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**Author/s:**

Sarant, J;Harris, D;Bennet, L;Bant, S

**Title:**

Bilateral versus unilateral cochlear implants in children: A study of spoken language outcomes

**Date:**

2014-01-01

**Citation:**

Sarant, J., Harris, D., Bennet, L. & Bant, S. (2014). Bilateral versus unilateral cochlear implants in children: A study of spoken language outcomes. *Ear and Hearing*, 35 (4), pp.396-409. <https://doi.org/10.1097/AUD.0000000000000022>.

**Persistent Link:**

<https://hdl.handle.net/11343/59600>

## 1 **Abstract**

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

9 **Method:** The receptive vocabulary and language development of 91 children was assessed  
10 when they were aged either five or eight years old using the Peabody Picture Vocabulary Test  
11 (Fourth Edition), and either the Preschool Language Scales (Fourth Edition) or the Clinical  
12 Evaluation of Language Fundamentals (Fourth Edition) respectively. Cognitive ability,  
13 parent involvement in children's intervention or education programs, and family reading  
14 habits were also evaluated. Language outcomes were examined using linear regression  
15 analyses. The influence of elements of parenting style, child characteristics and family  
16 background as predictors of outcomes were examined.

17 **Results:** Children using bilateral CIs achieved significantly better vocabulary outcomes and  
18 significantly higher scores on the Core and Expressive Language subscales of the CELF-4  
19 than did comparable unilateral children. Scores on the PLS-4 did not differ significantly  
20 between children with unilateral and bilateral CIs. Bilateral CI use was found to predict  
21 significantly faster rates of vocabulary and language development than unilateral CI use; the  
22 magnitude of this effect was moderated by child age at activation of the bilateral CI. In terms  
23 of parenting style, high levels of parental involvement, low amounts of screen time, and more  
24 time spent by adults reading to children facilitated significantly better vocabulary and  
25 language outcomes. In terms of child characteristics, higher cognitive ability and female

26 gender were predictive of significantly better language outcomes. When family background  
27 factors were examined, having tertiary-educated primary caregivers and a family history of  
28 hearing loss were significantly predictive of better outcomes. Birth order was also found to  
29 have a significant negative effect on both vocabulary and language outcomes, with each older  
30 sibling predicting between a 5-10% decrease in scores.

31 **Conclusions:** Children with bilateral CIs achieved significantly better vocabulary outcomes,  
32 and eight-year-old children with bilateral CIs had significantly better language outcomes than  
33 did children with unilateral CIs. These improvements were moderated by children's ages at  
34 both first and second CIs. The outcomes were also significantly predicted by a number of  
35 factors related to parenting, child characteristics, and family background. Fifty-one percent  
36 of the variance in vocabulary outcomes and between 59-69% of the variance in language  
37 outcomes was predicted by the regression models.

38

## 39 INTRODUCTION

40 Although many children with unilateral cochlear implants (CIs) have excellent speech  
41 perception abilities in a controlled listening environment such as a quiet room or sound proof  
42 booth (Leigh et al. 2008; Sarant et al. 2001) these environments do not represent listening  
43 conditions in the real world. In more difficult listening conditions, such as noisy classrooms  
44 or playgrounds and the family home, children with a unilateral CI and a severe-profound or  
45 profound hearing loss in the contralateral ear will experience significant difficulties, which  
46 will reduce the amount and quality of speech they are exposed to. Understanding speech that  
47 is soft, speech in background noise, and locating sound sources such as speakers in a group  
48 conversation are examples of such difficulties. With these perceptual limitations, it is less  
49 likely that children with unilateral CIs will have the ability to learn incidentally through  
50 'overhearing', as do children with normal hearing, which limits their acquisition of language,

51 world knowledge and social skills. Although many children with CIs have been able to  
52 develop spoken language and other skills that would not have been possible with  
53 conventional hearing aids, it has been well-documented for many years through to the present  
54 time that many children with unilateral CIs show delays in the development of language  
55 (Blamey et al. 2001; Geers 2002, Nittrouer et al. 2012), speech production (Connor et al.  
56 2006; Spencer et al. 2011; Tobey et al. 2003), literacy (Crosson & Geers 2001; Geers &  
57 Hayes 2011; Marschark et al. 2007), academic (Mukari et al. 2007; Spencer et al. 2003) and  
58 social skills (Bat-Chava et al.2005; Hintermair 2006). Although a number of children with  
59 unilateral CIs have been able to achieve age-appropriate development in many of these areas  
60 (Duchesne et al. 2009; Percy-Smith et al. 2008; Spencer et al. 2004), for a significant number  
61 of these children, developmental delays have been maintained or increased through to  
62 adulthood (Geers et al., 2008; Moeller et al., 2007; Mukari et al., 2007; Uziel et al., 2007).  
63 For this reason, the efficacy of bilateral CIs is being investigated, and bilateral cochlear  
64 implantation is becoming the standard of care for children with severe-profound hearing loss  
65 in developed countries around the world (National Institute on Deafness and other  
66 Communication Disorders, 2011).

67

### 68 **Perceptual Benefits of Bilateral Cochlear Implants**

69       Bilateral CIs offer additional benefits over a unilateral CI through the mechanisms of  
70 binaural redundancy (speech perception is improved with two ears, as the brain has two  
71 opportunities to process the signal), binaural summation (the signal when combined from two  
72 ears is slightly louder than from one ear) and the head-shadow effect (the head acts as a  
73 physical barrier to the sound, such that the signal will be softer at the ear that is furthest from  
74 the sound source). The benefits of bilateral CIs for speech perception in children have been  
75 evaluated in both noisy and quiet listening conditions. In noise, many studies have reported a

76 significant improvement in children's abilities to perceive speech (Galvin et al. 2008;  
77 Johnston et al. 2009; Litovsky et al. 2006; Lovett et al. 2010). In quiet listening conditions,  
78 improved speech perception has also been reported (Scherf et al. 2007; Zeitler et al. 2008).  
79 Advantages of bilateral CIs for sound localisation are not quite as clear, with some children  
80 reported to localise sound well (Litovsky et al. 2006; Lovett et al. 2010), and others  
81 demonstrating more limited localisation ability (ie. left-right lateralization, rather than true  
82 localization (Galvin et al. 2008; Grieco-Calub & Litovsky 2010). Many other children,  
83 particularly older children, have shown no ability to localise sound (Galvin et al. 2007).  
84 Further benefits have also been documented, with parents in some studies frequently  
85 reporting superior performance using bilateral CIs in everyday life, in situations such as  
86 group conversations, background noise and hearing at a distance (Galvin et al. (submitted);  
87 Sparreboom et al. 2012). There is also objective evidence that for some children with  
88 bilateral CIs, listening effort is reduced, suggesting that more attention can then be paid to the  
89 learning process (Hughes & Galvin in press). Despite the evidence of benefit for children  
90 from bilateral CIs cited above, it has yet to be determined whether these perceptual benefits  
91 facilitate significantly better broader outcomes in children with bilateral CIs, and if so, to  
92 quantify the degree of benefit received by children based on factors such as age at second  
93 implant.

