

# Early Sign Language Exposure and Cochlear Implantation Benefits

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abstract

**BACKGROUND:** Most children with hearing loss who receive cochlear implants (CI) learn spoken language, and parents must choose early on whether to use sign language to accompany speech at home. We address whether parents' use of sign language before and after CI positively influences auditory-only speech recognition, speech intelligibility, spoken language, and reading outcomes.

**METHODS:** Three groups of children with CIs from a nationwide database who differed in the duration of early sign language exposure provided in their homes were compared in their progress through elementary grades. The groups did not differ in demographic, auditory, or linguistic characteristics before implantation.

**RESULTS:** Children without early sign language exposure achieved better speech recognition skills over the first 3 years postimplant and exhibited a statistically significant advantage in spoken language and reading near the end of elementary grades over children exposed to sign language. Over 70% of children without sign language exposure achieved age-appropriate spoken language compared with only 39% of those exposed for 3 or more years. Early speech perception predicted speech intelligibility in middle elementary grades. Children without sign language exposure produced speech that was more intelligible (mean = 70%) than those exposed to sign language (mean = 51%).

**CONCLUSIONS:** This study provides the most compelling support yet available in CI literature for the benefits of spoken language input for promoting verbal development in children implanted by 3 years of age. Contrary to earlier published assertions, there was no advantage to parents' use of sign language either before or after CI.



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"Deaf" is capitalized here because it is customary when referring to people who culturally identify as deaf and embrace the values of the Deaf Community.

Dr Geers conceptualized and designed the analysis and drafted the initial manuscript; Ms Mitchell operationalized participant selection and definitions of key concepts through systematic retrieval of data from the CDaCI database, conducted all analyses, and reviewed and revised the manuscript; Dr Warner-Czyz provided speech intelligibility estimates, conducted initial comparisons of speech perception and production results, and reviewed and revised the manuscript; Dr Wang, principal investigator for the CDaCI Data Coordinating Center, developed the Speech Recognition Index in Quiet based on the speech perception hierarchy, provided guidance on statistical analytic approach, reviewed all analyses, and reviewed and revised the manuscript; Dr Eisenberg, principal investigator for the CDaCI project, conceived the speech perception hierarchy and reviewed and revised the manuscript; and all authors approved the full manuscript as submitted.

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**WHAT'S KNOWN ON THIS SUBJECT:** Cochlear implant use, even from a young age, does not insure that spoken language will develop normally. Controversy exists regarding whether sign language in combination with spoken language provides greater benefit from a cochlear implant than spoken language alone.

**WHAT THIS STUDY ADDS:** Outcomes were compared for early-implanted children from a prospective, national cohort differing in amount and duration of sign language use. Children exposed to sign language performed more poorly on auditory-only speech recognition, speech intelligibility, spoken language, and reading outcomes.

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In a national sample of more than 27 000 school-aged children who were deaf or hard of hearing, Mitchell and Karchmer<sup>1</sup> reported only 3.9% had 2 parents who were deaf or hard of hearing. Most parents with normal hearing would like their child who is deaf to learn to communicate using spoken language and choose to provide a cochlear implant (CI) to facilitate this outcome. A major question for parents and the professionals who work with them is whether speech recognition, speech production, spoken language, and reading skills are best developed by focusing exclusively on spoken language or if early exposure to sign language provides an important foundation for learning a spoken language. Children of Deaf\* parents are assumed to learn American Sign Language (ASL) at a normal rate through natural exposure to their parents' language.<sup>2,3</sup> However, most hearing parents and teachers are not ASL-proficient and typically learn an English-based sign language system that accompanies speech and maintains English word order and morphology, often referred to as "total" or "simultaneous" communication.<sup>4</sup> Proponents of this approach maintain that signing while talking aids development of spoken language and reading skills.

For example, a recent review in *Pediatrics*<sup>2</sup> noted "The benefits of learning sign language clearly outweigh the risks. For parents and families who are willing and able, this approach seems clearly preferable to an approach that focuses solely on oral communication." Despite these assertions, there is a paucity of data directly comparing spoken language outcomes in similar groups of children learning language with and without the addition of sign. A systematic review of studies

conducted between 1995 and 2013 concluded that "insufficient high-quality evidence exists to determine if sign language in combination with oral language is more effective than oral language therapy alone"<sup>5</sup>. The question we address is whether parents' use of sign language before and after cochlear implantation positively influences auditory-only speech recognition, speech intelligibility, spoken language, and reading outcomes.

1. Does an early exclusive focus on spoken language promote faster development of auditory speech recognition skills, leading to more intelligible speech in elementary grades?
2. Does early exposure to sign language in addition to speech promote the development of spoken language and reading in elementary grades?
3. Is the quantity of sign language exposure over the first 3 years postimplant important for age-appropriate spoken language and reading development?

