

# Bilateral Versus Unilateral Cochlear Implants in Children: A Study of Spoken Language Outcomes

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**Objectives:** Although it has been established that bilateral cochlear implants (CIs) offer additional speech perception and localization benefits to many children with severe to profound hearing loss, whether these improved perceptual abilities facilitate significantly better language development has not yet been clearly established. The aims of this study were to compare language abilities of children having unilateral and bilateral CIs to quantify the rate of any improvement in language attributable to bilateral CIs and to document other predictors of language development in children with CIs.

**Design:** The receptive vocabulary and language development of 91 children was assessed when they were aged either 5 or 8 years old by using the Peabody Picture Vocabulary Test (fourth edition), and either the Preschool Language Scales (fourth edition) or the Clinical Evaluation of Language Fundamentals (fourth edition), respectively. Cognitive ability, parent involvement in children's intervention or education programs, and family reading habits were also evaluated. Language outcomes were examined by using linear regression analyses. The influence of elements of parenting style, child characteristics, and family background as predictors of outcomes were examined.

**Results:** Children using bilateral CIs achieved significantly better vocabulary outcomes and significantly higher scores on the Core and Expressive Language subscales of the Clinical Evaluation of Language Fundamentals (fourth edition) than did comparable children with unilateral CIs. Scores on the Preschool Language Scales (fourth edition) did not differ significantly between children with unilateral and bilateral CIs. Bilateral CI use was found to predict significantly faster rates of vocabulary and language development than unilateral CI use; the magnitude of this effect was moderated by child age at activation of the bilateral CI. In terms of parenting style, high levels of parental involvement, low amounts of screen time, and more time spent by adults reading to children facilitated significantly better vocabulary and language outcomes. In terms of child characteristics, higher cognitive ability and female sex were predictive of significantly better language outcomes. When family background factors were examined, having tertiary-educated primary caregivers and a family history of hearing loss were significantly predictive of better outcomes. Birth order was also found to have a significant negative effect on both vocabulary and language outcomes, with each older sibling predicting a 5 to 10% decrease in scores.

**Conclusions:** Children with bilateral CIs achieved significantly better vocabulary outcomes, and 8-year-old children with bilateral CIs had significantly better language outcomes than did children with unilateral CIs. These improvements were moderated by children's ages at both first and second CIs. The outcomes were also significantly predicted by a number of factors related to parenting, child characteristics, and family back-

ground. Fifty-one percent of the variance in vocabulary outcomes and between 59 to 69% of the variance in language outcomes was predicted by the regression models.

**Key words:** Bilateral, Children, Cochlear implant, Spoken language, Unilateral.

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## INTRODUCTION

Although many children with unilateral cochlear implants (CIs) have excellent speech perception abilities in a controlled listening environment such as a quiet room or sound-proof booth (Sarant et al. 2001; Leigh et al. 2008) these environments do not represent listening conditions in the real world. In more difficult listening conditions, such as noisy classrooms or playgrounds and the family home, children with a unilateral CI and a severe to profound or profound hearing loss in the contralateral ear will experience significant difficulties, which will reduce the amount and quality of speech they are exposed to. Understanding speech that is soft, speech in background noise, and locating sound sources such as speakers in a group conversation are examples of such difficulties. With these perceptual limitations, it is less likely that children with unilateral CIs will have the ability to learn incidentally through “overhearing,” as do children with normal hearing, which limits their acquisition of language, world knowledge, and social skills. Although many children with CIs have been able to develop spoken language and other skills that would not have been possible with conventional hearing aids, it has been well-documented for many years through to the present time that many children with unilateral CIs show delays in the development of language (Blamey et al. 2001; Geers 2002; Nittrouer et al. 2012), speech production (Tobey et al. 2003; Connor et al. 2006; Spencer et al. 2011), literacy (Crosson & Geers 2001; Marschark et al. 2007; Geers & Hayes 2011), academic (Spencer et al. 2003; Mukari et al. 2007) and social skills (Bat-Chava et al. 2005; Hintermair 2006). Although a number of children with unilateral CIs have been able to achieve age-appropriate development in many of these areas (Spencer et al. 2004; Percy-Smith et al. 2008; Duchesne et al. 2009), for a significant number of these children developmental delays have been maintained or increased through to adulthood (Moeller et al. 2007; Mukari et al. 2007; Uziel et al. 2007; Geers et al. 2008). For this reason, the efficacy of bilateral CIs is being investigated, and bilateral cochlear implantation is becoming the standard of care for children with severe to profound hearing loss in developed countries around the world (National Institute on Deafness and other Communication Disorders, 2011).

### Perceptual Benefits of Bilateral CIs

Bilateral CIs offer additional benefits over a unilateral CI through the mechanisms of binaural redundancy (speech

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perception is improved with 2 ears, as the brain has 2 opportunities to process the signal), binaural summation (the signal when combined from 2 ears is slightly louder than from 1 ear), and the head-shadow effect (the head acts as a physical barrier to the sound, such that the signal will be softer at the ear that is farthest from the sound source). The benefits of bilateral CIs for speech perception in children have been evaluated in both noisy and quiet listening conditions. In noise, many studies have reported a significant improvement in children's abilities to perceive speech (Litovsky et al. 2006b; Galvin et al. 2008; Johnston et al. 2009; Lovett et al. 2010). In quiet listening conditions, improved speech perception has also been reported (Scherf et al. 2007; Zeitler et al. 2008). Advantages of bilateral CIs for sound localization are not quite as clear, with some children reported to localize sound well (Litovsky et al. 2006a; Lovett et al. 2010), and others demonstrating more limited localization ability (i.e., left–right lateralization, rather than true localization) (Galvin et al. 2008; Grieco-Calub & Litovsky 2010). Many other children, particularly older children, have shown no ability to localize sound (Galvin et al. 2007). Further benefits have also been documented, with parents in some studies frequently reporting superior performance using bilateral CIs in everyday life, in situations such as group conversations, background noise, and hearing at a distance (Sparreboom et al. 2012; Galvin et al. in press). There is also objective evidence that for some children with bilateral CIs, listening effort is reduced, suggesting that more attention can then be paid to the learning process (Hughes & Galvin 2013). Despite the above-cited evidence of benefit for children from bilateral CIs, it is yet to be determined whether these perceptual benefits facilitate significantly better broader outcomes in children with bilateral CIs, and if so, to quantify the degree of benefit received by children based on factors such as age at second implant.

### Effect of Bilateral CIs on Language Outcomes

Until recently, most of the research on outcomes for children with bilateral CIs was focused on speech perception and sound localization benefits. Evidence regarding whether bilateral CIs significantly improve broader outcomes such as language, literacy, academic and social skills, and overall quality of life is lacking, particularly regarding longer-term outcomes (Johnston et al. 2009; Sparreboom et al. 2010). The results of the few earlier studies comparing language outcomes for children with unilateral and bilateral CIs did not show a significant benefit to language development from bilateral implantation. One of the first studies to investigate the effect of bilateral CIs on vocabulary, receptive and expressive language of children assessed at 3.5 years of age (15 unilateral, 26 bilateral) concluded that the reported perceptual benefits from bilateral CI use “may not extend to generative language” (Nittrouer & Chappman 2009). A further study of language outcomes in preschool-age children implanted by 5 years of age (60 unilateral, 31 bilateral; average age 2 to 5 years) reported similarly that children with bilateral CIs did not receive a significant benefit to either receptive or expressive language development over children with unilateral CIs (Niparko et al. 2010). It was noted that this outcome may have been a result of the brief period of time that had elapsed between implantation of the second CI and follow-up. A third study of vocabulary and language development in children who had just completed preschool (13 unilateral, 14 bilateral) and

were up to 6 years of age also reported that having bilateral CIs had no effect on language outcomes (Nittrouer et al. 2012). In considering the results of these three studies it is worth noting that most of the children were of preschool age and many had sequential CIs, and therefore had not had a long time to use their bilateral hearing to develop language. Two of the three studies also had relatively small sample sizes, which makes detecting a significant difference in performance (if it exists) difficult, given the large variance in language outcomes commonly reported (Spencer et al. 2003; Connor et al. 2006; Sarant et al. 2009).

