Academic outcomes for school-aged children with severe-profound hearing loss and early unilateral and bilateral cochlear implants

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Abstract

**Purpose:** To (a) determine whether academic outcomes for early-implanted children with cochlear implants (CIs) are age-appropriate,
(b) determine whether bilateral CI use significantly improves academic outcomes, and (c) identify other factors that are predictive of these outcomes.

**Method:** Forty-four eight-year-old children with severe-profound hearing loss participated in this study. Their academic development in mathematics, oral language, reading and written language was assessed using a standardized test of academic achievement.

**Results:** (a) Across all academic areas, the proportion of children in the average or above-average ranges was lower than expected for children with normal hearing. The strongest area of performance was written language, and the weakest was mathematics.
(b) Children using bilateral CIs achieved significantly higher scores for oral language, math, and written language, after controlling for predictive factors, than did children using unilateral CIs. Younger ages at second CI predicted largest improvements.
(c) High levels of parental involvement and greater time spent by children reading significantly predicted academic success, although other factors were identified.

**Conclusions:** Average academic outcomes for these children were below those of children with normal hearing. Having bilateral CIs at younger ages predicted the best outcomes. Family environment was also important to children’s academic performance.

**Key Words:** academic outcomes, cochlear implants, bilateral cochlear implants, children, hearing loss
Since the 1980’s, cochlear implants (CIs) have been used to provide hearing to children with severe-profound hearing loss, as hearing aids do not give sufficient amplification for this degree of hearing loss. Outcomes for children with cochlear implants were measured for many years primarily in terms of speech perception ability, with the underlying assumption being that reasonable speech perception ability would facilitate other aspects of children’s development and learning (Geers, Brenner, & Davidson, 2003; Pyman, Blamey, Lacy, Clark, Dowell, 2000; Sarant, Blamey, Dowell, Clark & Gibson, 2001).

However, although there have been enormous improvements in speech perception outcomes for children with CIs (Blamey et al., 2001; Boothroyd, 1997), it has become apparent that good speech perception ability does not ensure age-appropriate outcomes in other areas. There have been documented improvements in outcomes in speech perception, speech production, language, literacy and social development for children with CIs, and it has been expected that similar improvements in academic outcomes would follow (Marschark, Rhoten & Fabich, 2007). Despite an increase in the proportion of children with CIs who are enrolled in mainstream educational settings (Geers & Brenner, 2003), the impact of CIs on academic outcomes is not yet clear. The proportion of children who achieve academic outcomes that are within the average range for children with normal hearing is still unknown, particularly for older children, since few children have had longer-term follow-up of academic outcomes (Marschark et al., 2007). There is a paucity of research into academic outcomes, with most studies focusing on literacy development, despite delays being observed across the curriculum (Marschark et al., 2007). There have also been methodological criticisms of some studies conducted to date, which have included small sample sizes, ‘estimated’ data, selection bias (the exclusion of inconsistent users and poor performers), bias in testing protocols (reliance on laboratory tasks, rather than those related to real-life), and a lack of prospective and longitudinal studies (Beadle et al., 2005; O’Donoghue, 2006). Failure to control for
variables such as age of implantation and consistency of CI use have also been cited as concerns (Marschark et al., 2007). A further limitation of many studies is that they also have not assessed cognitive ability, which is well-established as one of the most significant predictors of language and educational outcomes (Geers, 2003; Moog & Geers, 2003; Sarant, Holt, Dowell, Rickards, & Blamey, 2009). The effects of additional family factors on academic outcomes for children with CIs, such as parental involvement in children’s intervention/education, children’s reading habits and birth order have also not been investigated. Further prospective research that addresses these concerns is needed, as these are important outcomes that impact significantly on the longer-term futures of these children (Uziel et al., 2007). Academic outcomes are assessed in four categories - Oral Language, Math, Written Language and Reading – existing evidence for each of which is now summarized.

**Oral Language**

Oral language outcomes for children with CIs have been documented over many years (Blamey et al., 2001; Duchesne, Sutton & Bergeron, 2009; Nittouer, Caldwell, & Holloman, 2012). Initially, the performance of children with CIs was compared with that of their peers with severe-profound hearing loss using hearing aids, but as children with CIs demonstrated significantly better outcomes on average (Connor, Hieber, Arts, & Zwolan, 2000; Svirsky, Robbins, Kirk, Pisoni, & Miyamoto, 2000), their outcomes have since been compared with those of their peers with lesser degrees of hearing loss or normal hearing. Over time, with earlier diagnosis of hearing loss, implantation at younger ages improved speech processing technologies, language outcomes have improved. In the 1990’s, there were reports of slower rates of growth (around 50-60% of the rate for children with normal hearing) with around 50% of children having severe language delays (Blamey et al., 2001; Davis & Hind, 1999; Ramakalawan & Davis, 1992). More recently, however, oral language outcomes have
improved such that some children have been reported to acquire language as do children with mild to moderate hearing loss (Spencer et al., 2011).

Language outcomes remain highly variable around these improving averages. A recent report showed how widely distributed language outcomes still are, with superior vocabulary and language outcomes for children with moderately severe – severe hearing loss using hearing aids compared to children with CIs (Fitzpatrick et al., 2012). Further evidence of the wide distribution of outcomes are the reports of children who have received their CIs at very young ages (ie. under two years) achieving oral language development at the same rate as children with normal hearing (Schorr, Roth, & Fox, 2008; Spencer, Barker, & Tomblin, 2003; Tomblin, Barker, Spencer, Zhang, & Gantz, 2005). Although there is a consensus that for most children, CIs facilitate the development of oral language, there is still considerable variation in the rate of oral language development between children, and a significant proportion of children with CIs still exhibit significant delays (Nikopoulous, Dyar, Archbold, & O’Donoghue, 2004; Sarant et al., 2009; Sarant et al., 2009).