94

### 95 **The Effect of Bilateral CIs on Language Outcomes**

96 Until recently, most of the research on outcomes for children with bilateral CIs was  
97 focussed on speech perception and sound localization benefits. Evidence regarding whether  
98 bilateral CIs significantly improve broader outcomes such as language, literacy, academic  
99 and social skills and overall quality of life is lacking, particularly regarding longer-term  
100 outcomes (Johnston et al. 2009; Sparreboom et al. 2010). The results of the few earlier

101 studies comparing language outcomes for children with unilateral and bilateral CIs did not  
102 show a significant benefit to language development from bilateral implantation. One of the  
103 first studies to investigate the effect of bilateral CIs on vocabulary, receptive and expressive  
104 language of children assessed at age three and a half years (15 unilateral, 26 bilateral)  
105 concluded that the reported perceptual benefits from bilateral CI use “may not extend to  
106 generative language”(Nittrouer & Chappman 2009). A further study of language outcomes in  
107 preschool-aged children implanted by age five years (60 unilateral, 31 bilateral; average age  
108 2-5 years) reported similarly that children with bilateral CIs did not receive a significant  
109 benefit to either receptive or expressive language development over children with unilateral  
110 CIs (Niparko et al. 2010). It was noted that this outcome may have been a result of the brief  
111 period of time that had elapsed between implantation of the second CI and follow up. A third  
112 study of vocabulary and language development in children who had just completed preschool  
113 (13 unilateral, 14 bilateral) and were aged up to six years also reported that having bilateral  
114 CIs had no effect on language outcomes (Nittrouer et al. 2012). In considering the results of  
115 these three studies, it is worth noting that most of the children were of preschool age and  
116 many had sequential CIs, and therefore had not had a long time to use their bilateral hearing  
117 to develop language. Two of the three studies also had relatively small sample sizes, which  
118 makes detecting a significant difference in performance (if it exists) difficult, given the large  
119 variance in language outcomes commonly reported (Connor et al. 2006; Sarant et al. 2009;  
120 Spencer et al. 2003).

121 Two recent reports from the same population of children contradict the results of the  
122 first three studies. A retrospective study of 288 children implanted by age five years  
123 examined language outcomes each year over three years post-CI for up to 29 children with  
124 bilateral CIs, compared with up to 85 children with unilateral CIs, and up to 62 children with  
125 a CI and a hearing aid (Boons et al. 2012a). It was concluded that contralateral stimulation

126 (with bilateral CIs or unilateral CI plus hearing aid) contributed to significantly improved  
127 language outcomes. An unspecified post-hoc analysis separated the bilateral CI and hearing  
128 aid effects, with the finding that bilateral CIs led to better outcomes than unilateral CIs, and  
129 also than bimodal hearing (CI plus hearing aid). This study differed from two previous  
130 studies in that the children were slightly older (up to age six years), although the number of  
131 children with bilateral CIs was not greater. A limitation of the study was that participating  
132 children used different CIs (Cochlear Ltd. or Advanced Bionics), which have recently been  
133 reported to give significantly different perceptual results (Lazard et al. 2012) and could  
134 therefore have affected language outcomes. Although the effect of child age at first CI was  
135 considered, with children implanted before the age of two years performing significantly  
136 better on all measures, the effect of age at second CI was not investigated.

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

144

### 145 **The contribution of this study**

146 Evidence of the impact of bilateral implantation on language outcomes is vital if  
147 evidence-based pre-operative recommendations are to be made to parents considering CIs for  
148 their children. This evidence can also be used by governments around the world to make  
149 policy decisions on whether to fund bilateral implantation in children. The current evidence,  
150 as described above, is limited and shows mixed findings. There is therefore a need for further

151 research in this area, particularly with regard to longer-term outcomes, given most studies to  
152 date have involved children of preschool age with limited CI experience.

153         The present study offers a further comparison of vocabulary and language outcomes  
154 in children using unilateral and bilateral CIs. It has the advantages of being prospective, and  
155 including older children than in previous studies, thus offering some insight into longer-term  
156 outcomes. It includes a moderate sample size of 91 children, with a greater number of  
157 children with bilateral CIs than in previous studies. Although the current study has a cross-  
158 sectional design, eighty-four percent of the country's population was located in the area from  
159 which the study sample was recruited. Of the eligible children in this area, 51.6% were  
160 recruited to the study, consistent with reported recruitment rates in the last decade for  
161 epidemiological studies (Galea & Tracy, 2007). The sample can therefore be considered to  
162 be reasonably representative of Australian children with CIs. The current study also expands  
163 upon previous research in that the effects of age at CI on language outcomes for both the first  
164 and second CI are evaluated. The effects of some parenting practices (family reading habits  
165 and child screen time) that have not previously been investigated in any studies of language  
166 outcomes in children with CIs and their relationships to language outcomes are also  
167 examined. Finally, the proportion of variance in language outcomes accounted for in the  
168 current study (up to 69%) is higher than in most previous studies.

169

## 170 **Objectives**

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

174

## 175 **MATERIALS AND METHODS**

## 176 **Participants**

177           Ninety one children aged 5 to 8 years were recruited from three cochlear implant  
178 clinics and three early intervention centers in four states of the country, accounting for most  
179 of the country's pediatric CI-related service organisations and major intervention centers.  
180 Eighty-four percent of the country's population is located in the area from which the study  
181 sample was recruited. Of the eligible children in this area, 51.6% were recruited to the study.  
182 This figure is consistent with reported recruitment rates in the last decade for epidemiological  
183 studies (Galea & Tracy 2007). Given the fact that the study cohort was recruited from 84%  
184 of the country's population, with a recruitment rate similar to recently reported recruitment  
185 rates for epidemiological studies, these results can be considered to be reasonably  
186 representative of Australian children with cochlear implants.