## METHODS

### Participants

Ninety-seven children were selected from the Childhood Development after Cochlear Implantation (CDaCI) study,<sup>6</sup> a prospective multicenter, national cohort of 188 children recruited from 6 large CI centers between November 2002 and December 2004. Institutional Review Board approval of the protocol was obtained at each of the test sites (see Acknowledgments), and written informed consent was obtained from each participating family. The sample for this study was selected as follows: CI activated by 38 months of age ( $n = 137/188$ ), parents who consistently reported the communication mode used with their child ( $N = 129/137$ ), and returned for testing near early and late elementary grades

( $n = 97/129$ ). Over 95% of the children received specialized intervention during some or all of the first 3 years postimplant. The selected sample had statistically significantly younger age at implant activation (21.8 vs 37.3 months) and a higher percentage of families with white race (70% vs 55%) and with maternal education level at college graduate or higher (57% vs 42%) compared with those 91 families in the CDaCI database who were not included in these analyses.

### Sign Exposure Classification

Parents answered written questions about their child's exposure to sign language at baseline (just before CI surgery) and at 12, 24, and 36 months postimplant. A child was classified as negative for sign language exposure at a given rating period if the parent reported no sign language use by the parent or intervention program (ie, auditory-oral, auditory-verbal, or cued speech methodologies). A child was classified as positive for sign language exposure at that rating period if one of the following systems was reported by a parent as used at least 10% of the time at home and/or in the child's intervention program: ASL, Total/Simultaneous Communication, baby sign, Signing Exact English, Signed English, sign language, sign support, or Pidgin sign. Sign language exposure categorization was determined as follows: No sign = no report of parent or intervention program sign language use from baseline through the first 3 years postimplant ( $N = 35$ ); Short-term sign = positive for sign language use at baseline and/or 12 months postimplant but negative for sign language use at 24 and 36 months postimplant ( $N = 26$ ); Long-term sign = positive for sign language use at baseline and/or 12 months and at 24 and 36 months postimplant ( $N = 36$ ). Similar rates of sign language use at baseline were reported for the

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**TABLE 1** Baseline Characteristics of CI Recipients by Sign Language Exposure Group

Characteristic	No Sign ( <i>n</i> = 35)	Short-term Sign ( <i>n</i> = 26)	Long-term Sign ( <i>n</i> = 36)	Total ( <i>n</i> = 97)
Girl, <i>n</i> (%)	17 (49)	9 (35)	21 (58)	47 (48)
Household income, <i>n</i> (%) < \$50 k	11 (32)	11 (44)	15 (42)	37 (39)
Maternal education, <i>n</i> (%) graduated college	24 (69)	13 (50)	18 (50)	55 (57)
Parent-infant program, <i>n</i> (%)**	14 (40)	20 (77)	23 (64)	57 (59)
Aided PTA better ear, dB <sup>a</sup>	75.1 (22.0)	73.1 (23.6)	77.8 (21.8)	75.6 (22.2)
Age at onset of deafness, mo	0.3 (1.2)	1.2 (3.4)	1.3 (3.6)	0.9 (2.9)
Amplification age, mo	9.4 (8.6)	10.8 (8.3)	11.5 (7.5)	10.6 (8.1)
Activation age, mo	19.3 (8.3)	22.1 (7.3)	22.8 (8.3)	21.4 (8.1)
Maternal sensitivity <sup>b</sup>	5.5 (0.7)	5.3 (0.7)	5.4 (0.7)	5.4 (0.7)
Baseline IQ <sup>c</sup>	94.5 (19.3)	97.4 (21.2)	98.5 (14.2)	96.8 (18.1)
Vocabulary <sup>d</sup>	14.7 (41.6)	10.8 (18.4)	16.2 (59.4)	14.2 (44.6)
Auditory perception <sup>e</sup>	9.8 (9.4)	7.0 (6.9)	5.8 (7.3)	7.5 (8.1)

Data are expressed as mean (SD) unless otherwise noted. PTA, pure-tone average.

<sup>a</sup> Average of available thresholds for tested frequencies 500, 1000, 2000, and 4000 Hz, where at least 1 frequency was tested (88/97 of the participants had 4-frequency pure-tone average).

<sup>b</sup> Maternal sensitivity scale from the Eunice Kennedy Shriver National Institute of Child Health and Human Development Early Childcare Study codes.

<sup>c</sup> Bayley Scales of Infant Development (BSID II) (Bayley<sup>9</sup>).

<sup>d</sup> Spoken words both understood and said on the MacArthur-Bates Communicative Development Inventory (MBCDI: Words and Gestures Form; Fenson et al<sup>10</sup>).

<sup>e</sup> Infant-Toddler Meaningful Auditory Integration Scale (IT-MAIS) (Robbins et al<sup>11</sup>).

\*\*  $\chi^2$  *P* value = .01.

