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The effect of cochlear implant interval on spoken language skills of pediatric bilateral cochlear implant users

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Abstract

Objective: To examine the effects of cochlear implant (CI) interval (time between CI surgeries) on receptive vocabulary and receptive language skills for children with bilateral CIs.

Study Design: A prospective cross-sectional study design.

Setting: Participants were recruited from, and tested at, oral schools for the deaf and pediatric audiology clinics across the United States.

Patients: Eighty-eight children, 4 to 9 years of age, with bilateral CIs and known hearing histories. Twenty-three participants received CIs simultaneously and 65 received CIs sequentially. Of those implanted sequentially, 86% wore a hearing aid (HA) on the non-implanted ear during the CI interval.

Intervention: Bilateral cochlear implantation.

Main Outcome Measures: Receptive vocabulary was measured via the Peabody Picture Vocabulary Test (PPVT). Receptive language skills were measured via the Clinical Evaluation of Language Fundamentals (CELF).

Results: Multiple linear regression models indicate better receptive vocabulary and receptive language skills are associated with earlier ages at first CI (CI 1), but not with shorter CI intervals.

Conclusions: Early cochlear implantation (i.e., age at CI 1) is critical for better receptive vocabulary and receptive language skills. Shorter CI intervals are not associated with better receptive vocabulary and receptive language skills for these 88 children, who nearly all used bimodal hearing during the interval. Use of a HA at the non-implanted ear, prior to receipt of a second CI (CI 2), may mitigate the effects of early bilateral auditory deprivation.

INTRODUCTION

It is widely accepted that cochlear implants (CIs) offer substantial benefits for listeners with severe to profound hearing loss (HL), particularly for children receiving CIs early in life[1-7]. Although the auditory system typically receives signals at two ears, until recently unilateral cochlear implantation (one CI, and *no* device at the non-implanted ear) was the predominant protocol. Consequently, such listeners using a unilateral CI would be at a disadvantage for localizing sounds and listening in noise. Currently, stimulation at both ears, via either bilateral CIs (two CIs) or bimodal devices (CI with a hearing aid (HA) at the non-implanted ear), is recommended for most listeners[8-15].

Any absence of hearing, or auditory deprivation, can have degenerative consequences and may allow auditory areas of the brain to reorganize and reallocate to other senses[16-18]. For children with severe-to-profound hearing loss (HL), auditory deprivation begins at the onset of deafness and continues until they receive a CI (CI 1). If there is no auditory input at the other ear (no HA nor a CI 2), then bilateral stimulation of auditory pathways will not occur (the child experiences ‘*bilateral* auditory deprivation’; i.e., with unilateral stimulation there is a deprivation of *bilateral* auditory inputs). Such unilateral auditory input, during early childhood, may have negative consequences for bilateral hearing abilities and possibly for spoken language development[19-21]. If, however, there is auditory input at the other ear, i.e., the child uses bilateral cochlear implants[22, 23] or bimodal devices, then bilateral auditory deprivation might be mitigated.

For the development of speech and language skills in young deaf children, there is nearly undisputed evidence that receiving a CI (CI 1) at an earlier age is better. That is, the younger the age of the child when she/he receives CI 1 (ideally in the range from 9 months – 2 years of age), the better her/his speech and language skills[24]. There is, however, less consistent evidence on the importance of the age at which a child receives a second CI (CI 2) (mathematically, Age at CI 1 + Duration of CI interval = Age at CI 2). Importantly, the effects of age at CI 2 are likely influenced by age at first CI (CI 1), and both the severity of HL and use of a HA at the non-implanted ear during the CI interval (the time between CI 1 and CI 2). Use of a HA at the non-implanted ear for children with severe (less-profound) HL could maintain stimulation of auditory pathways bilaterally during critical developmental periods. As might be expected for comparisons of bilateral to unilateral hearing, there are numerous reports of HA use in children, at the non-implanted ear, providing a variety of benefits[8, 11, 12, 25]. There are also a few reports of HA use, at the non-implanted ear prior to receiving CI 2, providing benefits for some children when compared to children who did not use a HA during the CI interval. Again, such benefits might be attributable to maintained stimulation of auditory pathways bilaterally[8-12, 23, 26].