Two recent reports from the same population of children contradict the results of the first three studies. A retrospective study of 288 children implanted by 5 years of age examined language outcomes each year over 3 years post-CI for up to 29 children with bilateral CIs, compared with up to 85 children with unilateral CIs, and up to 62 children with a CI and a hearing aid (Boons et al. 2012a). It was concluded that contralateral stimulation (with bilateral CIs or unilateral CI plus hearing aid) contributed to significantly improved language outcomes. An unspecified post hoc analysis separated the bilateral CI and hearing aid effects, with the finding that bilateral CIs led to better outcomes than unilateral CIs, and also than bimodal hearing (CI plus hearing aid). This study differed from two previous studies in that the children were slightly older (up to 6 years of age), although the number of children with bilateral CIs was not greater. A limitation of the study was that participating children used different CIs (Cochlear Ltd., Sydney, Australia or Advanced Bionics, Valencia, CA), which have recently been reported to give significantly different perceptual results (Lazard et al. 2012) and could therefore have affected language outcomes. Although the effect of child age at first CI was considered, with children implanted before the 2 years of age performing significantly better on all measures, the effect of age at second CI was not investigated.

The same researchers also compared the spoken language outcomes of a smaller sample of children selected from the larger retrospective study (25 unilateral and 25 bilateral; Boons et al. 2012b). The children were matched for several auditory features, sex, implantation age, lack of additional disabilities, a monolingual family background with normal-hearing parents, and educational setting. As for the larger study, it was reported that the performance of children with bilateral CIs on spoken language comprehension and expression was superior.

### Contribution of This Study

Evidence of the impact of bilateral implantation on language outcomes is vital if evidence-based preoperative recommendations are to be made to parents considering CIs for their children. This evidence can also be used by governments around the world to make policy decisions on whether to fund bilateral implantation in children. The current evidence, as described earlier in the article, is limited and shows mixed findings. There is therefore a need for further research in this area, particularly with regard to longer-term outcomes, given that most studies to date have involved children of preschool age with limited CI experience.

The present study offers a further comparison of vocabulary and language outcomes in children using unilateral and bilateral CIs. It has the advantages of being prospective, and including older children than in previous studies, thus offering some insight into longer-term outcomes. It includes a moderate

sample size of 91 children, with a greater number of children with bilateral CIs than in previous studies. Although the present study has a cross-sectional design, 84% of the country's population was 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 in the last decade for epidemiological studies (Galea & Tracy 2007). The sample can therefore be considered to be reasonably representative of Australian children with CIs. The present study also expands upon previous research in that the effects of age at CI on language outcomes for both the first and second CI are evaluated. The effects of some parenting practices (family reading habits and child screen time) that have not previously been investigated in any studies of language outcomes in children with CIs and their relationships to language outcomes are also examined. Finally, the proportion of variance in language outcomes accounted for in the present study (up to 69%) is higher than in most previous studies.

### Objectives

1. To compare language abilities of children with unilateral and bilateral CI.
2. To quantify the rate of any improvement in language attributable to a bilateral CI.
3. To document other predictors of language development in children with CIs.

## PARTICIPANTS AND METHODS

### Participants

Ninety-one children 5 to 8 years of age were recruited from three CI clinics and three early-intervention centers in four states of the country, accounting for most of the country's pediatric CI-related service organizations 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. This figure is consistent with reported recruitment rates in the last decade for epidemiological studies (Galea & Tracy 2007). Given the fact that the study cohort was recruited from 84% of the country's population, with a recruitment rate similar to recently reported recruitment rates for epidemiological studies, these results can be considered to be reasonably representative of Australian children with CIs.

The study cohort consisted of 44 boys and 47 girls. All children were implanted early (first CI by 3.5 years of age and second CI, if bilaterally implanted, by 6 years of age), spoke English as their primary language, and had normal cognitive abilities. The age at CI criteria were chosen based on physiological studies that suggest that in the absence of normal auditory stimulation there is a period of about 3.5 years during which the central auditory system retains its maximum plasticity, and that this can extend in some children up to the age of approximately 6 to 7 years, after which it is significantly reduced (Sharma et al. 2002, 2005). Of the 91 children, 67 used bilateral CIs and 24 used a unilateral CI. All children used a CI from Cochlear Ltd., with Advanced Combined Encoder speech processing strategy. With the exception of 7 participants, for whom information regarding the number of active electrodes was unavailable at the time of writing, all children had between 19 to 22 active electrodes in their

arrays. Twelve children had been reimplanted. Mean lengths of device use at the time of language assessment were 5.20 years (SD = 1.79) for the bilateral group and 4.55 years (SD = 2.04) for the unilateral group. Hearing aid use, both before and after, CII (if applicable) for all children was documented through parent interviews (see Table 1).

### Demographic Measures

Table 1 provides demographic information. Of the 91 children, 38 presented with a family history of hearing loss, although only 2 children had deaf parents. Of these 38 children, 25 had a genetic cause of hearing loss, 1 had a viral cause, and the etiology for 12 children was unknown. Thirty-five of the children had a hearing loss of a genetic origin and 42 had a hearing loss of an unknown cause. The remaining 14 children presented with etiologies resulting from viral causes or medical complications at birth. Further information about child development was collected relating to birth order, birth weight, age at which children first walked, and history of concerns with fine motor skill development. Only 3 children had a diagnosed additional disability that may have impacted on their ability to learn language. The communication mode for all families was primarily spoken language, with 2 children using supplementary sign to communicate with immediate family members with a profound hearing loss.

### Procedure

The children in this study were part of a wider study examining outcomes for children with CIs. In accordance with the wider project's protocol, the children were assessed at 5 and 8 years of age. To ensure that children were entered into the analyses only once, the most recently collected data for each child was entered. Of the bilateral group, 41 of the children were assessed at 5 years and 26 were assessed at 8 years. Of the unilateral group, 15 were assessed at 5 years and 9 were assessed at 8 years. For each assessment point, language outcomes were measured using the standardized language tests described in Instruments by speech language pathologists. The cognitive ability of all children was assessed at 5 years of age by an educational psychologist using either the Wechsler Non-Verbal Scale of Ability (86 children; WNV; Wechsler & Naglieri 2006) or the Wechsler Preschool and Primary Scale of Intelligence—Third Edition (5 children; WPPSI-III; Wechsler 2002). Normal cognitive ability was defined as a Performance Scale score of 80 or more.

A questionnaire was designed specifically for this study to form a general picture of children's reading habits (see Questionnaire, Supplemental Digital Content 1, <http://links.lww.com/EANDH/A133>). Part of the questionnaire was based on items used in the study *Growing Up In Australia: The Longitudinal Study of Australian Children* (Australian Institute of Family Studies 2012). In addition, questions specific to the aims of the present study were developed. The questionnaire was administered using a Web-based form in the first instance. Where no response was received within a predetermined time frame, questionnaires were sent via mail or filled out during a telephone interview with a member of the research team.