Predictive factors for oral language development include ages at diagnosis and implantation (Geers, Tobey, Moog, & Brenner, 2008; Holt & Svirsky, 2008; Kennedy et al., 2006), degree of pre-operative hearing loss (Wake, Poulakis, Hughes, Carey-Sargeant, & Rickards, 2005), cognitive ability (Geers, Moog, Biedenstein, Brenner, & Hayes, 2009; Sarant, Hughes, & Blamey, 2010), socioeconomic status (Dollaghan et al., 1999; Holt & Svirsky, 2008; Niparko, et al., 2010), maternal education (Geers et al., 2009; Sarant et al., 2009) and gender, with females achieving better outcomes than males (Geers et al., 2009; Huttenlocher, Haight, Bryk, Seltzer, & Lyons, 1991). Birth order has also been found to significantly affect spoken language development, (Hoff-Ginsberg, 1998; Nelson, Nygren, Walker, & Panoscha, 2006) and also cognitive development, which impacts indirectly on language (Zajonc & Markus, 1975).
Math

Although studies of children with hearing loss have consistently shown poorer performance in mathematics (Davis & Kelly, 2003; Hyde, Zevenbergen, & Power, 2003; Kelly & Gaustad, 2006), there are very few studies of mathematical ability in children with CIs, with existing studies reporting conflicting results. A large study by Thoutenhoofd (2006), reporting 4-year aggregate National Curriculum Test scores for 152 school-aged children with CIs compared to national data and to data for children with hearing loss gave positive results, with attainment difference scores in Math comparable to those of children with a moderate hearing loss. As the level of attainment rose with age, difference scores for children with CIs increased less than did those for children with hearing loss who were using hearing aids, indicating less delay at higher levels of attainment. However, despite these positive findings, most children with CIs still performed below the national cumulative average.

A study of academic ability in a small sample of 20 Malaysian students with CIs reported superior performance in math examinations compared to language performance (Mukari, Ling, & Ghani, 2007). However, the mean math score was still low; only 62.67%, although 87.5% of the children scored either at or above the average level for children with normal hearing. Children who did well in Math were found to have similar language results.

Motasaddi-Zarandy and colleagues reported very good to excellent Math outcomes for all but one of the 27 Iranian children with CIs in their study, although these results were lower than for language (Motasaddi-Zarandy, Rezai, Mahdavi-Arab, & Golestan, 2009). However, their study did not use a standardized assessment tool, instead using examination marks collected from the different mainstream schools attended by the children. It is therefore questionable whether the same marking standards were used by the different schools, and whether these data can be directly compared in this way. Further, all of the children in their study were chosen as CI candidates based on a lack of additional disabilities.
and having other characteristics known to be favorable to optimal post-operative outcomes, and all were consistent CI users, so their outcomes may not be representative of those of the general population of children with CIs (Beadle et al., 2005; Uziel et al., 2007).

The most recent study of mathematical ability in children with CIs compared the abilities of 24 elementary school-aged children with CIs with that of 22 children with normal hearing of similar age (Edwards, Edwards, & Langdon, 2012). This is the first study that has examined different aspects of mathematical ability, namely arithmetic ability, which included tasks such as addition that required accuracy, and geometrical reasoning, which included approximations, mental rotation, and spatial memory tasks. Again, the performance of children with CIs on both types of math problems was significantly poorer than that of the control group. Similarly to the Mukari study, math skill was found to be mediated by language ability, as assessed by vocabulary knowledge. When vocabulary ability was controlled for, there was a difference in performance between the groups for only one subtest. In particular, it was found that mathematical deficits could be almost entirely explained by vocabulary ability rather than inherent mathematical inability, which is logical given the complex verbal explanations often involved with teaching math.

In summary, the limited literature on the mathematical abilities of children with CIs, has yielded conflicting results, with below average, average, and above average performance documented, and language ability identified as a key predictor of performance. Our current knowledge of mathematical outcomes for children with CIs is poor, and further research is required in this area (Huber & Kipman, 2012; Marschark et al., 2007).

**Written Language**

Studies of written language development in children with hearing loss have primarily examined the use of syntax. The writing of children with hearing loss has historically differed from that of their peers with normal hearing in that sentences are shorter (Kretchmer
phrasing is more repetitive, there are more errors of omission, substitution and word addition, and subject-object-verb constructions are overused (Marschark, Mouradian, & Halas, 1994; Myklebust, 1964; Wilbur, 1977). Errors of morphology, such as plurality, tense and verb agreement are also more common in children with hearing loss (Lichtenstein, 1998), and the use of fewer adjectives, adverbs, prepositions and conjunctions is also reported (Marschark et al., 1994).

There are few studies of written language in children with CIs, particularly those that compare outcomes for this group with those for their peers with normal hearing. One such study of 16 children with CIs (average age 9.8 years) showed a persistence of immature writing patterns that employ shorter and less complex sentences and contain more errors, and the use of an immature writing style that is similar to spoken language (Spencer et al., 2003). However, written productivity and written error rates were still less than one standard deviation from those of normal hearing children, indicating the children should have been able to perform at a similar level to their peers with normal hearing in a regular classroom.

A further study of attainment levels in English writing reported similar results; although children with CIs were not achieving at the same level as those with normal hearing, the performance gap of less than 10% between the two groups clearly illustrated the advantage of CIs over hearing aids, with the CI children functioning more like children with moderate to severe hearing loss, rather than severe-profound hearing loss (Thoutenhoofd, 2006).

A recent study of expository descriptive writing in 112 secondary students showed that fewer than half of the children with CIs scored within one standard deviation of the control group of children with normal hearing in each of the four categories on which scoring was based (organization, content, language use and vocabulary use; Geers & Hayes, 2011). When compared with children with normal hearing, these children with CIs had delayed spelling and expository writing skills.
The results of the first two studies suggest that CIs may have an attenuating effect on the development of writing skills in children with CIs; while many children are still delayed, this delay appears to be less severe than has been reported in the past for children with hearing loss using hearing aids. However, these results may not be representative of current outcomes for children with CIs, as in all of these studies, early implantation was not common. Further research, with children who have been implanted in the first few years of life, should clarify further what the potential in terms of writing development is for children with CIs.

**Reading**

Literacy development in children with severe-profound hearing loss has historically been poor, with reports of many children unable to read beyond fourth-grade elementary level (Geers et al., 2008; Moeller et al., 2007), and 30% of children functionally illiterate when they graduate from secondary school (Traxler, 2000). Even very recent studies of university students with hearing loss, who are presumably among students with the best academic outcomes, have revealed that reading abilities in this population are still below the sixth grade level (Albertini & Mayer, 2011; Parault & Williams, 2010). It has been documented that reading development in many children with hearing loss plateaus at Chall’s stage three (Marschark & Harris, 1996), resulting in ongoing difficulties with ‘reading to learn’, and a lack of ability to use both top-down and bottom-up processing, all of which reduce comprehension and the ability to acquire new knowledge.