Some researchers and clinicians suggest that children who are CI candidates receive bilateral CIs simultaneously when possible, and that children receiving sequential CIs should receive CI 2 at the earliest opportunity, preferably within 12 months of CI 1[22, 23, 27-29]. They suggest that bilateral CIs, received early in age, may lead to better speech perception skills, better language abilities, increased symmetry in central auditory development, and prevention of bilateral auditory deprivation. Physiological evidence, based on data from

children with unilateral CIs (N=245), indicates a critical period (birth to ~3.5 years old) for the development of normal P1 latencies in auditory evoked cortical potentials; children who receive a CI during this critical period develop normal P1 latencies[30]. With bilateral CIs, Bauer and colleagues[31] report faster trajectories to normal P1 latencies for children with simultaneously implanted CIs than for those with sequentially implanted CIs (N=4; CI intervals of 5 and 12 months; all four children received both CIs by 24 months of age). Behavioral evidence in favor of shorter, or 'zero' (i.e., simultaneous bilateral CIs), CI intervals is based on speech perception, localization, and listening-in-noise abilities of children with bilateral CIs[21, 32-36]. Results from these reports suggest that receiving CI 2 earlier yields better outcomes. For example, Gordon and Papsin[37] report that improvements (speech perception with bilateral CIs re: speech perception with 1st CI) were greatest for children with shorter CI intervals. Notably, however, in this study only 12 of the 52 children receiving sequentially-implanted CIs wore a HA for *any* length of time during the CI interval, and of those 12 children, six wore their HA for a very-limited number of months (e.g., 3 months).

While improved speech perception could facilitate better receptive language skills, evidence of an effect of CI interval on language skills is extremely limited. There are only two such studies. In the first, Boons and colleagues[38] assessed the effects of CIs on receptive and expressive language skills of Dutch-speaking children via retrospective chart review. Twenty-five children with bilateral CIs were matched carefully (e.g., age at diagnosis, etiology, and school environment) to 25 children with a unilateral CI (*no* device/auditory stimulation at the non-implanted ear). They report, as expected, that children with bilateral CIs have better receptive and expressive language skills than children with unilateral CIs. Additionally, for the group of children with bilateral CIs, they report that the CI interval between (range: 0-36 months) correlates negatively with language scores. However, for these children with bilateral CIs, use of a HA at the non-implanted ear during the CI interval is not specified. In the second study on language, Sarant and colleagues[39] compared receptive vocabulary, and receptive and expressive language skills of bilateral (N = 67) to unilateral (N = 24) CI users (5- or 8-year-old Australian children). Sarant and colleagues found that bilateral CI use, and earlier ages at both CI 1 and CI 2 were associated with better receptive vocabulary and receptive language skills, after controlling for parenting, child, and family characteristics. However, of the children with bilateral CIs, only 32% used a HA at the non-implanted ear during the CI interval. That is, 68% had no stimulation at the non-implanted ear. Although both studies indicate shorter CI intervals are associated with better language outcomes, these two studies' results should be interpreted within the context that there was limited, or no, information regarding stimulation of bilateral auditory pathways during the CI interval. Notably, there was a lack of HA use (or, no mention of HA use) at the non-implanted ears by most of the participants during the CI intervals. A third study of language abilities of children with CIs is relevant, though the researchers did not examine the effects of CI interval on outcomes. Moberly and colleagues[40] examined, at 4th grade, a group of 48 children with CIs. Children used unilateral CIs (*no* device/stimulation at the non-implanted ear; N = 8), bimodal devices (N = 8), or bilateral CIs (N=32). The group using bilateral CIs was evenly divided into those with at least 11 months of bimodal device experience before receiving CI 2 (N=16) and those who did not (N=16). Overall, children

with bimodal devices or bimodal experience outperformed the others on measures of phonemic awareness and language skills, suggesting that having some period of early bimodal device use may provide benefits for language skills.

In summary, there is evidence that bilateral CI use (compared to unilateral CI use) and receiving CI 1 earlier facilitate spoken language skills. The current evidence, however, is less clear and is limited regarding the effect of age at CI 2 (or, equivalently CI interval) on language skills, especially since HA use at the ear to receive CI 2 may moderate these effects.