187           The study cohort consisted of 44 boys and 47 girls. All children were implanted early  
188 (first CI by age 3.5 years and second CI (if bilaterally implanted) by age 6 years), spoke  
189 English as their primary language, and had normal cognitive abilities. The age at CI criteria  
190 were chosen based on physiological studies that suggest that in the absence of normal  
191 auditory stimulation there is a period of about 3.5 years during which the central auditory  
192 system retains its maximum plasticity, and that this can extend in some children up to the age  
193 of approximately 6-7 years, after which it is significantly reduced (Sharma et al. 2005;  
194 Sharma et al. 2002). Of the 91 children, 67 used bilateral cochlear implants and 24 used a  
195 unilateral cochlear implant. All children used a Cochlear Ltd. CI, with ACE speech  
196 processing strategy. With the exception of 7 participants, for whom information regarding  
197 the number of active electrodes was unavailable at the time of writing, all children had  
198 between 19-22 active electrodes in their arrays. Twelve children had been re-implanted.  
199 Mean lengths of device use at the time of language assessment were 5.20 years (SD=1.79) for  
200 the bilateral group and 4.55 years (SD= 2.04) for the unilateral group. Hearing aid use, both

201 prior to and after, CII (if applicable) for all children was documented through parent  
202 interviews (see Table 1).

203

#### 204 **Demographic Measures**

205 Table 1 provides demographic information. Of the 91 children, 38 presented with a  
206 family history of hearing loss, although only two children had deaf parents. Of these 38  
207 children, 25 had a genetic cause of hearing loss, one had a viral cause and the etiology for 12  
208 children was unknown. Thirty five of the children had a hearing loss of a genetic origin and  
209 42 had a hearing loss of an unknown cause. The remaining 14 children presented with  
210 etiologies resulting from viral causes or medical complications at birth. Further information  
211 about child development was collected relating to birth order, birth weight, age at which  
212 children first walked and history of concerns with fine motor skill development. Only three  
213 children had a diagnosed additional disability that may have impacted on their ability to learn  
214 language. The communication mode for all families was primarily spoken language, with  
215 two children using  
216 supplementary sign to communicate with immediate family members with a profound  
217 hearing loss.

218

#### 219 **Procedure**

220 The children in this study were part of a wider study examining outcomes for children  
221 with cochlear implants. In accordance with the wider project's protocol, the children were  
222 assessed at five and eight years of age. To ensure that children were entered into the  
223 analyses only once, the most recently collected data for each child was entered. Of the  
224 bilateral group, 41 of the children were assessed at 5 years and 26 were assessed at 8 years.  
225 Of the unilateral group, 15 were assessed at 5 years and 9 were assessed at 8 years. For each

226 assessment point, language outcomes were measured using the standardized language tests  
227 described in the Instruments section by speech language pathologists. The cognitive ability  
228 of all children was assessed at age five years by an educational psychologist using either the  
229 Wechsler Non-Verbal Scale of Ability (86 children; WNV; Wechsler & Naglieri 2006) or the  
230 Wechsler Preschool and Primary Scale of Intelligence – Third Edition (5 children; WPPSI-  
231 III; Wechsler 2002). Normal cognitive ability was defined as a Performance Scale score of 80  
232 or more.

233 A questionnaire was designed specifically for this study to form a general picture of  
234 children's reading habits (see Appendix 1). Part of the questionnaire was based on items used in  
235 the study *Growing Up In Australia: The Longitudinal Study of Australian Children*  
236 (Australian Institute of Family Studies, 2012). In addition, questions specific to the aims of the  
237 present study were developed. The questionnaire was administered using a web-based form in  
238 the first instance. Where no response was received within a pre-determined timeframe,  
239 questionnaires were sent via mail or filled out during a telephone interview with a member of  
240 the research team.

241 Parental involvement in each child's intervention program was assessed using the  
242 Moeller's Family Rating Scale (MFRS; Moeller 2000). Whether or not children had been  
243 slow in fine motor skill development, whether parents were tertiary educated, and whether  
244 there was a family history of hearing loss, difficulty learning to read or learning to speak  
245 were documented. All of this demographic information was obtained through a telephone  
246 interview with the primary caregiver.

247

## 248 **Instruments**

249 **Peabody Picture Vocabulary Test – Fourth Edition (PPVT-4; Australian Standardized**  
250 **Edition.** The PPVT-4 (Dunn & Dunn 2007) is a norm-referenced (mean = 100, SD = 15)

251 closed-set test of receptive vocabulary that can be used from the ages of 2.6 - 90+ years of  
252 age. Children are required to point to one of four pictures that best represents the meaning of  
253 a verbally presented stimulus word. All children were assessed using the PPVT-4, and their  
254 scores were compared to normative data using standard scores. The average reliability  
255 coefficient for this test, based on the normative sample, is 0.89.

256 **Preschool Language Scale – 4 (PLS-4; Australian Language Adaptation).** The PLS-4  
257 (Zimmerman et al. 2002) was used to assess the receptive and expressive language skills of  
258 five-year-old children. This assessment is norm-referenced (mean = 100, SD = 15), can be  
259 used from age 0 – 6 years, 11 months, and uses various stimulus materials such as toys,  
260 pictures and verbal prompts to elicit responses. Performance on this test is divided into  
261 subscale scores for Auditory Comprehension (AC), Expressive Communication (EC) and a  
262 Total Language Score. Each child's score is then compared with normative data using  
263 standard scores. The reliability coefficients for this test, based on the normative sample,  
264 range from 0.81–0.95.

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

274 **Wechsler Non-Verbal Scale of Ability (WNV).** The non-verbal cognitive skills of 86  
275 children were assessed using the WNV. The WNV is a cognitive assessment that uses

276 pictorial directions rather than language-based instructions, making it a suitable tool for  
277 children with hearing loss, perceptual difficulties or language delay. The test is norm-  
278 referenced and provides standard score measures of non-verbal cognitive skills. It has a full-  
279 scale score reliability of 0.91.