However, reading outcomes for a proportion of children with CIs appear to be better than for their peers with hearing aids, with some studies reporting almost four times as many children with CIs achieving a reading level beyond fourth grade (Spencer et al., 2003; Vermeulen, van Bon, Schreuder, Knoors, & Snik, 2007). There is also increasing evidence that some children with CIs can achieve reading scores that are within one standard deviation
of the mean for their peers with normal hearing (Archbold et al., 2008; James, Rajput, Brinton, & Goswami, 2007; Spencer et al., 2003).

Conversely, there are also reports that impressive early reading development in children with CIs may not necessarily be sustained as they grow older, with children who have been followed over time in a series of studies by Geers and colleagues demonstrating age-appropriate reading skills at age 8-9 years, but showing an average delay of almost two years behind grade level when aged 15-16 years (Geers et al., 2008). Thoutenhoofd (2006) also reported similar findings for a group of 152 older school-aged children with CIs, with children aged 11-13 years behind their hearing peers in reading ability by approximately 3 years, and older adolescents (15-17 years) by 4-5 years. This study also observed the wide distribution of equivalent reading ages commonly seen in children with hearing loss in general, and an overall downward curve in reading development over time when compared to children with normal hearing. Further studies have reported sustained or even increased delays in reading ability for other children with CIs (Archbold et al., 2008; Connor & Zwolan, 2004). This lack of consistency in outcomes leaves considerable doubt as to what might reasonably be expected as the ‘average’ reading outcome for children with CIs.

Factors found to affect reading development in children with CIs include higher non-verbal cognitive ability (Geers, 2002; Geers & Hayes, 2011), younger age at implantation (Harris & Terlektski, 2011; Johnson & Goswami, 2010;), female gender (Geers, 2003; Moog & Geers, 2003), later onset of hearing loss (Geers, 2002), family socioeconomic status (Connor & Zwolan, 2004; Geers, 2003) and the use of spoken language as opposed to sign language (Geers, 2002; Venail, Vieu, Artieres, Mondain, & Uziel, 2010). As with normally-hearing children, spoken language abilities have been found to correlate highly with reading abilities (Connor & Zwolan, 2004; Johnson & Goswami, 2010), with children who are competent oral narrators demonstrating better reading comprehension skills (Crosson &
Phonological development, auditory memory, and visual memory have also been linked with reading attainment (Johnson & Goswami, 2010), the first of which has been shown to be delayed in children with CIs, particularly for children with later implantation (Ambrose, Fey, & Eisenberg, 2012; James et al., 2007). Further, speech perception ability has been shown to be highly predictive of reading outcomes (Spencer & Oleson, 2008).

Implant-related characteristics that have been reported as being predictive of better reading development have included a greater number of active electrodes in the implant array, greater dynamic range, and use of recent speech processing software (Geers, 2002). However, as with overall reading outcomes, there is also disagreement between studies about the importance of some of these factors, with age at implantation, for example, not being significantly predictive of outcomes in some studies, particularly those involving older children, where the effect of this factor appears to be attenuated over time (Geers et al., 2008). There may be an indirect relationship between language ability and reading, resulting in the lack of a negative relationship between age at implantation and reading ability (Connor & Zwolan, 2004).

The effect of bilateral CIs

A further factor that has received no attention to date is the effect of bilateral CIs on academic outcomes. Bilateral CIs give significant perceptual benefits over unilateral CIs through binaural redundancy (improved speech perception through two ears, as the brain has two opportunities to process the signal), binaural summation (when combined from two ears, the signal is slightly louder), and the head-shadow effect (the head shields sound, such that the signal will be softer at the ear further from the source). It is also well-established that these perceptual benefits lead to significant improvements in children’s abilities to perceive speech in both quiet (Scherf et al., 2007; Zeitler et al., 2008) and noisy listening conditions (Galvin, Mok, Dowell, & Briggs, 2008; Johnston, Durieux-Smith, O’Connor, & Fitzpatrick,
2009; Lovett, Kitterick, Hewitt, & Summerfield, 2010). However, it is not known whether these perceptual advantages are sufficient to significantly change academic outcomes.

A small number of studies have compared language outcomes on standardized language tests for children with bilateral and unilateral CIs, and mixed findings have been reported. Earlier studies did not find a significant benefit to language development with bilateral implantation (Niparko et al., 2010; Nittrouer, Caldwell, Lowenstein, Tarr, & Holloman, 2012; Nittrouer & Chappman, 2009), however, most of the children in these studies were very young (preschool age), and many had sequential CIs, and therefore had not had long to develop their language using binaural hearing. Further, two of the studies had quite small sample sizes, which may have limited their power to detect a significant difference in outcomes if this existed. Three more recent studies with larger sample sizes and older children have all reported significantly superior performance for children with bilateral CIs on spoken language comprehension and expression, however (Boons et al., 2012a & b; Sarant, Harris, Bennet & Bant, 2014). As far as the authors are aware, there have been no other comparisons of the effects of bilateral versus unilateral CIs on other academic outcomes such as reading, written language or mathematical ability.

The Contribution of this study

This study provides new evidence about outcomes for children with early CIs across a wide range of academic areas, based on results for children with unilateral and bilateral CIs and using a standardized assessment tool (Sarant et al., 2014). In doing so, it contributes to current limited knowledge about academic outcomes for children with CIs in general, and is the only study that has assessed academic performance in this population using a broad standardized array of measures. It also addresses some of the previous concerns about methodological issues in that it is prospective, predictive variables have been controlled, there was no selection bias in which children received CIs, and no estimated data is included.
The current study includes a moderate sample size of 44 children, larger than some previous studies. This study is also the first to investigate the effect of bilateral CIs on academic outcomes, and provides evidence that has implications for parent choices and clinical management of children with CIs in the future. Lastly, the study expands on previous academic outcomes research in terms of identifying new factors that are significantly predictive of outcomes, and accounts for a significant proportion of the variance across all four academic areas examined, namely oral language, math, written language and reading.

Objectives

1. To compare the academic outcomes of children with early CIs to those of their typically developing peers with normal hearing.