Parental involvement in each child's intervention program was assessed using the Moeller's Family Rating Scale (MFRS; Moeller 2000). Whether or not children had been slow in fine motor skill development, whether parents were

**TABLE 1. Means, standard deviations, and differences between means for participant demographic variables**

	Bilateral			Unilateral			Difference	
	n	Mean	SD	n	Mean	SD	Mean	<i>p</i>
<b>Implant details</b>								
Age CI 1	67	1.37	0.75	24	1.99	0.83	-0.62	0.002
Age CI 2	67	2.94	1.37					
Unilateral years	67	1.57	1.26	24	4.55	2.04		
Bilateral years	67	3.64	1.44					
<b>Hearing aid use</b>								
Before CI 1	66	0.55	0.50	24	0.46	0.51	0.09	0.470
After CI 1	66	0.32	0.47	24	0.54	0.51	-0.22	0.062
<b>Parenting style</b>								
Parent involvement	67	4.52	0.62	24	4.33	0.74	0.18	0.277
Screen time (hr/weekday)	66	1.57	1.78	24	1.63	1.06	-0.06	0.853
Adult reading time (min/week)	67	74.58	59.89	24	70.25	63.27	4.33	0.770
<b>Child characteristics</b>								
Birth order	67	0.84	0.95	24	0.75	0.90	0.09	0.691
Intelligence quotient	67	105.36	13.13	24	103.67	12.07	1.69	0.564
Birth weight (kg)	66	3.22	0.82	24	3.36	0.78	-0.15	0.429
Male sex	67	0.48	0.50	24	0.50	0.51	-0.02	0.853
Age walked (mos)	67	15.31	4.64	24	13.92	3.80	1.40	0.147
Age diagnosis (mos)	67	6.86	7.46	24	6.99	8.19	-0.13	0.947
Fine motor problems	67	0.18	0.39	24	0.17	0.38	0.01	0.891
<b>Family background</b>								
Parent higher education	67	0.36	0.48	24	0.42	0.50	-0.06	0.621
Family history, hearing loss	67	0.36	0.48	24	0.58	0.50	-0.23	0.059
Family history, reading difficulties	67	0.18	0.39	24	0.25	0.44	-0.07	0.485
Family history, speech difficulties	67	0.15	0.36	24	0.25	0.44	-0.10	0.315

CI, cochlear implant.

tertiary educated, and whether there was a family history of hearing loss, difficulty learning to read or learning to speak were documented. All of this demographic information was obtained through a telephone interview with the primary caregiver.

### Instruments

**Peabody Picture Vocabulary Test, Fourth Edition (Australian Standardized Edition)** • The Peabody Picture Vocabulary Test—fourth edition (PPVT-4; Dunn & Dunn 2007) is a norm-referenced (mean = 100, SD = 15), closed-set test of receptive vocabulary that can be used from the ages of 2.6 to 90+ years of age. Children are required to point to one of four pictures that best represents the meaning of a verbally presented stimulus word. All children were assessed using the PPVT-4, and their scores were compared with normative data using standard scores. The average reliability coefficient for this test, based on the normative sample, is 0.89.

**Preschool Language Scale—4 (Australian Language Adaptation)** • The Preschool Language Scale—4 (PLS-4; Zimmerman et al. 2002) was used to assess the receptive and expressive language skills of 5-year-old children. This assessment is norm-referenced (mean = 100, SD = 15), can be used from age of 0 to 6 years, 11 months, and uses various stimulus materials such as toys, pictures, and verbal prompts to elicit responses. Performance on this test is divided into subscale scores for Auditory Comprehension, Expressive Communication, and a Total Language Score. Each child's score is then compared with normative data using standard scores. The reliability

coefficients for this test, based on the normative sample, range from 0.81 to 0.95.

**Clinical Evaluation of Language Fundamentals, Fourth Edition (CELF-4; Australian Standardized Edition)** • The Clinical Evaluation of Language Fundamentals, fourth edition (CELF-4; Semel et al. 2006) was used to assess the receptive, expressive, and global language development of 8-year-old children. This test is designed for use between the ages of 5 to 21 years. For the purpose of this study, the children completed subtests that provided a measure of their skills for Core Language, Receptive Language, and Expressive Language. Each child's standard score was compared with norm-referenced age-based standard scores (mean = 100, SD = 15). The reliability coefficients, based on the normative sample, are 0.69 to 0.91 for the subtests and 0.87 to 0.95 for the composite scores.

**Wechsler Non-Verbal Scale of Ability** • The nonverbal cognitive skills of 86 children were assessed using the Wechsler Non-Verbal Scale of Ability (WNV). The WNV is a cognitive assessment that uses pictorial directions rather than language-based instructions, making it a suitable tool for children with hearing loss, perceptual difficulties, or language delay. The test is norm-referenced and provides standard score measures of nonverbal cognitive skills. It has a full-scale score reliability of 0.91.

**Wechsler Preschool & Primary Scale of Intelligence—Third Edition** • The Performance IQ scale of the WPPSI—III was used to measure the nonverbal intelligence of 5 children. The Performance IQ scale examines nonverbal cognitive skills through the use of block design, matric reasoning, and picture concepts. The test is norm-referenced and provides

scaled score measures of nonverbal cognitive skills. The reliability coefficients for the WPPSI-III United States composite scales range from 0.89 to 0.95.

**Moeller's Family Rating Scale** • The MFRS was used to determine the quality of parental participation in each child's intervention and educational programs. Two professionals or interventionists (e.g., teachers of the deaf, teachers, or early-intervention specialists) were asked to rate the family's participation in the child's early intervention or specialist programs in the year before the time of assessment. Each family was rated on a scale from 1 to 5 (1 = limited participation through to 5 = ideal participation). Raters were asked to base their ratings on specific descriptions or characteristics that represented each participation category. Raters were also asked to estimate the confidence in their own ratings as questionable, okay, or good. Rater's scores were averaged to give an overall rating. If the raters specified different confidence levels, a weighted average was calculated, as specified by Moeller (2000). The MFRS was administered at the time of each child's language assessment.

### Statistical Analysis

**PPVT Linear Regression Analysis** • The sample of PPVT results for 91 children who were 5 and 8 years of age contained considerable variation in the lengths of time the children had used unilateral and bilateral CIs (see second panel of Table 1). The information in this variation was exploited in a regression to estimate the rate at which language ability accumulated over time, and whether this rate was different for children with unilateral and bilateral CIs. The language accumulation regression model had the general form:

$$\begin{aligned} \text{PPVT}_i = & \alpha_0 + \alpha_1 \text{bilat yrs}_i + \alpha_2 (\text{ageCI2}_i \times \text{bilat yrs}_i) \\ & + \alpha_3 \text{unilat yrs}_i + \alpha_4 (\text{ageCI1}_i \times \text{unilat yrs}_i) \\ & + \gamma_1 X_{1,i} + \gamma_2 X_{2,i} + \gamma_3 X_{3,i} + \gamma_4 X_{4,i} + U_i, \end{aligned} \quad (1)$$

in which  $X_{1,i}$ ,  $X_{2,i}$ , and  $X_{3,i}$  each represent predictors respectively chosen from the parenting style, child characteristics, and family background variables in Table 1. For a bilateral child, the variable "bilat yrs" is the length of time (in years) between activation of bilateral CIs and the date of the PPVT assessment, while "unilat yrs" is the length of time between activations of unilateral and bilateral CIs. Only unilat yrs is relevant for a child with a unilateral CI, and is the length of time between unilateral activation and the PPVT assessment.