40 children who met the implant age criteria but were excluded due to lack of sufficient follow-up data.

Parents were also asked to estimate how much of each day sign language was used in the home, to separate frequent signers (>50% of the day) from infrequent signers (<50%). In families providing long-term sign language exposure, the proportion of frequent signers decreased from 63% at baseline or 12 months postimplant to 29% at 24 or 36 months postimplant.

### Preimplant (Baseline) Characteristics

Table 1 summarizes sample characteristics at baseline. No statistically significant group differences emerged for sex, family income, percentage of mothers with college degrees, age at onset of deafness, age first aided, average aided hearing threshold, age at first CI activation, maternal sensitivity to communicative interactions,<sup>7,8</sup> nonverbal cognition,<sup>9</sup> spoken words rated as both understood and produced,<sup>10</sup> or auditory perceptual skills.<sup>11</sup> Families of children who used sign language were statistically significantly more likely to be enrolled in parent–infant

intervention preimplant than nonsigning families (*P* = .01).

### Postimplant Outcome Measures

Auditory development was tracked over the first 3 years after CI activation. Speech intelligibility was measured near the middle of elementary school (age = 6.0–8.9 years). Spoken language and reading outcomes were examined at a point near early (age = 5.0–7.9) and near late (age = 9.0–11.9) elementary grades. Tests were administered by certified audiologists and speech-language pathologists at each CI Center without previous knowledge of group assignment for this study.

#### Early Auditory Development

The Speech Recognition Index in Quiet (SRI-Q) combines multiple results from a hierarchical test battery into a single cumulative speech perception index, accounting for both the difficulty level and accuracy on a specific test.<sup>12</sup> SRI-Q values range from 0 to 600, with lower scores (0–100) representing parent report on the Meaningful Auditory Integration Scale.<sup>11,13</sup> Midrange scores (101–300) represent closed-set word recognition: Early Speech Perception Test<sup>14</sup> and Pediatric Speech

Intelligibility Test.<sup>15</sup> Highest values (301–600) delineate open-set speech recognition: Lexical Neighborhood Test,<sup>16</sup> Phonetically-Balanced Word Lists-Kindergarten,<sup>17</sup> and Hearing in Noise Test for Children (administered in quiet).<sup>18</sup>

#### Speech Intelligibility

Audio recordings were made of each child imitating 36 sentences (3, 5, or 7 syllables long).<sup>19</sup> Each sentence contained a key word that was either predicted by context (Read the book) or not (Get the cake). Normal hearing adults with no previous experience listening to the speech of individuals who are deaf were instructed to write down as much of the sentence as they understood. Three judges provided responses to each sentence, and no judge listened to more than 1 sentence from the same child. Each overall intelligibility score represents the percent of 36 key words correctly understood across a total of 108 judgments.

#### Spoken Language

The Core Composite standardized score (SS) on the Comprehensive Assessment of Spoken Language (CASL)<sup>20</sup> was used to assess language in relation to hearing age-mates in the normative sample. All children

**TABLE 2** Median (and interquartile range) of Speech Recognition in Quiet (SRI-Q) Scores at Baseline and Over the First 3 y of CI Use by Sign Language Exposure Group

Time of Assessment	No Sign ( <i>n</i> = 35)	Short-term Sign ( <i>n</i> = 26)	Long-term Sign ( <i>n</i> = 36)	Total ( <i>n</i> = 97)	<i>P</i> <sup>a</sup>
Baseline (pre-CI)	16.3 (5.0–50.0)	15.0 (2.5–22.5)	7.5 (1.3–18.8)	12.5 (2.5–30.0)	.11
12 mo post-CI	150.0 (90.0–223.0)	125.0 (72.5–250.0)	125.0 (80.0–200.0)	125.0 (85.0–200.0)	.39
24 mo post-CI	300.0 (260.0–365.0)	343.5 (273.0–359.0)	286 (125.0–359.5)	313.0 (150.0–359.3)	.25
36 mo post-CI	394.0 (341.0–556.0)	381.8 (351.0–444.0)	354.0 (291.0–392.0)	374.0 (334.5–509.0)	.03 <sup>b</sup>

<sup>a</sup> Kruskal-Wallis equality-of-populations rank test.

<sup>b</sup> Significant value (*P* = .03)

received the “core” language subtests appropriate for their age, drawn from the antonyms, syntax construction, paragraph comprehension, nonliteral language, pragmatic judgment, grammatical morphemes, and sentence comprehension subtests.

### Reading

The Passage Comprehension subtest of Woodcock-Johnson Tests of Achievement (WJ)<sup>21</sup> measures understanding of printed words and phrases at the early elementary level and paragraph comprehension at later grades. Results are expressed in SS in relation to hearing age-mates in the normative sample.