2. To determine whether children with unilateral versus bilateral CIs have significantly different academic outcomes.

3. To identify the factors that predict academic outcomes for these children.

This project was approved by the Royal Victorian Eye and Ear Hospital Human Research Ethics Committee (Project 08/846H), The Children, Youth and Woman’s Health service Human Research Ethics Committee (REC2360/3/2014), and the Royal Prince Alfred Hospital Ethics Review Committee (Protocol No. X11-0142 &HREC/13/RPAH/206), Bilateral Cochlear Implants for Children: Does a Second Implant Improve Language, Psychosocial and Other Outcomes? Informed consent for children to participate in this study was obtained from parents in writing.

Method

Participants

Forty-four children aged 8 years were selected from a wider study of outcomes for children with early CIs. The children were recruited from three cochlear implant clinics and
three early intervention centers in four states of Australia, accounting for most the country’s pediatric CI-related services and major intervention centers. Eighty-four percent of the country’s population is located in the area from which the study sample was recruited. Of the eligible children in this area, 51.6% were recruited to the study, consistent with reported recruitment rates for epidemiological studies (Galea & Tracy, 2007). The study cohort consisted of 23 boys and 21 girls. All the children were implanted early (first CI by age 3.5 years and second CI if bilaterally implanted by age 6 years), spoke English as their primary language and had normal cognitive abilities. The average age at diagnosis for children with unilateral CIs was 10.56 months, and the average age at diagnosis for children with bilateral CIs was 7.84 months. Of the 44 children, 34 used bilateral CIs and 10 used unilateral CIs. Seven children with unilateral CIs received their CI before age 2 years, and three over the age of 2 years. Twenty-eight children with bilateral CIs received their first CI before age 2 years and 6 over the age 2 years. Two children received two CIs simultaneously; the remaining 32 children with bilateral implants received them sequentially. Mean length of device use at the time of academic assessment was 7.31 years (SD = 0.13) for the bilateral group and 6.92 years (SD = 0.26) for the unilateral group. There was no significant difference between the groups in terms of pre-operative pure tone average in the better ear. Of the 10 children with unilateral CIs, 6 also used a hearing aid. All of these children had a profound hearing loss in their non-implanted ear, with an average PTA of 111dBSPL.

Twenty-seven of the children presented with a family history of hearing loss. Fifteen of the children had a hearing loss of a genetic origin, and 17 had a hearing loss of unknown cause. The remaining 11 children presented with etiologies resulting from viral causes or medical complications at birth. Further information was collected about the children relating to birth order, birth weight, and hearing aid use (age fitted and whether used pre- and/or post CI). The communication mode for all families was primarily spoken language, with one child
using supplementary sign to communicate with an immediate family member with a profound hearing loss. Information on parent education (see Table 1) was obtained through interviews with parents. In this study an indicator variable was defined stating whether or not the parent has completed a university qualification at or above Bachelor Degree level. That constituted 53% of parents in this study, relative to 43% in the general Australian population in 2013 (Australian Bureau of Statistics publication 6227 Table 12). An immediate family history (parents, grandparents, parent’s siblings) of hearing, reading or speaking difficulties was also recorded for each child three separate 0/1 indicator variables.

**Procedure**

The children in this study were part of a wider study examining outcomes for children with cochlear implants of elementary school age. In accordance with the protocol of the wider project, the children’s academic skills were assessed by speech-language pathologists at 8 years using the Wechsler Individual Achievement Test (WIAT-II; Wechsler, 2001). The cognitive ability of all children was assessed at either 5 or 8 years using one of the two measures listed below.

**Information provided by families**

A reading habits questionnaire designed for this study was mailed to participating families after the children’s eighth birthday (see Appendix 1 for relevant questions). Parents reported the age at which the children had learned to read, and the number of hours per week children spent on each of: reading books, reading aloud to their parents, and screen time (including TV, ipad and gaming devices) were recorded. Part of the questionnaire was based on items used in the study ‘Growing up in Australia: The Longitudinal Study of Australian Children’ (2011). Interviews with parents provided information about levels of parent education, whether there was a family history of hearing loss or a family history of reading
difficulties, rates of hearing aid usage (for children with unilateral CIs), birth order, and birth weight.

**Audiological and CI information**

Audiological information was obtained either through participant’s hospital files or directly from Australian Hearing, the government service provider for all children from birth through to age 26 years. Information about CIs (such as age at implant, mapping and speech processor details) were obtained directly through the children’s CI clinics.

**Academic Assessment**

The Wechsler Individual Achievement Test Second Edition (WIAT–II, Australian Standardized Edition; Wechsler, 2001) was used to assess a broad range of academic skills of the children. The test measures aspects of learning that are expected to take place within a traditional academic setting, is norm-referenced and gives age-based standard scores (mean = 100 and standard deviation = 15) across nine subtests and the four composite areas of learning (Oral Language, Mathematics, Written Language and Reading). Standard scores were used in the statistical analysis. The overall composite reliability coefficient is 0.98.

The Oral Language composite score is based on two subtests, Listening Comprehension and Oral Expression. For Listening Comprehension, the participant is required to match a verbally presented word or sentence to a corresponding picture (eg. “Which picture [out of two] describes the word ‘empty’?”). For Oral Expression, the participant is required to repeat sentences, provide an oral narrative relating to a sequence of pictures and provide oral directions relating to a specific task both with and without a picture prompt (eg. “Explain the steps required to make a peanut butter and honey sandwich”).

The Mathematics composite score is derived from two subtests, Numerical Operations and Maths Reasoning. Numerical Operations requires the participant to solve written math
problems of increasing difficulty requiring addition, subtraction, multiplication and division using whole numbers, fractions and decimals. An early example of a Numerical Operations task is to identifying a missing number in a rote count. Maths Reasoning requires the participant to solve word problems presented in single or multiple steps relating to time, money, measurement, geometry, probability and to read and interpret graphs (eg. using whole numbers to describe quantities).

The Written Language composite score is based on two subtests – Spelling and Written Expression. For the Spelling subtest, the participant is required to spell a target word that is presented in isolation and in a sentence. For Written Expression, tasks vary with age and include the writing of words, sentences and a short passage. Responses are scored for organization, vocabulary, theme development, punctuation and spelling.