Equation (1) relates language ability to years of unilateral CI use and, if applicable, years of bilateral CI use. It also allows the effect of each CI to vary according to its activation age. For example, the predicted marginal change in PPVT score due to an additional year spent with bilateral CIs, holding all other variables constant, is given by  $\alpha_1 + \alpha_2 \text{ age CI2}_i$ . It is hypothesized that  $\alpha_1 > 0$ , so that each year spent with bilateral CIs is beneficial for language, and  $\alpha_2 < 0$ , so that bilateral CIs have greater effect when implanted in younger children. The same interpretations apply to  $\alpha_3$  and  $\alpha_4$  for the unilateral CI. Inference can therefore be carried out to compare the rate of language accumulation with bilateral CIs ( $\alpha_1 + \alpha_2 \text{ age CI2}_i$ ) with the rate of language accumulation with a unilateral CI ( $\alpha_3 + \alpha_4 \text{ age CI1}_i$ ), controlled for all the other child, parent, and family characteristics.

Given the moderate sample sizes available and the large number of possible predictors that could be included in Eq. (1), a regression specification was chosen to minimize the Akaike information criterion (AIC), which is a bias-corrected estimator of the Kullback-Leibler divergence between the distribution implied by a statistical model and the distribution of the data (Akaike 1974; Claeskens & Hjort 2008). Specifically, each possible specification of the CI years variables (bilat yrs<sub>i</sub>, age CI2<sub>i</sub>, unilat yrs<sub>i</sub>, age CI1<sub>i</sub>), hearing aid use ( $X_{1,i}$ ), parenting variables ( $X_{2,i}$ ), child characteristics ( $X_{3,i}$ ), and family background variables ( $X_{4,i}$ ) was estimated, and the specification with minimum AIC selected. The statistical analyses were conducted using Eviews v7.1. Quantitative Micro Software, 2010.

### PLS-4 and CELF-4 Linear Regression Analyses

For each PLS-4 and CELF-4 language outcome, a regression was specified for average test scores for children with and without bilateral CIs, controlling for parenting, child, and family characteristics. The differences between predicted test scores with and without the bilateral CI were analyzed to evaluate the effectiveness of the bilateral CI. The language accumulation form of Eq. (1) for the combined sample of 5- and 8-year olds was inapplicable when these age groups were separated. Instead, for a language outcome  $Y_p$ , the regression model was

$$\begin{aligned} Y_i = & \beta_0 + \beta_1 \text{bilat}_i + \beta_2 (\text{ageCI1}_i \times \text{unilat}_i) \\ & + \beta_3 (\text{ageCI1}_i \times \text{bilat}_i) + \beta_4 \text{ageCI2}_i \\ & + \gamma_1 X_{1,i} + \gamma_2 X_{2,i} + \gamma_3 X_{3,i} + \gamma_4 X_{4,i} + U_i, \end{aligned} \quad (2)$$

with  $X_{1,i}$ ,  $X_{2,i}$ ,  $X_{3,i}$ , and  $X_{4,i}$  defined in Eq. (1). The coefficient  $\beta_1$  allowed the average language outcome to differ between children with unilateral and bilateral CIs. In addition, the distinct coefficients  $\beta_2$  and  $\beta_3$  on the interaction terms allowed the language effect of age of first CI to differ between children with unilateral and bilateral CIs. This flexibility is potentially important because age CI1 was, on average, significantly lower for the children with bilateral CIs, and its effect may differ between the two groups. The coefficient  $\beta_4$  allowed the language effect of bilateral CIs to depend on the age of its activation.

To interpret Eq. (2), consider the average effect of a bilateral CI activated at age  $a_2$ . The structure of Eq. (2) allows for the possibility that this effect depends on the age of first CI, denoted  $a_1$ . For given parenting, child, and family characteristics  $x_1$ ,  $x_2$ ,  $x_3$ , the predicted language score with the bilateral CI is

$$y^{\text{bi}} = \beta_0 + \beta_1 + \beta_3 a_1 + \beta_4 a_2 + \gamma_1 x_1 + \gamma_2 x_2 + \gamma_3 x_3 + \gamma_4 x_4,$$

while for a unilateral CI it is

$$y^{\text{uni}} = \beta_0 + \beta_2 a_1 + \gamma_1 x_1 + \gamma_2 x_2 + \gamma_3 x_3 + \gamma_4 x_4.$$

The difference between these predictions

$$y^{\text{bi}} - y^{\text{uni}} = \beta_1 + (\beta_3 - \beta_2) a_1 + \beta_4 a_2$$

is a measure of the effectiveness of the second CI. This decomposes the predicted difference in language due to bilateral CIs into three components: (1) a constant effect  $\beta_1$ , (2) any difference in the effect of age of first implant ( $\beta_3 - \beta_2$ )  $a_1$ , and (3) the effect of the age of the second implant  $\beta_4 a_2$ . These individual components can be tested and interpreted based on the

**TABLE 2. Results of difference of means *t* tests for standard scores on PPVT-4, PLS-4, and CELF-4**

	Language Outcomes							
	Bilateral			Unilateral			Difference	
	n	Mean	SD	n	Mean	SD	Mean	<i>p</i>
PPVT	67	94.57	17.17	24	85.21	11.98	9.36	0.0004
CELF-4 Core	26	93.42	22.88	9	88.78	16.48	4.65	0.512
CELF-4 EL	26	95.00	23.22	9	86.22	14.09	8.78	0.184
CELF-4 RL	24	93.58	17.99	9	92.11	10.48	1.47	0.772
PLS-4 TL	41	89.59	21.36	15	78.87	19.25	10.72	0.076
PLS-4 AC	41	92.10	19.95	15	83.87	17.69	8.23	0.139
PLS-4 EL	41	88.49	20.26	15	77.13	19.68	11.35	0.061

AC, Auditory Comprehension; CELF-4, Clinical Evaluation of Language Fundamentals, fourth edition; EL, Expressive Communication; PLS-4, Preschool Language Scales, fourth edition; PPVT, Peabody Picture Vocabulary Test—fourth edition; RL, receptive language; TL, Total Language.

individual coefficients. The overall effect of bilateral CIs, however, may depend on the activation ages, so inference proceeded by computing a “bilateral activation window,” which was the range of ages within which the activation of a bilateral CI produced a significant and positive effect on the language outcome. For a given unilateral activation age  $a_1$ , the window was defined to be the range  $[a_1, a_2]$ , where  $a_2$  was the oldest bilateral activation age such that the 95% confidence interval for the overall bilateral effect  $y^{bi} - y^{uni}$  contained only positive values. The window was empty if bilateral CIs had no significant positive effects.

## RESULTS

### Bilateral / Unilateral Differences of Means

Table 2 reports the difference of means *t* tests for each of the seven language outcomes, allowing for unequal variances between the two groups. A significant difference ( $p = 0.004$ ) was found for the PPVT results, with children with bilateral CIs scoring an average of 9.36 points (10.98%) higher than children with unilateral CIs. Neither the PLS-4 nor CELF-4 mean scores revealed significant differences between children with bilateral and unilateral CIs without controlling for other variables.

### Vocabulary (PPVT)

Mean scores for both groups of children with unilateral and bilateral CIs were within 1 SD of the mean for typically developing children with normal hearing, although this was the case for the former group only by a narrow margin. As is commonly observed (Blamey et al. 2001; Sarant & Garrard 2013), variability in scores was high, with some children scoring within or above the average range, and others well below it.

Table 3 gives results of the regression to predict bilateral/unilateral vocabulary differences while controlling for the parenting, child, and family characteristics listed in Table 1. The language accumulation specification for the PPVT score revealed that the number of years spent with bilateral CIs was a highly significant ( $p < 0.001$ ) predictor, with its effect moderated by bilateral activation age ( $p = 0.033$ ). For example, a bilateral CI activated at 2.93 years of age (the mean activation age for this sample) was estimated to result in a significant ( $p = 0.004$ ) rate of language improvement of  $3.95 - 0.69 \times 2.93 = 1.93$  points (95% confidence interval =  $[0.62-3.23]$ , 2.10% of the unilateral average) for each year thereafter. The joint semipartial  $R^2$  of the

two bilateral variables was 0.09 of a total  $R^2$  of 0.51. The variables measuring the unilateral CI years were not found to be significant, being excluded by the AIC from the final specification.