### Statistical Analyses

Pre- and postimplant characteristics and outcomes were compared across the 3 sign language exposure groups. Continuous variables were compared using analysis of variance F- or Kruskal-Wallis (when scores were not normally distributed or in interval scale) tests, and categorical variables were compared by using  $\chi^2$  or Fisher’s exact tests. Because the SRI-Q is an ordinal hierarchy created by scaling the different tests from 0 to 100 and stacking them, Kruskal-Wallis tests were used to compare the groups. Dunn’s test was used for posthoc comparison between groups. Spearman’s rank correlation  $\rho$  was used to correlate SRI-Q scores with speech intelligibility. Unadjusted logistic regressions were used to compare odds of language and reading outcomes below clinical thresholds (SS <85) for those exposed to short- or long-term sign to the no sign group. Stata 11

(StataCorp, College Station, TX) was used for all analyses, and all tests were 2-sided with  $\alpha$  = .05.

## RESULTS

### Does an Early Exclusive Focus on Spoken Language Promote Faster Development of Auditory Speech Perception Skills, Leading to More Intelligible Speech in Elementary Grades?

Median SRI-Q at baseline and over the first 3 years of CI use is reported in Table 2. A statistically significant difference between sign language exposure groups was identified at 36 months postimplant. Posthoc pairwise comparisons revealed a statistically significant advantage of the no sign group over the long-term sign language exposure group (*P* = .004).

Speech intelligibility scores obtained at a mean age of 8.2 years (SD = 0.7) statistically significantly differed for the 3-sign exposure groups (*P* < .001). Dunn’s pairwise posthoc comparison revealed that children with long-term sign exposure produced statistically significantly less intelligible speech (mean = 50.9%; SD = 23.5) than those with no sign exposure (mean = 70.4%; SD = 16.5; *P* < .001) and those with short-term sign exposure (mean = 63.0%; SD = 20.4; *P* = .01).

SRI-Q at baseline did not predict later speech intelligibility ( $\rho$  = 0.10). However, the rank correlation between post-CI speech recognition and later production was statistically significant at each

of the 3 postimplant intervals and increased over time ( $\rho$  = 0.24, *P* = .02 at 12 months;  $\rho$  = 0.32, *P* = .002 at 24 months; and  $\rho$  = 0.42, *P* < .001 at 36 months post-CI).

### Does Early Exposure to Sign Language in Addition to Speech Promote the Development of Spoken Language and Reading in Elementary Grades?

Language (CASL core composite) and reading comprehension (WJ) passage comprehension) SS near early and late elementary grades are plotted in Fig 1 for each of the 3 sign language exposure groups. Table 3 presents mean standard scores of the no sign language group and the average difference from this mean observed in the short-term and long-term groups. Posthoc tests compared the mean difference of the unadjusted linear regression for the 2 sign language exposure groups to the no sign group.

### Language

Language scores statistically significantly differed among sign language exposure groups at both test intervals, and statistical significance increased over time (early *P* = .04; late *P* = .002). Table 3 presents posthoc comparisons in early and late elementary grades. The no sign language exposure group scored statistically significantly higher than the long-term exposure group in spoken language at both tests and scored statistically significantly higher than the short-term exposure group in later grades. By late elementary grades, the average language score of children

without sign language exposure was 96.2, close to the normative mean of 100, whereas mean scores for the groups with sign language exposure remained delayed (83.8 and 76.4 for the short- and long-term exposure groups, respectively).

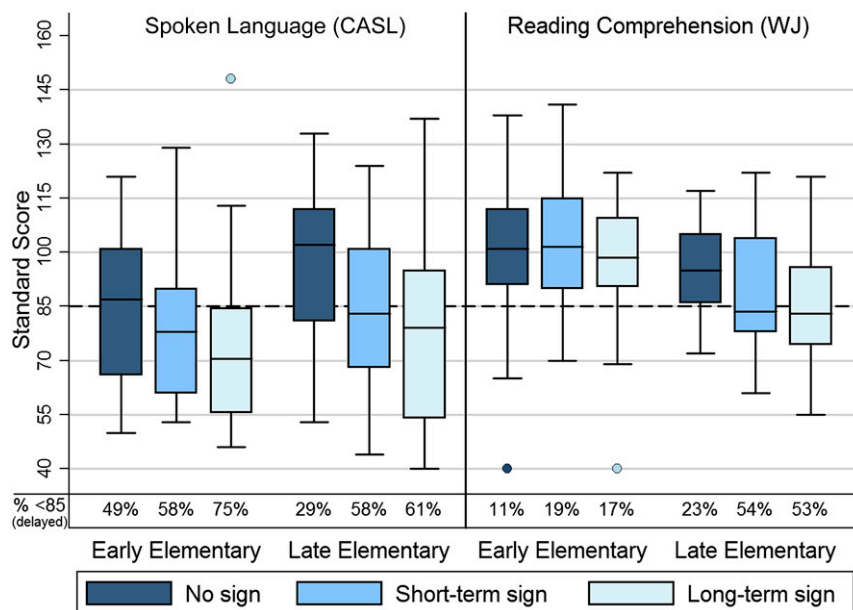
### Reading

All 3 CI groups achieved comprehension scores on par with hearing children in the early elementary years, with no group differences reaching statistical significance. However, children without sign language exposure (mean SS = 94.9) exhibited a statistically significant reading advantage over the long-term sign language group (mean SS = 86.0) in later elementary grades ( $P = .02$ ).