The Reading composite score is based on three subtests, the first of which is Word Reading. Tasks for this subtest vary with age, and include the identification of alphabet letters, beginning and ending sounds in words, rhyming pairs and a reading list. The second subtest, Reading Comprehension, requires the participant to read sentences and short passages and answer questions to determine their recognition of target words and understanding of specific content (eg. ‘Lee saw a duck in the water. Then she saw a frog.’). The questions aim to test the ability to make inferences, draw conclusions and use context clues to define unfamiliar words. The third Reading subtest, Pseudoword Decoding, requires the participant to use their phonetic knowledge to read aloud unfamiliar or nonsense words such as: droy /droi/, flid /flid/.

**Cognitive Ability**

The Wechsler Non-Verbal Scale of Ability (WNV; Wechsler & Naglieri, 2006) was used to assess the non-verbal cognitive abilities of 42 children. The WNV is a cognitive assessment that uses pictorial directions rather than language-based instructions, making it
suitable to use with children with hearing loss, language delay or perceptual difficulties. The test is norm-referenced and provides standard score measures of non-verbal cognitive skills. It has a full scale score reliability of 0.91, with performance validity correlations on comparable tests between 0.40 to 0.82.

The Wechsler Preschool & Primary Scale of Intelligence (Hamilton & Burns, 2003) was used to measure the non-verbal skills of two children. The performance IQ scale of the WPPSI-III was used. This scale examines non-verbal cognitive skills through the use of Block Design, Matrix Reasoning and Picture Concepts. The test is norm-referenced and provides scaled score measures of non-verbal cognitive skills. The reliability coefficients for the WPPSI-III United States composite scales range from 0.89 to 0.95. In estimating composite reliability, it is assumed that the items on each subtest share a common construct, and that reliability is a makeup of internal consistency for each subtest. Further, the level of reliability may not be the same between the hearing normative sample and the deaf or hard-of-hearing sample. With the exception of two children (who both scored in the borderline range) all of the children in this study scored either within or above the average range for typically developing children with normal hearing.

**Parent Involvement**

The quality and degree of parental involvement in the children’s intervention and educational programs was measured using the Moeller’s Family Rating Scale (Moeller, 2000). Two professionals who had worked closely with both the child and their family were asked to rate the family’s involvement in the child’s educational program over the course of the previous year. Each family was given a single global rating on a scale of one to five, and raters chose their ratings from specific descriptions of characteristics that represented each participation category. The descriptions included issues such as familial adjustment, session participation, effectiveness of communication with the child, and advocacy efforts of the
parents on behalf of their child. Ratings were: 1 = limited participation; 2 = below average participation; 3 = average participation; 4 = good participation; 5 = ideal participation. Raters were also asked to indicate their confidence in their ratings as questionable, okay or good. Ratings were compared for inter-rater reliability. Complete agreement was found when both raters gave the same score, and categorical agreement was found when raters placed families in the same participation category, as defined above (ie. Ratings 1-2 = below average; 3 = average; 4-5 = above average. Where raters disagreed, an average of the two scores was used. If the raters had different confidence levels, a weighted average was calculated. Ratings were obtained for all families whose children participated in the study.

Statistical Analysis

For each WIAT outcome, a regression was specified of the form

\[ WIAT_i = \alpha_0 + \alpha_1 \text{bilat}_i + \alpha_2 \text{ageCI1}_i + \alpha_3 (\text{bilat}_i \times \text{ageCI2}_i) + \beta_0 \text{IQ}_i + \beta_1'X_{1,i} + \beta_2'X_{2,i} + \beta_3'X_{3,i} + \beta_4'X_{4,i} + U_i, \]

where \( \text{bilat} \) is a 0/1 indicator for the presence of a bilateral CI, \( \text{ageCI1} \) and \( \text{ageCI2} \) are activation ages (in years) of the first and second CI (the latter disappears from the regression for unilateral children because of its interaction with \( \text{bilat} \), as illustrated in equation (3) below). The vectors \( X_{1,i}, X_{2,i}, X_{3,i} \) and \( X_{4,i} \) respectively represent predictors chosen from the Hearing Aid use, Parenting Style, Child Characteristics and Family Background listed in Table 1. The IQ of the child is included as a predictor in every regression.

Given the moderate sample size and relatively large number of possible predictors in \( X_{1,i}, X_{2,i}, X_{3,i} \) and \( X_{4,i} \), a specification for each WIAT outcome was chosen using Hurvich and Tsai’s (1989) bias-corrected Akaike Information Criterion (Akaike, 1974). Specifically, the regressors within each vector \( X_{1,i}, X_{2,i}, X_{3,i} \) and \( X_{4,i} \) were chosen to minimise the AICc. Shibata (1981) proves this produces an optimal predictive model for the dependent variable.
The regression models the *conditional mean* WIAT outcome given CI details and child and family characteristics. Interpretation is in terms of the signs, magnitudes, significance and confidence intervals for the coefficients and, where practically meaningful, additional *marginal effects* (Wang 2004) of particular explanatory variables. A marginal effect is the change in the conditional mean corresponding to a specified change in an explanatory variable, holding other explanatory variables constant.

To illustrate, the marginal effect of an single point of IQ is the difference between the means for a given IQ score and for that IQ score plus one, with all other variables constant:

\[
\left( \alpha_0 + \alpha_1 \text{bilat}_i + \alpha_2 \text{ageCI1}_i + \alpha_3 \text{ageCI2}_i + \beta_0 \text{IQ}_i + 1 + \beta_1 X_{1,i} + \ldots + \beta_4 X_{4,i} \right) \\
- \left( \alpha_0 + \alpha_1 \text{bilat}_i + \alpha_2 \text{ageCI1}_i + \alpha_3 \text{ageCI2}_i + \beta_0 \text{IQ}_i + \beta_1 X_{1,i} + \ldots + \beta_4 X_{4,i} \right) \\
= \beta_0. \tag{2}
\]

The marginal effect of one point of IQ is therefore given by its coefficient, and is the same regardless of the values of all explanatory variables. However it may be more interpretable to consider the marginal effect of 10 points of IQ, in which case the same calculation would give a marginal effect of \(10 \times \beta_0\). (Similarly a one S.D. increase in IQ has a marginal effect of \(\text{S.D.} \times \beta_0\), although standardised effects are not used here.) If \([\beta_{0,L}, \beta_{0,U}]\) is a 95% confidence interval for \(\beta_0\), a 95% confidence interval for \(10 \times \beta_0\) is given by \([10 \times \beta_{0,L}, 10 \times \beta_{0,U}]\).