Two of the parenting predictors were found to be significant. Parental involvement, as measured by the MFRS, was significant ( $p = 0.013$ ). A 1 SD increase (0.66) in the MFRS score predicted a  $0.66 \times 5.44 = 3.59$  point ( $[0.78-6.41]$ , 3.90%) increase in the PPVT outcome. Adult reading time was also a significant predictor ( $p = 0.048$ ), with an extra hour per week predicting an increase of  $60 \times 0.050 = 3.00$  points ( $[0.03-5.94]$ , 3.26%).

**TABLE 3. Summary of linear regression analysis results for PPVT standard scores**

Variable	PPVT	
	Coefficient	$R^2$
Constant	49.19*** (12.94)	
Bilateral years	3.95*** (1.07)	0.08
Bilateral Years $\times$ Age CI 2	-0.69* (0.32)	0.03
MFRS	5.44* (2.15)	0.04
Screen time	-0.42 (0.84)	0.01
Adult reading time	0.05* (0.02)	0.02
Birth order	-4.65** (1.41)	0.07
Intelligence quotient	0.12 (0.11)	0.01
Male	-4.64 (2.66)	0.02
Parent higher education	9.56** (2.88)	0.07
n	90	
$R^2$	0.51	
<i>F</i>	9.34***	

Coefficient standard errors in parentheses.

Significance at 5%, 1%, and 0.1% are denoted by \*, \*\*, and \*\*\*, respectively.

$R^2$  for each variable is the semipartial  $R^2$ , the amount by which the total  $R^2$  for the regression would decrease if that variable were omitted.

*F* is joint *F* test for overall significance of the regression.

CI, cochlear implant; MFRS, Moeller's Family Rating Scale; PPVT, Peabody Picture Vocabulary Test—fourth edition.

Children's birth order was found to be a highly significant predictor of vocabulary outcomes ( $p = 0.001$ ), and contributed 0.07 to the overall  $R^2$ . Each older sibling predicted a decreased PPVT score by 4.65 points ([1.85–7.45], 5.05%). The education level of the primary caregiver was a highly significant predictor of vocabulary outcomes ( $p = 0.001$ ), with the child of a tertiary qualified parent predicted to have a score of 9.56 points ([3.83–15.28], 10.38%) higher than otherwise.

### Language

Tables 4 to 6 give the results of the regressions to predict bilateral/unilateral language differences while controlling for the parenting, child, and family characteristics listed in Table 1.

### Language Outcomes for 5-Year-Old Children (PLS-4)

Group mean scores for the unilateral group of children on all three subtests of the PLS-4 were well below the average range for typically developing children, and there was great variability between children in outcomes.

Having bilateral CIs was not an individually significant predictor of outcomes in any of the PLS-4 regressions. The activation age of bilateral CIs, however, was a significant predictor of Total

Language and Auditory Comprehension scores ( $p = 0.002, 0.010$ ). The coefficients were negative, implying that earlier bilateral implantation predicted improved language outcomes; a reduction of 1 year in age at bilateral CI predicted an increase of 5.96 points ([2.31–9.61], 6.25%) on Total Language scores and 4.80 points ([1.20–8.39], 4.60%) on Auditory Comprehension scores. The effect of age at first CI followed a qualitatively similar pattern for the unilateral children for all three PLS subscales ( $p = 0.000, 0.000, 0.000$  for Total Language, Auditory Comprehension, and Expressive Language, respectively). For a child with a unilateral CI, a reduction of 1 year in age at implantation predicted increases of 13.82 points ([8.80–18.84], 19.94%), 11.37 points ([6.44–16.32], 17.10%), and 14.50 points ([9.62–19.39], 19.40%) on the Total Language, Auditory Comprehension, and Expressive Language scores, respectively. The activation age at first CI contributed most (0.22, 0.17, 0.25 for Total Language, Auditory Comprehension, and Expressive Language) to the overall  $R^2$  of each regression, while the significant contributions of the activation age at second CI were 0.08 (Total Language) and 0.06 (Auditory Comprehension).

The results for the joint inference based on the bilateral activation windows are given in Table 7. The results for the Total Language and Auditory Comprehension subscales show that bilateral CIs predicted significant language improvements by 5

**TABLE 4. Summary of linear regression analysis results for CELF-4 standard scores**

Variable	Core Language		Expressive Language		Receptive Language	
	Coefficient	$R^2$	Coefficient	$R^2$	Coefficient	$R^2$
Constant	-68.96 (37.02)		-70.23 (35.14)		-11.35 (24.40)	
Bilateral CI	58.32** (17.89)	0.20	61.19** (16.98)	0.22		
Age CI 1 (unilateral)					-13.91** (4.26)	0.17
Age CI 2	-11.36** (3.81)	0.17	-10.83** (3.61)	0.15	-4.11* (1.84)	0.08
MFRS	11.10* (4.84)	0.10	10.57* (4.59)	0.09	6.01 (3.35)	0.05
Birth order	-8.97* (4.06)	0.09	-10.83* (3.85)	0.13	-4.17 (2.85)	0.03
Intelligence quotient	0.89** (0.30)	0.16	0.96** (0.29)	0.19	0.92*** (0.21)	0.30
Birth weight	0.006 (0.01)	0.03	0.006 (0.00)	0.03	0.008* (0.00)	0.09
Male	-4.60 (6.54)	0.01	-5.48 (6.20)	0.01	-10.08* (4.66)	0.07
Age walked	-0.88 (0.90)	0.02	-1.20 (0.85)	0.03	-1.31 (0.64)	0.07
Fine motor problems	31.91* (11.39)	0.15	33.84** (10.81)	0.16	23.74** (7.08)	0.18
Family history (hearing loss)	8.77 (7.07)	0.03	10.01 (6.71)	0.04		
Family history (reading difficulties)	-11.45 (9.21)	0.03	-13.11 (8.74)	0.04	-8.61 (6.07)	0.03
Family history (speaking difficulties)	11.69 (9.72)	0.03	11.79 (9.22)	0.03		
N	35		35		33	
$R^2$	0.59		0.63		0.65	
F	2.63		3.17**		4.05**	

Significant at 5%, \* 1%, \*\* and 0.01%\*\*\* levels, respectively.

CELF-4, Clinical Evaluation of Language Fundamentals, fourth edition; CI, cochlear implant; MFRS, Moeller's Family Rating Scale.

**TABLE 5. Summary of linear regression analysis results for PLS-4 standard scores**

Variable	Total Language		Auditory Comprehension		Expressive Language	
	Coefficient	R <sup>2</sup>	Coefficient	R <sup>2</sup>	Coefficient	R <sup>2</sup>
Constant	98.34*** (19.53)		91.67*** (19.23)		107.14*** (18.88)	
Age CI 1 (bilateral)					-5.90 (3.70)	0.02
Age CI 1 (unilateral)	-13.82*** (2.49)	0.22	-11.38*** (2.45)	0.17	-14.50*** (2.42)	0.25
Age CI 2	-5.96** (1.81)	0.08	-4.80* (1.79)	0.06	-3.25 (2.58)	0.01
Screen time	-2.57* (1.21)	0.08	-1.88 (1.19)	0.07	-2.71* (1.17)	0.10
Adult reading time	0.10* (0.04)	0.05	0.09* (0.04)	0.05	0.09* (0.04)	0.05
Birth order	-8.39*** (1.99)	0.13	-8.13*** (1.96)	0.14	-7.18*** (1.93)	0.10
IQ	0.21 (0.15)	0.01	0.22 (0.15)	0.02	0.16 (0.15)	0.01
Male	-10.67** (3.95)	0.05	-9.55* (3.89)	0.05	-10.92** (3.81)	0.06
Age walked	-0.87 (0.43)	0.03	-0.59 (0.43)	0.02	-1.06* (0.42)	0.04
Family history (hearing loss)	14.21** (4.58)	0.07	13.55** (4.51)	0.07	11.92** (4.42)25	0.05
n	55		55		55	
R <sup>2</sup>	0.68		0.64		0.69	
F	10.63***		8.73***		9.87***	

Significant at 5%, \* 1%, \*\* and 0.01%\*\*\* levels, respectively.