### Is the Quantity of Sign Language Exposure Over the First 3 Years Postimplant Important for Age-Appropriate Language and Reading Development?

#### Short-term Versus Long-term Exposure

The bottom row of Fig 1 reports the percentage of children in each group scoring more than 1 SD less than their normal hearing age-mates, and the lower section of Table 3 compares the odds of children in the sign language groups exhibiting delays in spoken language



**FIGURE 1**

Language and reading scores of CI recipients by sign language exposure group near early and/or late elementary grades are depicted with box plots (25th, median, 75th percentiles; whiskers extend to highest and lowest value within  $\pm 1.5 \times$  interquartile range). Percentages of children  $>1$  SD less than the normative mean are listed at the bottom.

or reading relative to the no sign group. Between early and late elementary grades, the percentage of children with delayed language decreased from 49% to 29% in the no sign language exposure group, remained constant at 58% in the short-term group, and decreased from 75% to 61% in the long-term group. The percentage of children with less-than-average reading

scores increased from  $<20\%$  in early elementary grades to over 50% in late elementary grades in the groups exposed to sign language. For children without sign language exposure, only 11% were delayed in early elementary grades, increasing to 23% in late elementary grades. By late elementary school, participants exposed to sign language, regardless of short- or long-term exposure,

**TABLE 3** Mean Difference From Unadjusted Linear Regression and Odds Ratio (OR) From Unadjusted Logistic Regression Comparing CI Recipients Exposed to Sign Language to the No Sign (Reference) Group

Standard Scores	No Sign (Ref)		Short-term Sign vs No Sign (Ref)			Long-term Sign vs No Sign (Ref)		
	Mean SS	Mean Difference	95% Confidence Interval	<i>P</i>	Mean Difference	95% Confidence Interval	<i>P</i>	
<b>Spoken language</b>								
Early elementary	85.6	-8.15	-18.65 to 2.35	.13	-12.63	-22.25 to -3.00	.01	
Late elementary	96.2	-12.33	-24.20 to -0.45	.04	-19.81	-30.70 to -8.92	<.001	
<b>Reading comprehension</b>								
Early elementary	101.1	0.11	-9.11 to 9.33	.98	-3.20	-11.65 to 5.26	.46	
Late elementary	94.9	-6.51	-14.18 to 1.16	.10	-8.83	-15.86 to -1.80	.01	
<b>Delayed (SS &lt;85)</b>								
		OR	95% Confidence Interval	<i>P</i>	OR	95% Confidence Interval	<i>P</i>	
<b>Spoken language</b>								
Early elementary		1.44	0.52 to 4.01	.48	3.18	1.16 to 8.67	.02	
Late elementary		3.41	1.17 to 9.93	.03	3.93	1.45 to 10.61	.007	
<b>Reading comprehension</b>								
Early elementary		1.85	0.44 to 7.69	.40	1.55	0.40 to 6.05	.53	
Late elementary		3.94	1.31 to 11.87	.02	3.77	1.35 to 10.51	.01	

had more than 3 times higher odds of having less-than-average (SS <85) spoken language or reading comprehension scores compared with participants not exposed to sign language.

### *Frequent Versus Infrequent Sign Language Exposure*

No statistically significant differences were observed based on quantity of sign language exposure when collapsed across sign language exposure groups. Spoken language scores did not statistically significantly differ between children from families reporting use of sign language 10% to 50% of the day ( $N = 28$ ;  $SS = 75$  early CASL; 78 later CASL) and those whose family reported  $\geq 50\%$  sign language use ( $N = 33$ ;  $SS = 76$  early CASL; 82 later CASL). Similarly, there was no statistically significant reading comprehension advantage in children whose parents used sign language infrequently ( $SS = 99$  early WJ; 85 later WJ) compared with those with frequent sign language exposure ( $SS = 100$  early WJ; 90 later WJ).

## **DISCUSSION**

Three groups of children who differed in the amount and duration of early sign exposure provided in their homes and/or intervention programs were compared in their post-CI progress through elementary grades. Data were analyzed to address 3 questions. Below, we summarize and interpret these results.

### **Does an Early Exclusive Focus on Spoken Language Promote Faster Development of Auditory Speech Perception Skills, Leading to More Intelligible Speech in Elementary School?**

Children whose families used spoken language exclusively developed better auditory speech recognition skills after 3 years of CI use and had more intelligible speech than children whose families used sign language.