The marginal effect of a bilateral CI, relative to having only one CI, involves two explanatory variables: the indicator \(\text{bilat}_i\) and the age of implant \(\text{ageCI2}_i\). The difference between the conditional mean with a bilateral CI at age \(a\) (i.e. \(\text{bilat}_i = 1\) and \(\text{ageCI2}_i = a\)) and with no bilateral CI (i.e. \(\text{bilat}_i = 0\) and \(\text{ageCI2}_i\) irrelevant) is

\[
\left( \alpha_0 + \alpha_1 \times 1 + \alpha_2 \text{ageCI1}_i + \alpha_3 \times a + \beta_0 \text{IQ}_i + \beta_1 X_{1,i} + \ldots + \beta_4 X_{4,i} \right) \\
- \left( \alpha_0 + \alpha_2 \text{ageCI1}_i + \beta_0 \text{IQ}_i + \beta_1 X_{1,i} + \ldots + \beta_4 X_{4,i} \right) \\
= \alpha_1 + \alpha_3 \times a. \tag{3}
\]
The marginal effect of a bilateral CI therefore varies with the age of implant of the second CI. To demonstrate this variation, the marginal effects were calculated at ages 1, 2, 3 and 4 years.

**Results**

Table 1 provides summary statistics of the hearing aids and CIs of the participants along with their other demographic characteristics. Table 2 gives descriptive statistics for the WIAT test scores. Also provided for each score is the percentage of children whose scores fell below one standard deviation below the mean (i.e. with a standard score of less than 85), within the normal range (85-115) and above the normal range (>115). Table 2 also reports \( p \)-values for the difference of means \( t \)-test between the bilateral and unilateral samples, showing that no differences are statistically significant. Table 3 gives correlations between the variables, with accompanying \( p \)-values showing most are significant.

The regression results are presented in Table 4. There are four equations presented as the columns of the table, one for each of the four WIAT outcomes. For each predictor, the estimated regression coefficient is presented, along with its \( p \) value, the contribution of the predictor to the overall regression \( R^2 \), and a 95% confidence interval for the coefficient.

**Oral Language**

The mean Oral Language score for children in this study was 92.52, with 68% scoring within or above one standard deviation of the mean for typically developing children with normal hearing (Table 2). This is considerably less than the 85% of children in the normative population who score within this range. The mean Oral Language score for children with bilateral CIs was 93.82, insignificantly different from 88.10 for children with unilateral CIs.

The regression results in Table 4 for the Oral Language outcome show that both of the bilateral CI predictors were important. The presence of bilateral CIs predicted a significantly larger average Oral Language test outcome compared to only a unilateral CI, with the effect moderated by the age at second implant. Following equation (3), the marginal effect of a
bilateral CI activated at age $a$ years, relative to having no bilateral CI, was estimated to be $30.46 - 8.17 \times a$. Evaluating this for ages $a = 1, 2, 3$ and $4$ years gave marginal effects of $22.29, 14.12, 5.95, -2.22$ points respectively, which emphasizes that the largest gains occur for earlier implantation. The WIAT tests are standardized to have mean 100 and standard deviation 15 in the normal hearing population, so effect sizes of 22.29 and 14.12 points are both statistically and practically significant. Such gains in Oral Language by age 8 were not found for the older implant ages. Child age at first implant had a negative coefficient as may be expected, but was insignificant and is therefore not further interpreted.

Average Oral Language outcomes were also predicted by IQ and by the amount of time the children spent reading books. The marginal effect of an extra 10 points of IQ was to increase the average Oral Language score by 6.3 points, with 95% confidence interval [2.8, 9.8]. The estimated effect of the bilateral CI could be expressed in terms of IQ points – a bilateral CI activated at age 2 is equivalent in its effect size on Oral Language scores to an extra $14.12 \div 0.63 = 22.41$ points of IQ.

Time spent reading books predicted higher average Oral Language outcomes. An additional 15 minutes per day (approximately 1 S.D.) predicted an increase in average Oral Language scores of 7.3 points, with 95% confidence interval [3.0, 11.6] points.

Math

The mean Math score for the children in this study was 85.52 (Table 2), and only 57% scored within the average to above-average range, lower than on any other section of this test. The mean scores for children with bilateral CIs (86.56) and unilateral CIs (82.00) were not significant different. The marginal effect on the Math score of a bilateral CI activated at age $a$ years, relative to having no bilateral CI, was estimated to be $20.94 - 4.54 \times a$. Evaluating this for ages $a = 1, 2, 3$ and $4$ years gave marginal effects of 16.40, 11.86, 7.32 and 2.78 points respectively. Child age at first CI was not statistically significant.
Higher IQ predicted higher average Math scores. Each 10 points of IQ predicted an increased average Math score of 6.8 points, with 95% confidence interval [3.9, 9.7].

**Written Language**

Mean Written Language scores were 96.66 (Table 2), with no significant different between the means for those with bilateral CIs (97.06) and unilateral CIs (95.40). The 80% of children within 1SD of the normative mean was close to the 85% of children who would be expected to score within this range in the normative population.

Bilateral CIs had a significant effect on average Written Language outcomes only through the age of activation predictor, and the marginal effect size was relatively small. For example, a bilateral CI at age two years predicted an average Written Language score $9.87 - 3.92 \times 2 = 2.03$ points higher than that for children with only a unilateral CI. This is not as practically significant as the larger effects found for the Oral Language and Math scores.

The most important predictor of Written Language outcomes was the amount of time children spent reading, with the largest unique contribution (0.25) to the overall $R^2$. An extra 15 minutes of time spent reading books each day predicted an increase of 9.65 points, with 95% confidence interval [5.9, 13.4], a substantial improvement for a small time commitment.