CI, cochlear implant; IQ, intelligence quotient; PLS-4, Preschool Language Scales, fourth edition.

years of age, provided they were activated early enough. As with the CELF Receptive Language scores, the windows became wider as the age at first CI increased. The windows based on PLS-4 scores were narrower than those based on CELF-4 scores for the older children, implying that bilateral CI activation needs to occur early to give sufficient time to yield significant language improvement by 5 years of age. The absence of significant bilateral variables in the PLS-4 Expressive Language equation translated to an empty bilateral activation window—there was no evidence of improved Expressive Language scores for children with bilateral CIs at any activation age.

Two of the parenting predictors were found to be significant. The adult reading variable was significant in all three equations ( $p = 0.012, 0.014, 0.012$  for Total Language, Auditory Comprehension, and Expressive Language, respectively). The marginal effect on the Total Language score of an extra hour per week of adult time spent reading to the child was an increase of  $60 \times 0.096 = 5.76$  points ([1.33–10.17], 6.64%). The results of

the Auditory Comprehension and Expressive Language equations were similar. Screen time was found to have a significant effect on Total Language and Expressive Language scores ( $p = 0.039, 0.025$ ), with an extra hour of screen time per day predicting a decrease in Total Language scores of 2.57 points [0.13–5.00] and 2.71 points [0.36–5.06] for the Expressive score. A joint interpretation of these two parenting variables is that the predicted effect of a parenting intervention that reduces a child's screen time by half an hour every weekday, and substitutes with half an hour of adult reading time (an extra 150 min per week), would be to significantly ( $p = 0.005$ ) increase the Total Language score by  $150 \times 0.096 + 0.5 \times 2.57 = 15.66$  points ([5.01–26.30], 18.08%).

Two of the child characteristics were highly significant predictors of language outcomes on this test, and showed effects of a substantial magnitude. The effect of birth order was highly significant in each equation ( $p < 0.001$ ), with each older sibling predicting a decrease in Total Language, Auditory Comprehension, and

**TABLE 6. Bilateral activation age windows for CELF-4 standard scores**

First Implant Age	Core Language	Expressive Language	Receptive Language
0.5	3.75	4.33	1.04
1.0	3.75	4.33	2.09
1.5	3.75	4.33	3.13
2.0	3.75	4.33	4.18

CELF-4, Clinical Evaluation of Language Fundamentals, fourth edition.

**TABLE 7. Bilateral activation age windows for PLS-4 standard scores**

First Implant Age	Total Language	Auditory Comprehension	Expressive Language
0.5	0.78	0.73	—
1.0	1.55	1.46	—
1.5	2.33	2.20	—
2.0	3.10	2.93	—

PLS-4, Preschool Language Scales, fourth edition.

Expressive Language scores of 8.39 points [4.39–12.40], 8.13 points [4.19–12.08] and 7.18 points [3.29–11.06], respectively. Birth order also made a substantial contribution to the  $R^2$  of all three equations (0.13, 0.14, and 0.10 to Total Language, Auditory Comprehension, and Expressive Language, respectively). Sex was also significant in each equation ( $p = 0.010, 0.018, 0.006$ ) with boys predicted to score 10.67 points [2.71–18.63], 9.55 points [1.71–17.38], and 10.92 points [3.24–18.60] lower on the Total Language, Auditory Comprehension, and Expressive Language scores, holding all other characteristics constant.

A family history of hearing loss was the only family history variable to be found significant in any of the equations. It was highly significant for all three subscales ( $p = 0.003$ , Total Language; 0.004, Auditory Comprehension; 0.010, Expressive Language) and had a positive effect on scores. For example, having a family history of hearing loss predicted a 14.21-point ([4.98–23.44], 16.39%) higher Total Language score, relative to a child with all the same characteristics but with no family history of hearing loss.

### Language Outcomes for 8-Year-Old Children (CELF-4)

Language outcomes on both measures for the children in this study again showed great variability, as has been reported in previous studies using these measures (Spencer et al. 2003; Tobey et al. 2013). Group mean scores for all of the CELF-4 subtests were within 1 SD of the means for typically developing children, but there was great variability in results between children.

Bilateral CIs were a significant predictor for both the Core Language and Expressive language subscales of the PLS-4 ( $p = 0.004, 0.002$ , respectively), although not for the Receptive Language subscale. The magnitude of the effect was moderated by the age at which the second CI was activated ( $p = 0.007, 0.007$ ). The predicted effect of a bilateral CI activation at the mean activation age of 2.94 years was  $58.31 - 11.36 \times 2.94 = 24.91$  points [6.42–43.41] on the CELF Core Language subscale, or 28.06% higher than a unilateral child with all the same other characteristics. The same calculation for the CELF-4 Expressive Language subscale showed a 34% higher result (29.34 points, [11.78–46.89]) for a child with bilateral CIs implanted at the average age. For a child with bilateral CIs, each additional year of delay of implantation of the bilateral CI predicted a decrease in CELF Core Language and Expressive Language scores of 11.36 points ([3.47–19.25], 12.16%) and 10.83 points ([3.34–18.32], 11.40%), respectively.

The CELF-4 regressions accounted for between 59 to 65% of the variability in outcomes on this measure. The contributions of having bilateral CIs and the bilateral activation age to the predictive ability of the regressions are given by the semipartial  $R^2$  statistics in Table 3. These two variables were, by definition, highly collinear (correlation coefficient of 0.74), measuring two aspects of bilateral CIs. The overall predictive ability of bilateral CIs was therefore best evaluated using the joint semipartial  $R^2$  of the bilateral indicator and CI age, which were 0.20 (Core Language) and 0.22 (Expressive Language). That is, the overall  $R^2$  of 0.59 and 0.63 for the Core and Expressive regressions would decrease by 0.20 and 0.22, respectively, if the two bilateral CI variables were omitted.

The bilateral CI indicator was not selected in the CELF Receptive Language equation, but the child's age at bilateral implantation was selected, and was statistically significant ( $p = 0.036$ ), indicating that bilateral implantation is predictive of these outcomes, with a unique contribution of 8% of the

variance in outcomes. A delay of 1 year in bilateral implantation predicted a 4.11 point ([0.29–7.93], 4.39%) decrease in the CELF Receptive Language score. Child age at first CI was found to be significant ( $p = 0.004$ ) for children with unilateral CIs, but not for children with bilateral CIs. A delay of 1 year in first CI for a child with a unilateral CI predicted a 13.91 point ([5.07–22.75], 15.10%) decrease in CELF-4 Receptive Language score.

These results suggest that bilateral CIs had a statistically significant effect on CELF-4 language scores of the 8-year-old children in this study, the magnitude of which varied with the ages at which children received their CIs. The above-outlined prediction comparison approach provides a readily interpretable summary of the predicted effectiveness of bilateral CIs.