280 **Wechsler Preschool & Primary Scale of Intelligence – Third Edition.** The Performance  
281 IQ scale of the WPPSI –III was used to measure the non-verbal intelligence of five children.  
282 The Performance IQ scale examines non-verbal cognitive skills through the use of Block  
283 Design, Matric Reasoning and Picture Concepts. The test is norm-referenced and provides  
284 scaled score measures of non-verbal cognitive skills. The reliability coefficients for the  
285 WPPSI-III US composite scales range from .89 to .95.

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

## 298 **Statistical Analysis**

### 299 **PPVT linear regression analysis**

300 The sample of PPVT results for 91 five and eight-year-old children contained  
 301 considerable variation in the lengths of time children had used unilateral and bilateral CIs  
 302 (see second panel of Table 1). The information in this variation was exploited in a regression  
 303 to estimate the rate at which language ability accumulated over time, and whether this rate  
 304 was different for children with unilateral and bilateral CIs. The language accumulation  
 305 regression model had the general form:

$$306 \quad \text{PPVT}_i = \alpha_0 + \alpha_1 \text{ bilat yrs}_i + \alpha_2 (\text{age CI2}_i \times \text{bilat yrs}_i) + \alpha_3 \text{ unilat yrs}_i \\
 307 \quad + \alpha_4 (\text{age CI1}_i \times \text{unilat yrs}_i) + \gamma_1' X_{1,i} + \gamma_2' X_{2,i} + \gamma_3' X_{3,i} + \gamma_4' X_{4,i} + U_i, (1)$$

308 in which  $X_{1,i}$ ,  $X_{2,i}$ ,  $X_{3,i}$  each represent predictors respectively chosen from the Parenting style,  
 309 Child characteristics and Family background variables in Table 1. For a bilateral child, the  
 310 variable ‘bilat years’ is the length of time (in yrs) between activation of bilateral CIs and the  
 311 date of the PPVT assessment, while ‘unilat yrs’ is the length of time between activations of  
 312 unilateral and bilateral CIs. Only ‘unilat yrs’ is relevant for a child with a unilateral CI, and is  
 313 the length of time between unilateral activation and the PPVT assessment.

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

324 Given the moderate sample sizes available and the large number of possible predictors  
 325 that could be included in equation (1), a regression specification was chosen to minimise the  
 326 Akaike Information Criterion (AIC), which is a bias-corrected estimator of the Kullback-  
 327 Leibler divergence between the distribution implied by a statistical model and the distribution  
 328 of the data (Akaike 1974; Claeskens & Hjort 2008). Specifically, each possible specification  
 329 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 ( $X_{1,i}$ ),  
 330 parenting variables ( $X_{1,i}$ ), child characteristics ( $X_{2,i}$ ) and family background variables ( $X_{3,i}$ )  
 331 was estimated, and the specification with minimum AIC selected. The statistical analyses  
 332 were conducted using Eviews v7.1. Quantitative Micro Software, 2010.

333

#### 334 **PLS-4 and CELF-4 Linear regression analyses**

335 For each PLS-4 and CELF-4 language outcome, a regression was specified for  
 336 average test scores for children with and without bilateral CIs, controlling for parenting, child  
 337 and family characteristics. The differences between predicted test scores with and without the  
 338 bilateral CI were analysed to evaluate the effectiveness of the bilateral CI. The language  
 339 accumulation form of equation (1) for the combined sample of five and eight year olds was  
 340 inapplicable when these age groups were separated. Instead, for a language outcome  $Y_i$ , the  
 341 regression model was

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

344 with  $X_{1,i}$ ,  $X_{2,i}$ ,  $X_{3,i}$ ,  $X_{4,i}$  defined in equation (1). The coefficient  $\beta_1$  allowed the average  
 345 language outcome to differ between children with unilateral and bilateral CIs. In addition, the  
 346 distinct coefficients  $\beta_2$  and  $\beta_3$  on the interaction terms allowed the language effect of age of  
 347 first CI to differ between children with unilateral and bilateral CIs. This flexibility is  
 348 potentially important since age CI1 was, on average, significantly lower for the children with

349 bilateral CIs, and its effect may differ between the two groups. The coefficient  $\beta_4$  allowed the  
 350 language effect of bilateral CIs to depend on the age of its activation.

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

$$355 \quad y^{\text{bi}} = \beta_0 + \beta_1 + \beta_3 a_1 + \beta_4 a_2 + \gamma_1' x_1 + \gamma_2' x_2 + \gamma_3' x_3 + \gamma_4' x_4 ,$$

356 while for a unilateral CI it is

$$357 \quad y^{\text{uni}} = \beta_0 + \beta_2 a_1 + \gamma_1' x_1 + \gamma_2' x_2 + \gamma_3' x_3 + \gamma_4' x_4.$$

358 The difference between these predictions

$$359 \quad y^{\text{bi}} - y^{\text{uni}} = \beta_1 + (\beta_3 - \beta_2) a_1 + \beta_4 a_2$$

360 is a measure of the effectiveness of the second CI. This decomposes the predicted difference  
 361 in language due to bilateral CIs into three components: (i) a constant effect  $\beta_1$ , (ii) any  
 362 difference in the effect of age of first implant  $(\beta_3 - \beta_2) a_1$  and (iii) the effect of the age of the  
 363 second implant  $\beta_4 a_2$ . These individual components can be tested and interpreted based on the  
 364 individual coefficients. The overall effect of bilateral CIs, however, may depend on the  
 365 activation ages, so inference proceeded by computing a “bilateral activation window”, which  
 366 was the range of ages within which the activation of a bilateral CI produced a significant and  
 367 positive effect on the language outcome. For a given unilateral activation age  $a_1$ , the window  
 368 was defined to be the range  $[a_1, a_2]$ , where  $a_2$  was the oldest bilateral activation age such that  
 369 the 95% confidence interval for the overall bilateral effect  $y^{\text{bi}} - y^{\text{uni}}$  contained only positive  
 370 values. The window was empty if bilateral CIs had no significant positive effects.