**Reading**

The mean Reading score for these children was 91.70 (Table 2) with only 67% of children within the average-to-above-average range. Mean Reading mean scores for children with bilateral versus unilateral CIs were 92.64 and 88.6 respectively. Neither bilateral CIs nor age at first CI were found to have any statistically significant effect. Reading scores were predicted by IQ, time spent reading books, parental involvement (MFRS) and birth order. The signs of these effects were all as expected, with higher IQ, more time spent reading, increased parental involvement and fewer older siblings each predicting increased average Reading scores. Estimated effect sizes are summarized by the confidence intervals in the table.
Discussion

The academic achievement of children with severe-profound hearing loss lags that of their peers with normal hearing (Traxler, 2000; Schick, Williams, & Kupermintz, 2006). The academic performance of this group of children with CIs was also lower than would be expected for typically developing children with normal hearing, even though their cognitive ability was (with two exceptions) within or above the average range. However, the proportions of children scoring within one standard deviation of the means (see Table 2) suggest that having a CI attenuates the lag relative to those with hearing aids (Marschark et al., 2007), consistent with earlier studies (Spencer et al., 2003; Geers & Hayes, 2011).

Oral Language

For Oral Language, 68% of the children in this study scored higher than one standard deviation below the normative mean, more than many reported in the past based on results of standardized language assessment tools, which range from 39% to 58% (Blamey et al., 2001; Geers et al., 2009; Nicholas & Geers, 2008). The mean standardized score for Oral Language (92.52) was also higher than in most past reports, but is comparable with a more recent study (Geers & Nicholas, 2013) in which, like the present study, children received CIs at an early age. The outcomes still lag those of children with normal hearing though.

The regressions found a significant positive effect of bilateral CIs on the mean Oral Language composite score relative to only unilateral CIs, reflected in the percentages above one SD below the mean (74% vs 50% for bilateral vs unilateral CIs) Superior oral language results on standardized assessments have previously been found. (Boons et al., 2012 a & b and Sarant et al. (2014). Bilateral CIs provide hearing in both ears, conferring perceptual advantages and greater ease of listening and significantly improved speech perception in quiet and noisy conditions. Also 84% of children report increased confidence in group
conversations after bilateral implantation (Redfern & McKinley, 2011). These factors should facilitate faster language learning.

Earlier bilateral implantation predicted better outcomes and contributed most to the regression $R^2$ (16%). The period of auditory deprivation is a highly influential predictor of CI outcomes (Connor et al., 2006; Archbold et al., 2008; Sharma, Dorman, & Spahr, 2002). Recent studies comparing cortical function between ears show that despite the fact that many children with unilateral CIs develop reasonably mature brainstem and thalamo-cortical responses to sound after long-term CI use, there is a loss of normal cortical response in the deprived ear if implantation of a second CI does not occur either simultaneously or with limited delay (Gordon, Jiwani, & Papsin, 2013).

Time spent reading books was a significant positive predictor for Oral Language, explaining 9% of total variation. Reading books has been established as a strong predictor of reading outcomes for children with normal hearing (Stanovich, 1986). One study has examined the effects of the reading habits of adult university students with CIs (Marschark et al., 2012), but the authors are unaware of previous investigations of this factor in children with CIs. Cognitive ability was the only other significant predictor for Oral Language, and is a well-established predictor of language outcomes (Geers et al., 2009; Sarant et al., 2009).

**Math**

Children in this study performed worst in Math, with only 57% within or above 1 S.D. of the normative mean, well below the 85% standard. Other studies have reported similar findings for children with CIs (Hyde et al., 2003; Kelly & Gaustad, 2006; Motasaddi-Zarandy et al., 2009). Conversely, Mukari and colleagues reported 20 children with CIs performing at a poorer level in language than in Math (Mukari et al., 2007). A prime limiting factor for Math performance in children with hearing loss is language (Edwards et al., 2012), since
complex language is often used to explain Math. Language delay is common in the CI population.

The regression found a significant positive marginal effect of a bilateral CI on Math scores, with largest effects for younger bilateral implantation. This is a novel finding, the Math ability of children with unilateral and bilateral CIs not having been compared before.

Cognitive ability also predicted significantly higher Math scores, with the highest variance contribution of any factor across all four academic areas (26%). Cognitive executive functions in normal hearing children, such as processing speed and spatial ability have also been shown to be predictive of mathematical ability (Rhode & Thompson, 2007).

**Written Language**

Written language outcomes for the children in this study were very encouraging, with an average standard score of 96.66, and 80% of the children scoring either within or above the average range for children with normal hearing. The Written Language score is based on an assessment of linguistic elements (ie. vocabulary and syntax), but also on organization, vocabulary, theme development, punctuation and spelling. These outcomes are very close to those for children with normal hearing. However, there is little comparable CI literature. Motasaddi and colleagues (2009) reported very high exam scores on Persian literature for a small sample of ideal CI recipients, but these results were not standardized assessments, and it is unclear what ‘Persian literature’ was. A further study by Thoutenhoofd (2006) reported national attainment data showing that students with CIs performed at a lower level than the national average for written English, but data were not provided for comparison.

The marginal effect of bilateral CIs relative to CIs was smaller for Written Language than for Oral Language or Math. Nevertheless, earlier bilateral implantation was a significant positive predictor for Written Language outcomes, a novel finding in this literature.
The amount of time children spent reading was the most important predictor of Written Language. Quantifying the contribution of this factor to written language development for children with CIs is a novel contribution, although it is well-known that for children with normal hearing, reading has a significant positive effect on the development of writing skills (Hafiz & Tudor, 1989). Given as little as an additional 15 minutes per day of reading time has a significant effect on writing development, it is important that the effectiveness of practical strategies such as this are communicated with parents and educators alike.

**Reading**

Sixty-seven percent of the children in this study scored within or above the average range for children with normal hearing for Reading. A wide range (19-70%) of children in other studies achieved reading scores within one SD of the mean for normally-hearing children (Geers, 2003; Harris & Terlektski, 2011; Johnson & Goswami, 2010; Moog, 2002). There are also other reports of children achieving similar standards of reading ability to their peers with normal hearing (Archbold et al., 2008; Spencer et al., 2003; Spencer & Oleson, 2008), however, as mentioned earlier, there are still many who are delayed (Archbold et al., 2008; Geers et al., 2008), and some who do not progress at all (James et al., 2007).