Table 6 presents results for several first implant activation ages. For the Core Language and Expressive Language subscale results, bilateral CIs were found to have a significant and positive effect, provided they were activated before ages of 3.75 and 4.33 years, respectively.

The results for the child, parent, and family characteristics were similar for the Core and Expressive Language subscales. Birth order was significant in both cases ( $p = 0.038, 0.010$ ), with each extra older sibling predicting decreases of 8.97 points ([0.56, 17.39], 9.73%) and 10.83 points ([2.84, 18.82], 11.68%), respectively. Parent involvement was also significant for both measures ( $p = 0.032, 0.031$ ). A 1 SD increase in parent involvement predicted a 7.29 point ([0.70–13.87], 7.94%) and 6.94 point ([0.69–13.19], 7.52%) increase in the Core Language and Expressive Language subscales, respectively. Children's cognitive ability was significant for all three CELF-4 language subscales ( $p = 0.008, 0.003, 0.001$  for Core Language, Expressive Language, and Receptive Language, respectively), with an extra 10 points of cognitive ability (slightly less than 1 SD), holding all else constant, predicting increases of 8.86 points ([2.57–15.16], 9.61%), 9.63 points ([3.66–15.60], 10.38%), and 9.20 points ([4.82–13.59], 9.66%), respectively. Cognitive ability also made an important contribution to the predictive power of the regressions, especially for the Receptive Language subscale, for which cognitive ability contributed 0.30 to the overall  $R^2$  of 0.65. The presence of fine motor problems was significant ( $p = 0.010, 0.005, 0.003$ ), predicting higher scores of 31.91 points ([8.29–55.52], 34.61%), 33.84 points ([11.43–56.25], 36.48%), and 23.74 points ([9.06–38.41], 25.48%) for the Core, Expressive, and Receptive subscales, respectively.

## DISCUSSION

### Vocabulary Outcomes

The results of this study showed that children with bilateral CIs achieved significantly better vocabulary outcomes than did comparable children with unilateral CIs. This significant finding contrasts with those of the only other two studies, whose statistical power was likely reduced by reliance on samples containing relatively few children with sufficient bilateral CI experience to develop their listening skills and subsequent language ability.

### Language Outcomes

In terms of the comparative performance of children with unilateral and bilateral CIs, the language results were not as unequivocal as the vocabulary outcomes, with no significant differences between the mean scores for unilateral and bilateral

CI children for any of the CELF-4 and PLS-4 subtests. However, once other influential factors were controlled for in the regression analysis, bilateral CI use was found to predict significantly faster rates of language development in both of the PLS-4 and CELF-4 tests.

The weaker effect of bilateral CI use on the simple difference of means for the language measures is likely to reflect both the increased complexity of language skills required, the substantial length of time required to master these, and the fact that the sample sizes for both these measures were smaller than for the vocabulary measure, where results for both the 5- and 8-year-old children were included. An illustration of the need for more time to develop higher-level language skills can be seen in the Core Language and Expressive Language CELF-4 regression results, where bilateral CIs were found to have a significant and positive effect on scores, provided they were activated before 3.75 and 4.33 years of age, respectively. Activation of bilateral CIs after these ages may not have allowed sufficient time to produce a significant benefit relative to the unilateral implant for the 8-year-old participants of this study.

### **Effect of Bilateral Hearing on Vocabulary and Language Outcomes**

It was speculated earlier that the perceptual benefits reported for children with bilateral CIs such as improved speech perception in both quiet and noisy listening conditions (Scherf et al. 2007; Zeitler et al. 2008), improved sound localization ability for some children (e.g., Lovett et al. 2010), and reduced listening effort (and therefore reduced tiredness and a greater ability to concentrate; Hughes & Galvin 2013) may facilitate a greater ability to access the spoken language of others and to learn from these increased opportunities. Despite the large variability in language outcomes, a moderate sample size, and multiple factors that influence these outcomes, significantly faster rates of vocabulary and language development were found for the children with bilateral CIs in this study. This finding suggests that the perceptual benefits of bilateral hearing through two CIs conferred a significant advantage, in terms of learning, to these children. It must be emphasized, however, that it is not known whether the improved results for children with bilateral CIs are due simply to having bilateral auditory input, or whether they are the result of true binaural processing. This study was not designed to address this question, but it will be an important area for future investigation.

### **Predictors of Outcomes**

**Age at First CI** • Age at CI has been documented extensively a significant predictor of child language outcomes (Connor et al. 2006; Schorr et al. 2008; Geers et al. 2009), and marks the beginning of functional auditory input to the auditory cortex and the subsequent development of the auditory processing abilities that facilitate spoken language development. A surprising finding of this study was that neither age at first CI nor duration of unilateral CI use were significant predictors of PPVT scores. This does not imply that a unilateral implant is ineffective, but that there was insufficient information in this sample (with only 24 children with unilateral CIs) to precisely estimate the rate of vocabulary accumulation. It is also worth noting that for many children in this sample, 4 to 7 years had elapsed since their first CI implantation. Although several studies have

reported better language outcomes with very early implantation (Dettman et al. 2007; Nicholas & Geers 2007), follow-up in these studies occurred when the children had limited experience with their CIs (Geers 2004). More recent findings have been mixed; one study has since reported that the effect of age at first implant reduces over time (Hay-McCutcheon et al. 2008), while a very recent study has shown that age at first CI is still a strong predictor of outcomes after approximately 8 years of CI use (Geers & Nicholas 2013).

Child age at first CI had expected effects for the PLS-4 and CELF-4 tests. Earlier age at first CI was predictive of a large increase (17 to 19% for each year) in PLS-4 scores across all subscales, and explained the greatest amount of variance in outcomes on this measure (22%). For the 8-year-old children, age at first CI was found to be a significant predictor of outcomes on the CELF-4 Receptive Language subtest only for children with unilateral CIs.

**Age at Second CI** • As far as the authors are aware, the effect of age at second CI on language outcomes has not been examined in any other studies. Here it was found to have an important effect, with earlier ages of second implantation predicting significant vocabulary and language improvements for both 5- and 8-year olds. This effect was quantified in different ways for the different tests.

For the PPVT test, vocabulary ability was shown to accumulate more quickly for children with bilateral CI compared with unilateral CI, but the magnitude of the difference was reduced with each year of delay before second implantation. Therefore early bilateral implantation would be recommended for maximum vocabulary benefit.

A bilateral CI predicted statistically significant language improvements on all PLS-4 and CELF-4 subtests except the CELF-4 Expressive, and the extent of these improvements was influenced by various interactions between the ages of first and second implant. These results are summarized in the bilateral activation age windows. For each subtest, the bilateral activation age window provides the range of ages within which the activation of a bilateral CI predicts a statistically significant advantage over a unilateral CI. That these windows exist for all but the CELF-4 Expressive subtest demonstrates the diverse benefits of the bilateral CI for both 5- and 8-year olds.

To illustrate, activation windows with fixed upper age limits were found for the CELF-4 Core (3.75 years) and Expressive (4.33 years) subtests, implying that bilateral implantation before these ages was sufficient to produce significant gains by 8 years of age, regardless of age of first CI (at least within the range in this sample). In all cases, the magnitude of these language gains decreased as age of bilateral CI increased. For example, a child receiving a bilateral CI at 2 years of age is predicted to score 35.60 points higher on the CELF-4 Core test relative to a unilateral CI child, but if the activation age were 3.75 years then this gain reduces to 15.72 points. Therefore, early bilateral implantation would be recommended for maximum language benefit.