A strong relationship that increased over time was documented between early speech recognition and later speech intelligibility. Previous studies have documented a relation between the perception and production of speech sounds in children with CIs,<sup>22–24</sup> and more intelligible speech has been associated with oral-only instruction.<sup>23,25</sup> The current findings further suggest that parental sign language use from an early age, if continued after receipt of a CI, is associated not only with slower development of speech recognition, but also with substantially less intelligible speech in elementary grades (50% in children of long-term signers compared with 70% for children of nonsigning parents). Although short-term use of sign language did not enhance development, it did not appear to have deleterious effects on either speech perception growth or later speech intelligibility.

It is likely that parents in the long-term group continued to use sign language because their child was slow to develop speech perception abilities, and families in the short-term group stopped using sign language because their child's auditory gains made the use of sign language unnecessary. Nonsigning families did not report switching to sign language use later, presumably because their child's listening and spoken language skills continued to develop. Although the groups appeared well-matched initially with similar auditory and vocabulary skills preimplant and no speech perception differences for the first 2 years postimplant, it is possible that use of sign language interfered with auditory and speech development.

### **Does Early Exposure to Sign Language in Addition to Speech Promote the Development of Spoken Language and Reading in Elementary Grades?**

Spoken language development is negatively affected by delaying access to linguistic input until auditory input is initiated through hearing

aids and/or CIs. Proponents of early sign language use assert that children with Deaf parents who are exposed to ASL from birth have a firmer foundation for the development of spoken language once the CI is activated, although empirical data supporting this conclusion are limited.<sup>26</sup> Most hearing parents do not know sign language when their child is diagnosed with hearing loss, and acquiring proficiency is a long and arduous process for them. In this study, early exposure to sign language did not enhance either spoken language or reading. In fact, children whose parents signed were statistically significantly more likely than children of nonsigning parents to exhibit spoken language delays in elementary grades and to fall behind age-mates in reading comprehension by late elementary grades. Long-term parental signing was associated with greater delay throughout elementary grades, and children from families who discontinued signing after a year of CI use still were 3.5 to 4 times more likely than nonsigners to score more than 1 SD less than age-mates in the normative sample in spoken language and reading near the end of elementary school.

These results are in line with previous findings from a nationwide sample of 181 children who received a CI between 1.6 and 5.3 years of age and were tested when they were 8 to 9 years old.<sup>27</sup> Each child's communication mode was ranked to reflect educational emphasis on spoken versus sign language input beginning preimplant and continuing into elementary grades. Children enrolled in an educational environment that emphasized spoken language and minimized accompanying signs exhibited a statistically significant language advantage over children enrolled in sign language programs.<sup>28</sup> As in the current study, the effect of communication mode on reading comprehension was not statistically

significant in early elementary grades.<sup>29</sup> However, 8 years later, when 112 of the original 181 participants returned for assessment (ages 15.0–18.5 years),<sup>30</sup> those students who continued to rely on sign language in their teenage years had statistically significantly worse overall English language outcomes<sup>31</sup> as well as overall literacy levels.<sup>32</sup>

The relatively high proportion of children in the no sign language exposure group achieving scores within 1 SD of normal hearing age-mates replicated results observed previously for a nationwide sample of 60 children who had no early sign language exposure, received a CI within the same age range (ie, by 38 months), and were approximately the same age at assessment near early and late elementary grades.<sup>33</sup> The percentage of children exhibiting a language delay that persisted through elementary grades was strikingly similar in these 2 studies (29% and 32%), indicating generalizability of this result.

Parents in the long-term exposure group may have continued signing with their children because of their children's lack of spoken language progress, and sign language skills (not measured here) may have excelled. Measuring only spoken language outcomes may have underestimated total language abilities in spoken and signed language together. However, increasing lags in reading comprehension scores of children exposed to sign language suggest that their overall language skill was not sufficient to compensate for verbal achievement deficits.

### **Is the Quantity of Sign Language Exposure Over the First 3 Years Post-Implant Important for Age-Appropriate Spoken Language and Reading Development?**

To examine this question, we first compared outcomes of children with short-term and long-term

exposure to sign language with those from nonsigning families. Children with long-term sign language exposure were at a significant disadvantage compared with those from nonsigning families across all outcomes, whereas short-term exposure was associated with spoken language and reading delays that emerged only in late elementary grades. This result suggests a sensitive period may exist for early sensory experience and a focus on early auditory input capitalizing on phonologically relevant articulatory events plays an important and persisting role in verbal development.<sup>34</sup>

