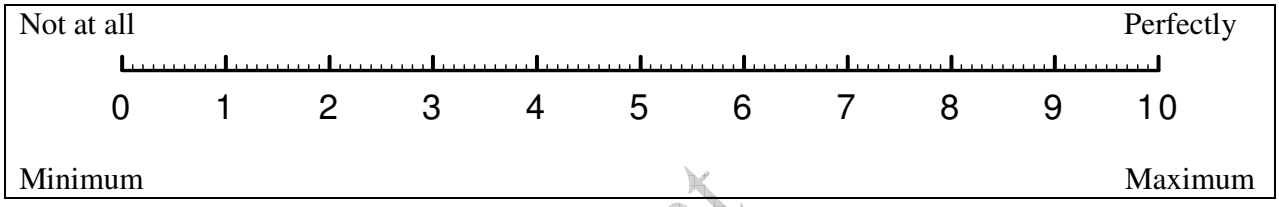
Section C: Quality of Hearing

1. Think about when there are two noises in or around the home at once, for example, water running into the bath and a radio playing, OR a truck driving past and the sound of knocking at the door. Is your child able to identify the two separate sounds?
2. You are in a room with your child and music is playing. Will your child be aware of your voice if you start speaking? Note that the child does not have to understand what you say.
3. Can your child recognise family members or other very familiar people by the sound of each one's voice without seeing them?
4. Can your child distinguish between pieces of music such as different nursery rhymes played on a cassette tape or CD? Note that producing relevant words or movements can indicate recognition.
5. Can your child tell the difference between sounds that are somewhat similar, for example, a car versus a bus, OR water boiling in a pot versus food cooking in a frypan?
6. Can your child easily judge another person's mood from the sound of their voice?

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7. Does your child have to put in a lot of effort to hear what is being said in conversation with others?
8. Can your child easily ignore other sounds when trying to listen to something?

Sample Response Format:



Would not hear it Do not know Not applicable

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