Some bilateral activation age windows depended on the age of first CI. For example, the upper age limit for the CELF-4 Receptive Language subtest was found to vary with the age of first CI. If a child received their first implant at 1 year of age, then a subsequent bilateral implant received before 2.09 years of age predicted a significant and positive effect on the Receptive Language subscale score. If the first

CI were received at 2 years of age, then the bilateral activation window extended to 4.18 years of age. This longer window for children with later age at first CI reflects the considerably larger implant age effect for the first CI relative to the second—a child first implanted at 2 years of age is acquiring hearing and language from a point substantially (predicted 15.10%) behind a child first implanted at 1 year of age; a deficiency that the bilateral implant can significantly reduce. Note, however, that the predicted language scores are always higher for children with lower implant ages, so this longer bilateral activation window does not imply better language scores for children implanted later, rather that a bilateral CI has the potential to help catch up some (not all) of the delay due to a delayed first implant.

Two of the three PLS-4 bilateral age activation windows were nonempty (Total Language and Auditory Comprehension), implying that a bilateral child will already be demonstrating improved language outcomes by 5 years of age, provided they receive their second implant sufficiently early. These activation windows were shorter than those for the CELF-4, implying that, compared with the 8-year olds, earlier bilateral implantation is required for bilateral CI children to show significant language gains relative to unilateral CI children by 5 years of age.

Overall, these age-at-CI results underscore the importance of having the shortest possible time between CIs for children who are sequentially implanted, and support the findings of physiological studies of abnormalities in spatial patterns of cortical brain activity in children implanted sequentially after a long time (Gordon et al. 2010), and theories of reduced central auditory plasticity at older ages (Sharma et al. 2002, 2005).

### Parenting Style

The effect of parental involvement on both vocabulary and language development was found to be strongly predictive of better outcomes, as has been documented previously (Moeller 2000; Sarant et al. 2009). The amount of time adults in the family spent reading to their child, and the amount of time children spent watching a screen each week were identified as new factors that significantly affected both vocabulary and language outcomes. An example of a joint interpretation of these two parenting variables is that the predicted effect of a parenting intervention that reduced a child's screen time by 30 min each weekday and substituted this with 30 min of adult-to-child reading time (150 extra min/week) would be a significant increase ( $p = 0.005$ ) in the PLS-4 Total Language score by 15.66 points (18.08%;  $150 \times 0.096 + 0.5 \times 2.57$ ). This large predicted increase in scores demonstrates how effective relatively small changes in parenting can be in facilitating significant changes in children's language development.

### Child Characteristics

Birth order was found to have a significant effect on PPVT scores, with each older sibling decreasing scores by approximately 5%. It also exerted a significant negative effect on language scores for five of the six subscales of both language measures. This has been found to be the case in other studies of children with normal hearing, and likely relates to the demands on parents' time and resources with increasing numbers of children (Hoff Ginsberg 1998; Nelson et al. 2006).

Although it has also been found that older siblings can enrich aspects of younger children's language development, it has also been reported that older children monopolize more parent-child conversations than their younger siblings (Wellen 1985; Oshima-Takane et al. 1996). Children with significant hearing loss are likely to miss out on the enriching aspects of "over-hearing" conversations due to their auditory limitations, and are also likely to be disadvantaged by the reduced number of opportunities to talk directly with their parents and learn language in this manner.

In common with other reports for both children with normal hearing (Fenson et al. 2000) and children with hearing loss (Moog & Geers 2003; Geers et al. 2009), male sex was significantly predictive of poorer language PLS-4 outcomes for 5-year-old children. However, this effect was no longer evident for the 8-year-old children on the CELF-4 results. This finding fits with reports that the gap in language ability found between boys and girls in the early life closes with increasing age (Gadde & Crockett 1975; Ely 2005).

Nonverbal cognitive ability also significantly affected language outcomes for the 8-year-old children in this study. Cognitive ability has been identified as a primary predictive factor of language development in children with hearing loss, accounting for large proportions of the variance in outcomes in many studies (e.g., Holt & Kirk 2005; Geers et al. 2009; Sarant et al. 2010).

An unexpected and puzzling finding was that 8-year-old children who were reported by their parents to have had difficulties with fine motor development showed significantly better language development than children whose fine motor development had been viewed by their parents as normal. This finding contradicts those of other large studies that have shown a strong synchrony between fine motor development and language development (Bavin et al. 2008; Taylor 2010). Closer examination of the data showed that despite 14 parent reports of difficulty with fine motor skills, only 4 of these children were receiving assistance with the development of these skills. It is possible that the concern levels of these parents were inconsistent with the degree of delay the children were experiencing. While this explanation could account for the lack of a negative effect of this factor on language development, it does not account for the positive effect found. It could be that this predictor is a proxy for another factor that may be identified with a larger sample size and further research in the future.

### Family Background

As has been reported previously (Dollaghan et al. 1999; Sarant et al. 2009; Niparko et al. 2010), there was a significant effect of maternal education level (which can be considered a reasonable proxy for socioeconomic status) on vocabulary scores, with children of tertiary-educated primary caregivers achieving significantly (approximately 10%) higher scores.

Having a family history of hearing loss also had a surprisingly large positive effect on language outcomes for 5-year-old children. This may reflect the value of having a family member with an understanding of the challenges of hearing loss and knowledge of strategies to access assistance in the early years of parenting a child with a hearing loss. While family histories of spoken language or reading difficulties have historically been reported as risk factors for language development (Nelson et al. 2006), the authors are unaware of any evidence regarding

family history of hearing loss. It is interesting that this factor did not influence language outcomes for the 8-year-old children, where a high level of parent involvement accounted for 10% of the variance in outcomes. It may be that by the time children are 8 years old, parents have developed sophisticated parenting practices that exceed the effect of the knowledge possessed by parents with a family history of hearing loss, or simply that different predictors were found for these different groups of children.

### Study Limitations

As this was a cross-sectional study, its findings may not be representative of other populations of children with CIs. However, given the fact that the study cohort was recruited from the majority of the country's highly populated areas, and the fact that the recruitment rate was comparable with that of epidemiological studies, these results should be reasonably representative of Australian children with CIs. A further potential limitation of this study was the imbalance in numbers between children with unilateral and bilateral CIs, and the resulting limited representation of children with unilateral CIs. With larger numbers of children in the future, it is hoped that this limitation may be addressed.

### CONCLUSIONS

Overall, bilateral CIs contributed to significantly better language outcomes for the children in this study, with improvements moderated by children's ages at second CI. Earlier implantation, both unilateral and bilateral, was found to be beneficial to language outcomes. With larger sample sizes in the future, possible nonlinearities in implant age effects, such as whether there is a discontinuity between simultaneous and sequential implantation could also be investigated. Examining whether there is an asymptote to the negative effect of delaying implantation, in addition to determining a window of implant ages of maximum effectiveness, would provide valuable clinical information.

The influence of parenting style on language outcomes for children was also extremely important. This study identified parenting practices, not previously investigated in studies of language outcomes in children, that exerted a significant influence on language outcomes. All three parenting variables examined in this study were found (in different regressions) to have statistically and practically significant effects on language development. This could be a particularly important consideration for parents of hearing-impaired children with several older siblings, because the presence of these children may slow language development in a younger child with a CI.

Despite the large reported variability in language outcomes, the present study was able to account for a significant proportion of the variance in language outcomes (up to 69%), which was higher than that in many studies.

The findings of this study add significantly to the body of knowledge regarding outcomes for children with bilateral versus unilateral CIs, and also to that of factors that facilitate improved spoken language outcomes for children with CIs. It is imperative that this knowledge is accessible to, and is used by parents, clinicians, and governments in making evidence-based decisions that are translated into clinical practice and governmental

policy development. In this way, it would be possible to most effectively assist the many children around the world for whom bilateral CIs can contribute to closing the gap, relative to those with normal hearing, in terms of language development, and hopefully, subsequent quality of life.

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