371

## 372 **RESULTS**

### 373 **Bilateral / Unilateral differences of means**

374 Table 2 reports the difference of means *t*-tests for each of the seven language  
375 outcomes, allowing for unequal variances between the two groups. A significant difference ( $p$   
376 = 0.004) was found for the PPVT results, with children with bilateral CIs scoring an average  
377 of 9.36 points (10.98%) higher than children with unilateral CIs. Neither the PLS-4 nor  
378 CELF-4 mean scores revealed significant differences between children with bilateral and  
379 unilateral CIs without controlling for other variables.

380

### 381 **Vocabulary (PPVT)**

382 Mean scores for both groups of children with unilateral and bilateral CIs were within one  
383 SD of the mean for typically developing children with normal hearing, although this was the  
384 case for the former group only by a narrow margin. As is commonly observed (Blamey et al.  
385 2001; Sarant & Garrard 2013), variability in scores was high, with some children scoring  
386 within or above the average range, and others well below it.

387 Table 3 gives results of the regression to predict bilateral / unilateral vocabulary  
388 differences while controlling for the parenting, child and family characteristics listed in Table  
389 1. The language accumulation specification for the PPVT score revealed that the number of  
390 years spent with bilateral CIs was a highly significant ( $p < 0.001$ ) predictor, with its effect  
391 moderated by bilateral activation age ( $p = 0.033$ ). For example, a bilateral CI activated at age  
392 2.93 years (the mean activation age for this sample) was estimated to result in a significant ( $p$   
393 = 0.004) rate of language improvement of  $3.95 - 0.69 \times 2.93 = 1.93$  points (95% confidence  
394 interval = [0.62, 3.23], 2.10% of the unilateral average) for each year thereafter. The joint  
395 semi-partial  $R^2$  of the two bilateral variables was 0.09 out of a total  $R^2$  of 0.51. The variables  
396 measuring the unilateral CI years were not found to be significant, being excluded by the AIC  
397 from the final specification.

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

403 Children's birth order was found to be a highly significant predictor of vocabulary  
404 outcomes ( $p = 0.001$ ), and contributed 0.07 to the overall  $R^2$ . Each older sibling predicted a  
405 decreased PPVT score by 4.65 points ( [1.85, 7.45], 5.05%). The education level of the  
406 primary caregiver was a highly significant predictor of vocabulary outcomes ( $p = 0.001$ ),  
407 with the child of a tertiary qualified parent predicted to have a score 9.56 points ( [3.83,  
408 15.28], 10.38%) higher than otherwise.

409

## 410 **Language**

411 Tables 4, 5, and 6 give the results of the regressions to predict bilateral /unilateral  
412 language differences while controlling for the parenting, child and family characteristics  
413 listed in Table 1.

### 414 **Language outcomes for 5-year-old children (PLS-4)**

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

418 Having bilateral CIs was not an individually significant predictor of outcomes in any  
419 of the PLS-4 regressions. The activation age of bilateral CIs, however, was a significant  
420 predictor of Total Language and Auditory Comprehension scores ( $p = 0.002, 0.010$ ). The  
421 coefficients were negative, implying that earlier bilateral implantation predicted improved  
422 language outcomes; a reduction of one year in age at bilateral CI predicted an increase of

423 5.96 points ([2.31, 9.61], 6.25%) on Total Language scores and 4.80 points ( [1.20, 8.39],  
424 4.60%) on Auditory Comprehension scores. The effect of age at first CI followed a  
425 qualitatively similar pattern for the unilateral children for all three PLS subscales ( $p = 0.000$ ,  
426 0.000, 0.000 for Total Language, Auditory Comprehension and Expressive Language  
427 respectively). For a child with a unilateral CI, a reduction of one year in age at implantation  
428 predicted increases of 13.82 points ( [8.80, 18.84], 19.94%), 11.37 points ( [6.44, 16.32],  
429 17.10%) and 14.50 points ( [9.62, 19.39], 19.40%) on the Total Language, Auditory  
430 Comprehension and Expressive Language scores respectively. The activation age at first CI  
431 contributed most (0.22, 0.17, 0.25 for Total Language, Auditory Comprehension and  
432 Expressive Language) to the overall  $R^2$  of each regression, while the significant contributions  
433 of the activation age at second CI were 0.08 (Total Language) and 0.06 (Auditory  
434 Comprehension).

435 The results for the joint inference based on the bilateral activation windows are given  
436 in Table 7. The results for the Total Language and Auditory Comprehension subscales show  
437 that bilateral CIs predicted significant language improvements by age five years, provided  
438 they were activated early enough. As with the CELF Receptive Language scores, the  
439 windows became wider as the age at first CI increased. The windows based on PLS-4 scores  
440 were narrower than those based on CELF-4 scores for the older children, implying that  
441 bilateral CI activation needs to occur early to give sufficient time to yield significant  
442 language improvement by age five years. The absence of significant bilateral variables in the  
443 PLS-4 Expressive Language equation translated to an empty bilateral activation window –  
444 there was no evidence of improved Expressive Language scores for children with bilateral  
445 CIs at any activation age.

446 Two of the parenting predictors were found to be significant. The adult reading  
447 variable was significant in all three equations ( $p = 0.012$ , 0.014, 0.012 for Total Language,

448 Auditory Comprehension, and Expressive Language respectively). The marginal effect on the  
449 Total Language score of an extra hour per week of adult time spent reading to the child was  
450 an increase of  $60 \times 0.096 = 5.76$  points ( [1.33, 10.17], 6.64%). The results of the Auditory  
451 Comprehension and Expressive Language equations were similar. Screen time was found to  
452 have a significant effect on Total Language and Expressive Language scores ( $p = 0.039$ ,  
453 0.025), with an extra hour of screen time per day predicting a decrease in Total Language  
454 scores of 2.57 points [0.13, 5.00] and 2.71 points [0.36, 5.06] for the Expressive score. A  
455 joint interpretation of these two parenting variables is that the predicted effect of a parenting  
456 intervention that reduces a child's screen time by half an hour every weekday, and substitutes  
457 with half an hour of adult reading time (an extra 150 minutes per week), would be to  
458 significantly ( $p = 0.005$ ) increase the Total Language score by  $150 \times 0.096 + 0.5 \times 2.57 =$   
459 15.66 points ( [5.01, 26.30], 18.08%).