Second, we compared children in families with frequent parental sign language use with those of infrequent signers. Children whose parents reported using sign language more frequently did not achieve better outcomes than those of less frequent signers. It is possible that the sign exposure provided by these hearing parents was not sufficient to promote spoken language development. The diminished performance of children of hearing parents learning sign may not adequately represent the potential benefits of early sign language input from accomplished signers.<sup>26</sup> On the other hand, when this issue was addressed in “Language Choices for Deaf Infants: Advice for Parents Regarding Sign Languages,”<sup>35</sup> parents were encouraged to sign regardless of their skill level:

*[P]arents do not have to be perfect language models or even very good language models...even if not fluent, the parents' language use is still important to the language development of the child... When a hearing mother signs with her deaf child, the child shows early language expressiveness on a par with hearing peers regardless of her signing abilities (p2).*

Results of the current investigation indicate that hearing parents' attempts to expose their child to sign language more frequently or for longer periods of time did not benefit,

and may have detracted from, development of auditory, speech, and spoken language skills. However, the proportion of parents using sign language more than half of the day decreased from 63% at baseline or 12 months post implant to 29% at 24 or 36 months postimplant. We do not know whether more intensive use of sign language would have had different outcomes.

## **CONCLUSIONS**

These results shed new light on a number of assertions regarding the benefits of early sign exposure cited in a review by Napoli et al.<sup>2</sup> (1) “[E]arly sign language, when used for a short time preimplant as a bridge to spoken language, cannot hurt and may be beneficial.” Current results indicate no lasting advantage to using sign before and immediately after a CI, and these children were more likely to experience delayed language and reading in late elementary grades than children with no sign exposure. (2) “With sign language, the deaf child is able to travel through various social situations and communities without difficulty and not be confined to communicating only with family and friends, as is often the case for deaf children who have no knowledge of sign language.” Children not exposed to sign language developed speech that was, on average, 70% intelligible to hearing listeners, suggesting they can use speech to communicate effectively in the wider hearing world. Children whose families signed for the first 3 years after CI averaged considerably less intelligible speech (50%), which likely affects the ease of spoken communication. (3) “[S]igning deaf children, with or without a CI, perform better on literacy skills.” Children without sign language scored significantly better in reading in late elementary grades compared with children whose families provided early exposure to sign language.

Some caveats apply when interpreting these results. First, children in the current study were all from families with normal hearing who were not native signers, and at least some children with Deaf parents who used ASL have reportedly achieved age-appropriate spoken language,<sup>26</sup> as did some children with sign exposure in the current investigation. Second, although there were no differences among the 3 groups of parents and children on a number of key predictors measured before cochlear implantation, differences may have existed that were not measured here. Third, more than half of mothers in this study had college degrees, and results may not apply equally to less advantaged populations. Nevertheless, based on results from this nationwide sample of children who differed in amount of early sign language exposure, if the long-term development of spoken English communication and literacy is the primary objective for a child with a CI, focus on early spoken input increases the probability of achieving those goals.

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## ABBREVIATIONS

ASL: American Sign Language

CASL: Comprehensive Assessment of Spoken Language

CDAci: Childhood Development after Cochlear Implantation

CI: cochlear implant

SRI-Q: speech recognition index in quiet

SS: standardized score

WJ: Woodcock-Johnson Tests of Achievement

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## REFERENCES

1. Mitchell R, Karchmer M. Chasing the mythical ten percent: parental hearing status of deaf and hard of hearing students in the United States. *Sign Lang Stud*. 2004;4(2):138–163
2. Napoli DJ, Mellon NK, Niparko JK, et al. Should all deaf children learn sign language? *Pediatrics*. 2015;136(1):170–176
3. Humphries T, Kushalnagar P, Mathur G, et al. Ensuring language acquisition for deaf children: what linguists can do. *Language*. 2014;90(2):e31–e52