Thus, there is a need for further research examining the effects of timing of CI implantation on language skills for children with known bilateral device histories. The goals of this current study are to examine the effect of CI interval on receptive vocabulary and receptive language in a large number of children ($N = 88$) with bilateral CIs and for whom bilateral hearing device usage is known.

MATERIALS AND METHODS

Participants

Participants were recruited from, and tested at, oral schools for the deaf and pediatric audiology clinics across the United States. This research was approved by the Human Research Protection Offices of Washington University School of Medicine in St. Louis. A parent/guardian of each participant gave informed consent and each participant gave informed assent to participate in this research. Eighty-eight children (42 female) with the following characteristics were included: primarily severe to profound HL pre-CI (in the ear(s) to be implanted), currently using bilateral CIs, educated in programs that focused on listening and spoken language, HL as primary disability, and no device failures lasting > 30 days. Participants were 4 to ~ 9 years of age at testing (mean = 7; SD = 1.2). See Table 1 for details.

Audiologic and Device Characteristics

Etiologies of HL included genetic, syndromic, enlarged vestibular aqueduct (EVA), cytomegalovirus (CMV), ototoxic exposure, Auditory Neuropathy Spectrum Disorder (ANSD), and one child with meningitis acquired at 20 months of age. Ten participants had progressive HL. All participants, except one (the one child who had meningitis received bilateral CIs quickly due to concerns of cochlear ossification), were fit with HAs prior to CI surgery. Twenty-three participants received their CIs simultaneously (both CIs activated the same day), while 65 received their CIs sequentially. Unaided pre-CI pure tone averages (PTA: thresholds at 500, 1000, 2000 Hz), obtained from participants' audiologists, represent thresholds of the better-ear pre-CI for simultaneously implanted CI recipients and thresholds of the CI 2-ear pre-CI2 for sequentially implanted CI recipients. As such, participants' unaided pre-CI PTAs ranged from 50 to 125 dB HL (mean = 100.4; SD = 15). Two participants had a pre-CI PTA < 70 dB HL. These participants were implanted due to their diagnosis of ANSD. All participants received their initial CI before 55 months of age (mean = 21.6 months; SD = 11.3). Across all participants, the CI interval ranged from 0 to 67

months (mean = 10 months; SD = 11.8). Of the sequential CI users, 86% (n = 56) had a period of bimodal device use pre-CI 2, and their duration of bimodal use ranged from 0.3 to 36 months (mean = 11.2; SD = 8.6).

Language Outcomes: Receptive Vocabulary and Receptive Language

The Peabody Picture Vocabulary Test (PPVT) was administered to assess receptive vocabulary ability[41]. Three subtests of the Clinical Evaluation of Language Fundamentals (CELF) were administered to assess comprehensive receptive language: Concepts and Following Directions, Receptive Word Classes, and Sentence Structure[42]. Raw scores from both the PPVT and CELF tests were converted to standard scores (mean = 100; SD = 15).

Non-verbal IQ

Non-verbal IQ was assessed via the Block Design subtest of one of two assessments. Children younger than 6 years of age were given the Wechsler Preschool and Primary Scale of Intelligence (WPPSI)[43] while those who were 6 years or older were administered the Wechsler Abbreviated Scale of Intelligence (WASI)[44]. Again, raw scores were converted to standard scores (mean = 100; SD = 15).

Maternal Education

Maternal education (in years) was reported by a parent or guardian via a family characteristics and demographics questionnaire, created for this current study.

Statistical Analysis

Results from the Kolmogorov-Smirnov Test ($p > 0.05$) revealed data were normally distributed. Pearson product correlations were used to guide multivariable linear regression models to examine the effects of CI interval on receptive vocabulary and receptive language skills. Confidence intervals of estimates, in addition to statistical significance, are reported[45].

RESULTS

Receptive vocabulary scores from the PPVT ranged from 56 to 138 (mean = 93.4; SD = 17.1). Twenty-nine participants scored at or above the normative mean (PPVT ≥ 100), 32 scored within 1 SD below the normative mean ($85 \leq \text{PPVT} < 100$), 23 scored within 1 to 2 SD below the normative mean ($70 \leq \text{PPVT} < 85$), and 4 scored below 2 SD of the normative mean (PPVT < 70). Receptive language scores from the CELF ranged from 51 to 131 (mean = 91.5; SD = 19.6). Thirty-six participants scored above the normative mean, 19 scored within 1 SD below the normative mean, 17 scored within 1 to 2 SD below the normative mean, and 16 scored below 2 SD of the normative mean. Non-verbal IQ scores ranged from 65 to 140 (mean = 99; SD = 15.3). Maternal education ranged from 11 to 20 years (mean = 16; SD = 2.2).