460 Two of the child characteristics were highly significant predictors of language  
461 outcomes on this test, and showed effects of a substantial magnitude. The effect of birth order  
462 was highly significant in each equation ( $p < 0.001$ ), with each older sibling predicting a  
463 decrease in Total Language, Auditory Comprehension and Expressive Language scores of  
464 8.39 points [4.39, 12.40], 8.13 points [4.19, 12.08] and 7.18 points [3.29, 11.06] respectively.  
465 Birth order also made a substantial contribution to the  $R^2$  of all three equations (0.13, 0.14,  
466 0.10 to Total Language, Auditory Comprehension and Expressive Language respectively).  
467 Gender was also significant in each equation ( $p = 0.010, 0.018, 0.006$ ) with boys predicted to  
468 score 10.67 points [2.71, 18.63], 9.55 points [1.71, 17.38], and 10.92 points [3.24, 18.60]  
469 lower on the Total Language, Auditory Comprehension and Expressive Language scores,  
470 holding all other characteristics constant.

471 A family history of hearing loss was the only family history variable to be found  
472 significant in any of the equations. It was highly significant for all three subscales ( $p = 0.003$ ,

473 Total Language; 0.004, Auditory Comprehension; 0.010, Expressive Language) and had a  
474 positive effect on scores. For example, having a family history of hearing loss predicted a  
475 14.21 point ( [4.98, 23.44], 16.39%) higher Total Language score, relative to a child with all  
476 of the same characteristics but with no family history of hearing loss.

477

#### 478 **Language outcomes for 8-year-old children (CELF-4)**

479 Language outcomes on both measures for the children in this study again showed  
480 great variability, as has been reported in previous studies using these measures (Spencer et al.  
481 2003; Tobey et al. 2013). Group mean scores for all of the CELF-4 subtests were within one  
482 SD of the means for typically developing children, but there was great variability in results  
483 between children.

484 Bilateral CIs were a significant predictor for both the Core Language and Expressive  
485 language subscales of the PLS-4 ( $p = 0.004, 0.002$  respectively), although not for the  
486 Receptive Language subscale. The magnitude of the effect was moderated by the age at  
487 which the second CI was activated ( $p = 0.007, 0.007$ ). The predicted effect of a bilateral CI  
488 activation at the mean activation age of 2.94 years was  $58.31 - 11.36 \times 2.94 = 24.91$  points  
489 [6.42, 43.41] on the CELF Core Language subscale, or 28.06% higher than a unilateral child  
490 with all of the same other characteristics. The same calculation for the CELF-4 Expressive  
491 Language subscale showed a 34% higher result (29.34 points, [11.78, 46.89]) for a child with  
492 bilateral CIs implanted at the average age. For a child with bilateral CIs, each additional year  
493 of delay of implantation of the bilateral CI predicted a decrease in CELF Core Language and  
494 Expressive Language scores of 11.36 points ( [3.47, 19.25], 12.16%) and 10.83 points ( [3.34,  
495 18.32], 11.40%) respectively.

496 The CELF-4 regressions accounted for between 59-65% of the variability in outcomes  
497 on this measure. The contributions of having bilateral CIs and the bilateral activation age to

498 the predictive ability of the regressions are given by the semi-partial  $R^2$  statistics in Table 3.  
499 These two variables were, by definition, highly collinear (correlation coefficient of 0.74),  
500 measuring two aspects of bilateral CIs. The overall predictive ability of bilateral CIs was  
501 therefore best evaluated using the joint semi-partial  $R^2$  of the bilateral indicator and CI age,  
502 which were 0.20 (Core Language) and 0.22 (Expressive Language). That is, the overall  $R^2$  of  
503 0.59 and 0.63 for the Core and Expressive regressions would decrease by 0.20 and 0.22  
504 respectively if the two bilateral CI variables were omitted.

505 The bilateral CI indicator was not selected in the CELF Receptive Language equation,  
506 but the child's age at bilateral implantation was selected, and was statistically significant ( $p =$   
507 0.036), indicating that bilateral implantation is predictive of these outcomes, with a unique  
508 contribution of 8% of the variance in outcomes. A delay of one year in bilateral implantation  
509 predicted a 4.11 point ([0.29, 7.93], 4.39%) decrease in the CELF Receptive Language score.  
510 Child age at first CI was found to be significant ( $p = 0.004$ ) for children with unilateral CIs,  
511 but not for children with bilateral CIs. A delay of one year in first CI for a child with a  
512 unilateral CI predicted a 13.91 point ([5.07, 22.75], 15.10%) decrease in CELF-4 Receptive  
513 Language score.

514 These results suggest that bilateral CIs had a statistically significant effect on CELF-4  
515 language scores of the 8-year-old children in this study, the magnitude of which varied with  
516 the ages at which children received their CIs. The prediction comparison approach outlined  
517 above provides a readily interpretable summary of the predicted effectiveness of bilateral CIs.  
518 Table 6 presents results for several first implant activation ages. For the Core Language and  
519 Expressive Language subscale results, bilateral CIs were found to have a significant and  
520 positive effect, provided they were activated before ages 3.75 and 4.33 years respectively.

521 The results for the child, parent and family characteristics were similar for the Core  
522 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 one 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.

539

## 540 **DISCUSSION**

541

### 542 **Vocabulary Outcomes**

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

548

**549 Language Outcomes**

550 In terms of the comparative performance of children with unilateral and bilateral CIs,  
551 the language results were not as unequivocal as the vocabulary outcomes, with no significant  
552 differences between the mean scores for unilateral and bilateral CI children for any of the  
553 CELF-4 and PLS-4 subtests. However, once other influential factors were controlled for in  
554 the regression analysis, bilateral CI use was found to predict significantly faster rates of  
555 language development in both of the PLS-4 and CELF-4 tests.

556 The weaker effect of bilateral CI use on the simple difference of means for the  
557 language measures is likely to reflect both the increased complexity of language skills  
558 required, the substantial length of time required to master these, and the fact that the sample  
559 sizes for both of these measures were smaller than for the vocabulary measure, where results  
560 for both the five and eight-year-old children were included. An illustration of the need for  
561 more time to develop higher level language skills can be seen in the Core Language and  
562 Expressive Language CELF-4 regression results, where bilateral CIs were found to have a  
563 significant and positive effect on scores, provided they were activated before ages 3.75 and  
564 4.33 years respectively. Activation of bilateral CIs after these ages may not have allowed  
565 sufficient time to produce a significant benefit relative to the unilateral implant for the 8-year-  
566 old participants of this study.