The standardized reading scores in the current study where slightly higher than for the Geers (2003) study and slightly lower than for James and colleagues (2007). Such variations may be partly explained by age differences, as children often do not maintain their rate of reading development, with widening lags due to the increasing difficulty of the task with increasing grade level at school (Kroese, Lotz, Puffer, & Osberger, 1986; Marschark & Harris, 1996). Although not found in the current study, age at CI has also been shown by others to have a significant effect on reading outcomes, with later-implanted children developing reading more slowly, and this factor may also account for some of the observed differences (Archbold et al., 2008; Connor & Zwolan, 2004; James et al., 2007).
Although average Reading Composite scores for children with bilateral CIs (92.64) were higher than for children with unilateral (88.60) CIs, Reading was the only area of the four assessed in this study where a significant difference between children with unilateral and bilateral CIs was not seen. Given the trend towards better reading performance for children with bilateral CIs, their significantly better results for Oral Language, Math, and Written Language, and the moderate sample size, a significant difference may be found in a larger sample. As this study is ongoing, this may be determined in the near future.

Higher levels of parental involvement predicted significantly higher Reading scores and was the most important predictor, explaining 12% of the overall variance. This emphasizes the influence of parent involvement in children’s education, and highlights the importance of professionals facilitating and encouraging parents to provide this support to their children. This factor also predicts better language outcomes (Moeller, 2000; Sarant et al., 2009), with involved parents demonstrating better communication skills (Fallon & Harris, 1991).

As for Oral and Written Language, the amount of time children spent reading was significantly predictive of reading abilities. Practice improves reading skill, with reading frequency previously found to explain reading ability in children with hearing loss (Limbrick, McNaughton, & Clay, 1992). Children born earlier in the family also achieved better reading results. Birth order has previously been identified as an influential factor in outcomes for all children, regardless of their hearing status (Hoff & Ginsberg, 1998; Zajonc & Markus, 1975). The theory is that for children born earlier in the family parents have greater personal and financial resources available with which to facilitate children’s development. Finally, cognitive ability also significantly affected reading performance, consistent with other research (Johnson & Goswami, 2010; Spencer & Oleson, 2008).

Conclusions
This study extends the literature showing that children with CIs can achieve academic outcomes similar to their peers with normal hearing, although there are no guarantees that this will occur. The outcomes of this study suggest that giving children binaural hearing through two CIs provides significant benefits in Oral Language, Written Language and Math over a single CI and can assist a number of profoundly hearing impaired children to achieve age-appropriate academic performance. The benefit is greatest when the second CI is implanted at a younger age. These benefits were statistically significant despite the relatively small number of children with a single CI in the sample. This aspect of the sample may imply imprecise estimates of the effect sizes of a bilateral implant, although any such imprecision is reflected in the reported confidence intervals. Further research with a larger sample size should increase the precision of the estimates of the effect sizes.

Findings of practical clinical relevance in this study include the importance of high levels of parental involvement in children’s intervention and educational programs, and of helping children to establish a regular reading habit. These factors were found to be highly influential on academic outcomes in this group of children. For example, an intervention such an additional 15 minutes per day of reading time has a significant beneficial effect on writing development, and it is important that the effectiveness of practical strategies such as this are communicated with parents and educators alike.

Follow up of children who have received early CIs, and bilateral CIs, is required to evaluate future academic progress. It has been reported that as children grow older and the demands of the curriculum increase, their rate of progress may slow, and learning difficulties may become more apparent (Blamey et al., 2001; Geers et al., 2008; Thoutenhoofd, 2006).

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Johnston, J. C., Durieux-Smith, A., Angus, D., O'Connor, A., & Fitzpatrick, E. M. 


Table 1. Descriptive statistics for CI and HA details and demographic variables.

<table>
<thead>
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<th>Bilateral</th>
<th>Unilateral</th>
<th>Difference</th>
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<tbody>
<tr>
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<td>SD</td>
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</table>

^a Ages at first and second CI in decimalized years; ^b age of first hearing aid fitting in months; ^c 0/1 indicators for hearing aid use before / after first CI; ^d MFRS ratings 1-5; ^e times spent looking at screens or reading in hours per week; ^f IQ standard score; ^g birth weight in kgs; ^h 0/1 indicators for family history of hearing, reading and speaking difficulties. ^i parent higher education indicator = 1 for Bachelor’s degree and above, = 0 otherwise.
Table 2. Statistics for WIAT outcomes.

<table>
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<tr>
<th></th>
<th>Full sample (n = 44)</th>
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<th>Unilateral (n = 10)</th>
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<tr>
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<sup>a</sup> p value for difference of means t test between bilateral and unilateral samples.
Table 3. Correlations between WIAT composite scores and predictive factors

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Table 4. Regression results

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For each explanatory variable, the estimated regression coefficient is on the first line, its $p$-value on the second line, its $R^2$ contribution on the second line, and 95% confidence interval for the coefficient on the third line.

*, **, ***: $F$ statistic significant at 5%, 1%, 0.01% levels respectively
Appendix 1.

Excerpts from Reading Habits Questionnaire

**Reading to parents**

12. At what age did your child start reading to you? Please respond in years and months.

13. Currently, in a typical week, how many days per week does your child read out loud to you?

a) None  b) 1-2 days  c) 3-5 days  d) Every day

14. Currently, for about how many minutes does your child read out loud to you at a sitting?

a) Child doesn’t like to read out loud at all  b) less than 5 minutes  c) 6-10 minutes  
d) 11-15 minutes  e) 16-21 minutes  f) 21-40 minutes  g) 41-60 minutes

**Reading Alone/to themselves**

15. Currently, in a typical week, how many days per week does your child read books, or look at books, on their own?

a) None  b) 1-2 days  c) 3-5 days  d) Every day

16. Currently, on average, for about how many minutes does your child look at books, or read on their own, at a sitting?

a) Child doesn’t like to read at all  b) less than 5 minutes  c) 6-10 minutes  
d) 11-15 minutes  e) 16-20 minutes  f) 21-40 minutes  g) 41-60 minutes

**Screen Time**

21. On a typical weekday, for how many hours does your child watch TV, DVDs, computer games, Wii etc?
Minerva Access is the Institutional Repository of The University of Melbourne

Author/s:
Sarant, JZ; Harris, DC; Bennet, LA

Title:
Academic Outcomes for School-Aged Children With Severe-Profound Hearing Loss and Early Unilateral and Bilateral Cochlear Implants

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2015-06-01

Citation:

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