Pearson Product Correlations

Pearson product correlations were computed amongst demographic, audiologic and language outcomes, to guide selection for use in multivariable linear regression models. Age at CI 1 ($r = -0.32$, $p < 0.01$ and $r = -0.25$, $p < 0.05$), Non-verbal IQ ($r = 0.53$, $p < 0.01$ and $r = 0.55$, $p < 0.01$), and Maternal Education ($r = 0.29$, $p < 0.01$ and $r = 0.24$, $p < 0.05$) were significantly correlated with PPVT and CELF, respectively. Neither CI interval nor unaided pre-CI PTA was significantly correlated with either language outcome. And, notably, although unaided pre-CI PTA was significantly correlated with age at CI 1 ($r = -0.46$, $p < 0.01$), it was not correlated with CI interval ($r = -0.04$).

Linear Regression Models

Linear regression models were explored for receptive vocabulary (PPVT) and receptive language (CELF) outcomes. For both outcomes, the independent variables assessed in the models were: Age at CI 1, CI interval, Non-verbal IQ, and Maternal Education. These variables were selected due to their statistically significant Pearson product correlations.

The results of the regression models are summarized in Table 2. These models account for 40% (PPVT outcome) and 32% (CELF outcome) of the variance in data, and the pattern of results is the same for these two language outcomes, with one exception. Overall, Age at CI 1 and Non-verbal IQ are significant predictors of PPVT and CELF, while CI interval is not. The exception is that Maternal Education is also a significant predictor of receptive vocabulary (PPVT) scores, but is not for receptive language (CELF) scores. Shown in Figure 1 is an illustrative example of the dependence of receptive vocabulary (PPVT) scores on Age at CI 1, but not on CI interval. The results of the regression models remain unchanged when the nine participants who received sequential CIs, but did not use a HA during the CI interval, are excluded.

DISCUSSION

CI interval is not statistically related to receptive vocabulary nor to receptive language skills for these 88 children who nearly all used bimodal devices during the CI interval. This result contrasts, in kind, with those that indicate shorter CI intervals are associated with better speech perception and localization[32-37]. This apparent discrepancy might be due to differences in bilateral device use amongst the pediatric populations (with severe-to-profound bilateral hearing loss) in these studies. Children who do not use bilateral hearing devices before receiving their 2nd CI would experience bilateral auditory deprivation *during* the CI interval. That is, CI interval would be highly correlated with duration of bilateral auditory deprivation. In this study, however, most of the children wore two devices since the age at which they received their 1st CI. Twenty-three children were simultaneously implanted (CI interval is '0') and hence wore bilateral CIs since the time of their initial CI activation. Of the 65 sequentially-implanted children, a large majority (86%) used two devices, a CI and a HA (at the non-implanted ear), during their CI interval. Our result (CI interval is not related to language outcomes) and our pediatric population's history of using two devices contrast with those in the two previous studies of language abilities in pediatric bilateral CI recipients[38, 39]. Both studies report better language outcomes with shorter CI

intervals. However, Sarant and colleagues[39] reported that only 32% of their children wore a HA during the CI interval. Boons and colleagues[38] do not specify HA use during the CI interval: bilateral stimulation, or the lack thereof, is not addressed during the CI interval. Thus, the statistically insignificant effect of CI interval on language in this study might be attributable to 'prevented' bilateral auditory deprivation in most of the 88 children in this study. HA use in the non-implanted ear may maintain neural pathways for subsequent CI use, and, in turn, allow for better receptive language skills. This idea is consistent with reports of better language skills[25, 26], verbal cognition[46], speech perception[8, 10], and localization[8, 47] for CI users with contralateral stimulation (via either a 2nd CI or a HA) than for CI users without contralateral stimulation (i.e., bilateral devices are better than a unilateral device).