4. Spencer L, Tomblin B. Speech production and spoken language development of children using “total communication”. In: Spencer P, Marschark M, eds. *Advances in Spoken Language Development of Deaf and Hard-of-Hearing Children*. New York, NY: Oxford University Press; 2006:166–192
5. Fitzpatrick EM, Hamel C, Stevens A, et al. Sign language and spoken language for children with hearing loss: a systematic review. *Pediatrics*. 2016;137(1):e20151974
6. Fink NE, Wang NY, Visaya J, et al; CDaCI Investigative Team. Childhood Development after Cochlear Implantation (CDaCI) study: design and baseline characteristics. *Cochlear Implants Int*. 2007;8(2):92–116
7. NICHD Early Child Care Research Network. Chronicity of maternal depressive symptoms, maternal sensitivity, and child functioning at 36 months. *Dev Psychol*. 1999;35(5):1297–1310
8. Quittner AL, Cruz I, Barker DH, Tobey E, Eisenberg LS, Niparko JK; Childhood Development after Cochlear Implantation Investigative Team. Effects of maternal sensitivity and cognitive and linguistic stimulation on cochlear implant users’ language development over four years. *J Pediatr*. 2013;162(2):343–348.e3
9. Bayley N. *Bayley Scales of Infant Development*. 2nd ed. San Antonio, TX: The Psychological Corp; 1993
10. Fenson L, Dale P, Reznick J. *The MacArthur Communicative Development Inventories: User’s Guide and Technical Manual*. San Diego, CA: Singular Press; 1993
11. Robbins AM, Renshaw JJ, Berry SW. Evaluating meaningful auditory integration in profoundly hearing-impaired children. *Am J Otol*. 1991;12(suppl):144–150
12. Wang NY, Eisenberg LS, Johnson KC, et al; CDaCI Investigative Team. Tracking development of speech recognition: longitudinal data from hierarchical assessments in the Childhood Development after Cochlear Implantation Study. *Otol Neurotol*. 2008;29(2):240–245
13. Zimmerman-Phillips S, Robbins AM, Osberger MJ. Assessing cochlear implant benefit in very young children. *Ann Otol Rhinol Laryngol Suppl*. 2000;185:42–43
14. Moog J, Geers A. *Early Speech Perception Test*. St Louis, MO: Central Institute for the Deaf; 1990
15. Jerger S, Jerger J. *Pediatric Speech Intelligibility Test*. St Louis, MO: Auditec of St Louis; 1984
16. Kirk KI, Pisoni DB, Osberger MJ. Lexical effects on spoken word recognition by pediatric cochlear implant users. *Ear Hear*. 1995;16(5):470–481
17. Haskins H. *A Phonetically Balanced Test of Speech Discrimination for Children*. Evanston, IL: Northwestern University; 1949
18. Gelnett D, Sumida A, Nilsson M, Soli SD. Development of the Hearing in Noise Test for Children (HINT-C). In: Annual meeting of the American Academy of Audiology; April, 1995; Dallas, TX
19. McGarr NS. The effect of context on the intelligibility of hearing and deaf children’s speech. *Lang Speech*. 1981;24(pt 3):255–264
20. Carrow-Woodfolk E. *Comprehensive Assessment of Spoken Language*. Circle Pines, MN: American Guidance Service; 1999
21. Woodcock R, McGrew K, Mather N. *Woodcock-Johnson III Tests of Achievement*. Rolling Meadows, IL: Riverside Publishing Co; 2001
22. Mahshie J, Core C, Larsen MD. Auditory perception and production of speech feature contrasts by pediatric implant users. *Ear Hear*. 2015;36(6):653–663
23. Tobey EA, Geers AE, Brenner C, Altuna D, Gabbert G. Factors associated with development of speech production skills in children implanted by age five. *Ear Hear*. 2003;24(suppl 1):36S–45S
24. Tobey EA, Geers AE, Sundarajan M, Lane J. Factors influencing elementary and high-school aged cochlear implant users. *Ear Hear*. 2011;32(1):27S–38S
25. Montag JL, AuBuchon AM, Pisoni DB, Kronenberger WG. Speech intelligibility in deaf children after long-term cochlear implant use. *J Speech Lang Hear Res*. 2014;57(6):2332–2343
26. Davidson K, Lillo-Martin D, Chen Pichler D. Spoken English language development among native signing children with cochlear implants. *J Deaf Stud Deaf Educ*. 2014;19(2):238–250
27. Geers A, Brenner C. Background and educational characteristics of prelingually deaf children implanted by five years of age. *Ear Hear*. 2003;24(suppl 1):2S–14S
28. Geers AE, Nicholas JG, Sedey AL. Language skills of children with early cochlear implantation. *Ear Hear*. 2003;24(suppl 1):46S–58S
29. Geers AE. Predictors of reading skill development in children with early cochlear implantation. *Ear Hear*. 2003;24(suppl 1):59S–68S
30. Geers AE, Brenner C, Tobey EA. Article 1: long-term outcomes of cochlear implantation in early childhood: sample characteristics and data collection methods. *Ear Hear*. 2011;32(suppl 1):2S–12S
31. Geers AE, Sedey AL. Language and verbal reasoning skills in adolescents with 10 or more years of cochlear implant experience. *Ear Hear*. 2011;32(suppl 1):39S–48S
32. Geers AE, Hayes H. Reading, writing, and phonological processing skills of adolescents with 10 or more years of cochlear implant experience. *Ear Hear*. 2011;32(suppl 1):49S–59S
33. Geers AE, Nicholas J, Tobey E, Davidson L. Persistent language delay versus late language emergence in children with early cochlear implantation. *J Speech Lang Hear Res*. 2016;59(1):155–170
34. Lachs L, Pisoni DB, Kirk KI. Use of audiovisual information in speech perception by prelingually deaf children with cochlear implants: a first report. *Ear Hear*. 2001;22(3):236–251
35. Humphries T, Kushalnagar P, Mathur G, et al. Language choices for deaf infants: advice for parents regarding sign languages. *Clin Pediatr (Phila)*. 2016;55(6):513–517