567

**568 The effect of bilateral hearing on vocabulary and language outcomes**

569 It was speculated earlier that the perceptual benefits reported for children with  
570 bilateral CIs such as improved speech perception in both quiet and noisy listening conditions  
571 (Scherf et al. 2007; Zeitler et al. 2008), improved sound localization ability for some children  
572 (eg. Lovett et al. 2010), and reduced listening effort (and therefore reduced tiredness and a

573 greater ability to concentrate) (Hughes & Galvin, 2013) may facilitate a greater ability to  
574 access the spoken language of others and to learn from these increased opportunities. Despite  
575 the large variability in language outcomes, a moderate sample size, and multiple factors that  
576 influence these outcomes, significantly faster rates of vocabulary and language development  
577 were found for the children with bilateral CIs in this study. This finding suggests that the  
578 perceptual benefits of bilateral hearing through two CIs conferred a significant advantage, in  
579 terms of learning, to these children. It must be emphasized, however, that it is not known  
580 whether the improved results for children with bilateral CIs are due simply to having bilateral  
581 auditory input, or whether they are the result of true binaural processing. This study was not  
582 designed to address this question, but it will be an important area for future investigation.

583

## 584 **Predictors of Outcomes**

### 585 **Age at First CI**

586 Age at CI has been documented extensively a significant predictor of child language  
587 outcomes (Connor et al. 2006; Geers et al. 2009; Schorr et al. 2008), and marks the beginning  
588 of functional auditory input to the auditory cortex and the subsequent development of the  
589 auditory processing abilities that facilitate spoken language development. A surprising  
590 finding of this study was that neither age at first CI nor duration of unilateral CI use were  
591 significant predictors of PPVT scores. This does not imply that a unilateral implant is  
592 ineffective, but that there was insufficient information in this sample (with only 24 children  
593 with unilateral CIs) to precisely estimate the rate of vocabulary accumulation. It is also worth  
594 noting that for many children in this sample, four to seven years had elapsed since their first  
595 CI implantation. Although several studies have reported better language outcomes with very  
596 early implantation (Dettman, et al. 2007; Nicholas & Geers 2007), follow up in these studies  
597 occurred when the children had limited experience with their CIs (Geers 2004). More recent

598 findings have been mixed; one study has since reported that the effect of age at first implant  
599 reduces over time (Hay-McCutcheon et al. 2008), while a very recent study has shown that  
600 age at first CI is still a strong predictor of outcomes after approximately 8 years of CI use  
601 (Geers & Nicholas 2013).

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

607

#### 608 **Age at Second CI**

609 As far as the authors are aware, the effect of age at second CI on language outcomes  
610 has not been examined in any other studies. Here it was found to have an important effect,  
611 with earlier ages of second implantation predicting significant vocabulary and language  
612 improvements for both five and eight year olds. This effect was quantified in different ways  
613 for the different tests.

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

618 A bilateral CI predicted statistically significant language improvements on all PLS-4  
619 and CELF-4 subtests except the CELF-4 Expressive, and the extent of these improvements  
620 was influenced by various interactions between the ages of first and second implant. These  
621 results are summarised in the bilateral activation age windows. For each subtest, the bilateral  
622 activation age window provides the range of ages within which the activation of a bilateral CI

623 predicts a statistically significant advantage over a unilateral CI. That these windows exist for  
624 all but the CELF-4 Expressive sub-test demonstrates the diverse benefits of the bilateral CI  
625 for both five and eight year olds.

626 To illustrate, activation windows with fixed upper age limits were found for the  
627 CELF-4 Core (3.75 years) and Expressive (4.33 years) subtests, implying that bilateral  
628 implantation before these ages was sufficient to produce significant gains by age 8, regardless  
629 of age of first CI (at least within the range in this sample). In all cases, the magnitude of these  
630 language gains decreased as age of bilateral CI increased. For example, a child receiving a  
631 bilateral CI at age 2 is predicted to score 35.60 points higher on the CELF-4 Core test relative  
632 to a unilateral CI child, but if the activation age were 3.75 years then this gain reduces to  
633 15.72 points. Therefore early bilateral implantation would be recommended for maximum  
634 language benefit.

635 Some bilateral activation age windows depended on the age of first CI. For example,  
636 the upper age limit for the CELF-4 Receptive Language subtest was found to vary with the  
637 age of first CI. If a child received their first implant at age 1 year, then a subsequent bilateral  
638 implant received before age 2.09 years predicted a significant and positive effect on the  
639 Receptive Language subscale score. If the first CI were received at age 2, then the bilateral  
640 activation window extended to age 4.18. This longer window for children with later age at  
641 first CI reflects the considerably larger implant age effect for the first CI relative to the  
642 second – a child first implanted at age two years is acquiring hearing and language from a  
643 point substantially (predicted 15.10%) behind a child first implanted at age one year; a  
644 deficiency that the bilateral implant can significantly reduce. Note, however, that the  
645 predicted language scores are always higher for children with lower implant ages, so this  
646 longer bilateral activation window does not imply better language scores for children

647 implanted later, rather than a bilateral CI has the potential to help catch up some (not all) of  
648 the delay due to a delayed first implant.

649 Two of the three PLS-4 bilateral age activation windows were non-empty (Total  
650 Language and Auditory Comprehension), implying that a bilateral child will already be  
651 demonstrating improved language outcomes by age five, provided they receive their second  
652 implant sufficiently early. These activation windows were shorter than those for the CELF-4,  
653 implying that, compared to the eight year olds, earlier bilateral implantation is required for  
654 bilateral CI children to show significant language gains relative to unilateral CI children by  
655 age five.

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

661

## 662 **Parenting Style**

663 The effect of parental involvement on both vocabulary and language development  
664 was found to be strongly predictive of better outcomes, as has been documented previously  
665 (Moeller 2000; Sarant et al. 2009). The amount of time adults in the family spent reading to  
666 their child, and the amount of time children spent watching a screen each week were  
667 identified as new factors that significantly affected both vocabulary and language outcomes.  
668 An example of a joint interpretation of these two parenting variables is that the predicted  
669 effect of a parenting intervention that reduced a child's screen time by 30 minutes each  
670 weekday and substituted this with 30 minutes of adult-to-child reading time (150 extra  
671 minutes/week) would be a significant increase ( $p = 0.005$ ) in the PLS-4 Total Language score

672 by 15.66 points (18.08%;  $150 \times 0.096 + 0.5 \times 2.57$ ). This large predicted increase in scores  
673 demonstrates how effective relatively small changes in parenting can be in facilitating  
674 significant changes in children's language development.