This study's results also indicate that receiving CI 1 early is associated with better receptive vocabulary and receptive language, consistent with those of many others[1-5]. That is, this study confirms that an earlier age at CI 1 is related to better receptive vocabulary and receptive language skills. Also consistent with previous studies, non-verbal IQ was found to be a significant predictor of receptive language skills[39, 48-50]. Finally, in this study maternal education was a significant predictor of PPVT scores, but not of CELF scores. The reason for slightly different results for the two receptive language outcomes is unclear. Perhaps the intensive therapy received by most participants and the relatively high level of maternal education in this sample (mean = 16 years) had an effect on receptive vocabulary, but not on receptive language.

The results of this current study emphasize the importance of receiving a CI at an early age, as earlier age at CI 1 was significantly related to better receptive vocabulary and receptive language skills. For the 88 pediatric participants in this sample, CI interval was not significantly related to receptive vocabulary and receptive language skills. Notably, as a group, the vast majority (~90%) of these pediatric bilateral CI users wore two hearing devices for most of their hearing history. They were either simultaneously implanted with bilateral CIs or used bimodal devices during their CI interval (i.e., the time between CI 1 and CI 2). Consequently, auditory stimulation was preserved at each ear, possibly preventing bilateral auditory deprivation. Conceivably, CI interval may not matter, if the non-implanted ear uses a HA with benefit during the interval. Future studies should examine the effects of degree of hearing and bilateral device configuration (i.e., bimodal devices versus bilateral CIs) on a variety of perceptual abilities, and ultimately on receptive vocabulary and language skills.

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Figure 1:
Scatter plots of PPVT Standard Score (receptive vocabulary) vs. Age at CI 1 (Panel A), and
PPVT Standard Score (receptive vocabulary) vs. CI interval (Panel B).

Demographic and audiologic information, and language outcomes for bilateral CI participants, reported as, mean (SD) [min - max], when appropriate.

Table 1:

DEMOGRAPHIC		<u>All Bilateral CI users</u>	
No. of Participants		88	
No. of Female Participants		42	
Age at Test (years)		7.0 (1.2) [4.8 - 9.0]	
Non-verbal IQ		99.0 (15.3) [65 - 140]	
Maternal Education (years)		16.0 (2.2) [11 - 20]	
LANGUAGE OUTCOMES			
PPVT Receptive Vocabulary (standard scores)		93.4 (17.1) [56 - 138]	
CELF Receptive Language (standard scores)		91.5 (19.6) [51 - 131]	
AUDIOLOGIC			
Age at 1 st HA (months) [n = 87]		10.0 (7.8) [1 - 28]	
Unaided pre-CI PTA (0.5, 1, 2 kHz) (dB HL)		100.4 (15.0) [50 - 125]	
CI Manufacturer {Cochlear, AB, Med-El}		{68, 17, 3}	
BILATERAL-DEVICES HISTORY			
No. of Participants		88	23
Age at 1st CI (months) [CI 1]		21.6 (11.3) [8 - 55]	16.5 (6.4) [8 - 33]
Age at 2nd CI (months) [CI 2]		31.6 (17.2) [8 - 80]	16.5 (6.4) [8 - 33]
CI [surgery] interval (months)		10.0 (11.8) [0 - 67]	0
No. of Participants who used a HA at the non-implanted ear (during CI surgery interval)		--	N/A
Duration of HA use at the non-implanted ear during CI [surgery] interval (months) [n=56]		--	N/A
			11.2 (8.6) [0.3 - 36]

Table 2:

Results of linear regression models to predict receptive vocabulary (PPVT) and receptive language (CELF) outcomes. Coefficient 'B'; [95% confidence interval for 'B']; and p values are given for statistically significant predictors. NS indicates non-statistical significance.

Independent Variable	OUTCOME	
	PPVT	CELF
Age at CI 1	-0.37; [-0.63, -0.11]; < 0.05	-0.34; [-0.65, -0.03]; < 0.05
CI Interval	NS	NS
Non-verbal IQ	0.51; [0.32, 0.71]; < 0.01	0.66; [0.43, 0.89]; < 0.01
Maternal Education	1.34; [0.04, 2.72]; < 0.05	NS
% variance	40	32