675

## 676 **Child Characteristics**

677 Birth order was found to have a significant effect on PPVT scores, with each older  
678 sibling decreasing scores by approximately five percent. It also exerted a significant negative  
679 effect on language scores for five of the six subscales of both language measures. This has  
680 been found to be the case in other studies of children with normal hearing, and likely relates  
681 to the demands on parents' time and resources with increasing numbers of children (Hoff  
682 Ginsberg 1998; Nelson et al. 2006). Although it has also been found that older siblings can  
683 enrich aspects of younger children's language development, it has also been reported that  
684 older children monopolize more parent-child conversations than their younger siblings  
685 (Oshima-Takane et al. 1996; Wellen 1985). Children with significant hearing loss are likely  
686 to miss out on the enriching aspects of 'overhearing' conversations due to their auditory  
687 limitations, and are also likely to be disadvantaged by the reduced number of opportunities to  
688 talk directly with their parents and learn language in this manner.

689 In common with other reports for both children with normal hearing (Fenson et al.  
690 2000) and children with hearing loss (Geers et al. 2009; Moog & Geers 2003), male gender  
691 was significantly predictive of poorer language PLS-4 outcomes for five-year-old children.  
692 However, this effect was no longer evident for the eight-year-old children on the CELF-4  
693 results. This finding fits with reports that the gap in language ability found between boys and  
694 girls in the early life closes with increasing age (Ely, 2005; Gaddes & Crockett, 1975).

695 Non-verbal cognitive ability also significantly affected language outcomes for the 8-  
696 year-old children in this study. Cognitive ability has been identified as a primary predictive

697 factor of language development in children with hearing loss, accounting for large  
698 proportions of the variance in outcomes in many studies (eg. Geers et al. 2009; Holt & Kirk  
699 2005; Sarant et al. 2010).

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

713

#### 714 **Family Background**

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

719 Having a family history of hearing loss also had a surprisingly large positive effect on  
720 language outcomes for five-year-old children. This may reflect the value of having a family  
721 member with an understanding of the challenges of hearing loss and knowledge of strategies

722 to access assistance in the early years of parenting a child with a hearing loss. While family  
723 histories of spoken language or reading difficulties have historically been reported as risk  
724 factors for language development (Nelson et al. 2006), the authors are unaware of any  
725 evidence regarding family history of hearing loss. It is interesting that this factor did not  
726 influence language outcomes for the eight-year-old children, where a high level of parent  
727 involvement accounted for 10% of the variance in outcomes. It may be that by the time  
728 children are 8 years old, parents have developed sophisticated parenting practices that exceed  
729 the effect of the knowledge possessed by parents with a family history of hearing loss, or  
730 simply that different predictors were found for these different groups of children.

731

### 732 **Study Limitations**

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

741

### 742 **Conclusions**

743 Overall, bilateral CIs contributed to significantly better language outcomes for the  
744 children in this study, with improvements moderated by children's ages at second CI. Earlier  
745 implantation, both unilateral and bilateral, was found to be beneficial to language outcomes.  
746 With larger sample sizes in the future, possible non-linearities in implant age effects, such as

747 whether there is a discontinuity between simultaneous and sequential implantation could also  
748 be investigated. Examining whether there is an asymptote to the negative effect of delaying  
749 implantation, in addition to determining a window of implant ages of maximum effectiveness  
750 would provide valuable clinical information.

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

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

762 The findings of this study add significantly to the body of knowledge regarding  
763 outcomes for children with bilateral versus unilateral CIs, and also to that of factors which  
764 facilitate improved spoken language outcomes for children with CIs. It is imperative that this  
765 knowledge is accessible to, and is used by parents, clinicians and governments in making  
766 evidence-based decisions that are translated into clinical practice and governmental policy  
767 development. In this way, we can most effectively assist the many children around the world  
768 for whom bilateral CIs can contribute to closing the gap, relative to those with normal  
769 hearing, in terms of language development, and hopefully, subsequent quality of life.

770

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964 **Appendix 1**965 **Reading Habits Questionnaire for a child aged five years**

966

- 967 1. Your child's name (first name and surname)
- 968 2. Your child's current age. Please respond in years and months, e.g. 7 years and 6  
969 months
- 970 3. Your name (first name and surname)
- 971 4. Your date of birth (the date of birth of the person filling out this survey)
- 972 5. Your current work status
- 973 6. Have you been able to establish a reading routine with your child (for example,  
974 where an adult in your family reads to the child as part of a nap, bedtime or other  
975 routine)?
- 976 7. If yes, at what age was a reading routine established? Please respond in years and  
977 months
- 978 8. When you started this routine, how many days in a typical week was your child  
979 read to?
- 980 9. When you started this routine, for about how many minutes was your child read to  
981 at a sitting?
- 982 10. Currently, in a typical week, how many days per week does an adult in your  
983 family read to your child?
- 984 11. Currently, for about how many minutes is your child read to at a sitting?
- 985 12. Has your child started reading? (ie. able to recognise and read out loud single  
986 words or text)
- 987 13. At what age did your child start reading to you? (Please respond in years and  
988 months)

- 989 14. Currently, in a typical week, how many days per week does your child read out  
990 loud to you?
- 991 15. Currently, for about how many minutes does your child read out loud to you at a  
992 sitting?
- 993 16. Currently, in a typical week, how many days per week does your child read books,  
994 or look at books, on their own?
- 995 17. Currently, on average, for about how many minutes does your child look at books,  
996 or read on their own, at a sitting?
- 997 18. Is there a family history of reading difficulties?
- 998 19. Is there a family history of speech and/or language difficulties?
- 999 20. How many children's books do you have in your home now, including library and  
1000 school books?
- 1001 21. In the past month, has your child visited a library?
- 1002 22. Currently, on a typical weekday, for how many hours does your child watch TV,  
1003 DVDs , computer games, Wii etc.?
- 1004 23. Do you have any additional comments?