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## Spoken Language Development in Children Following Cochlear Implantation

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### Abstract

**Context**—Cochlear implantation (CI) is a surgical alternative to traditional amplification (hearing aids) that can facilitate spoken language development in young children with severe-to-profound sensorineural hearing loss (SNHL).

**Objective**—To prospectively assess spoken language acquisition following CI in young children with adjustment of co-variables.

**Design, Setting, and Participants**—Prospective, longitudinal, and multidimensional assessment of spoken language growth over a 3-year period following CI. Prospective cohort study of children who underwent CI before 5 years of age (n=188) from 6 US centers and hearing children of similar ages (n=97) from 2 preschools recruited between November, 2002 and December, 2004. Follow-up completed between November, 2005 and May, 2008.

**Main Outcome Measures**—Performance on measures of spoken language comprehension and expression.

**Results**—Children undergoing CI showed greater growth in spoken language performance (10.4; [95% confidence interval: 9.6–11.2] points/year in comprehension; 8.4; [7.8–9.0] in expression) than would be predicted by their pre-CI baseline scores (5.4; [4.1–6.7] comprehension; 5.8; [4.6–7.0] expression). Although mean scores were not restored to age-appropriate levels after 3 years, significantly greater annual rates of language acquisition were observed in children who were younger at CI (1.1; [0.5–1.7] points in comprehension per year younger; 1.0; [0.6–1.5] in expression), and in children with shorter histories of hearing deficit (0.8; [0.2, 1.2] points in comprehension per year shorter; 0.6; [0.2–1.0] for expression). In multivariable analyses, greater

residual hearing prior to CI, higher ratings of parent-child interactions, and higher SES associated with greater rates of growth in comprehension and expression.

**Conclusions**—The use of cochlear implants in young children was associated with better spoken language learning than would be predicted from their pre-implantation scores. However, discrepancies between participants' chronologic and language age persisted after CI, underscoring the importance of early CI in appropriately selected candidates.

## INTRODUCTION

Young children who experience severe-to-profound sensorineural hearing loss (SNHL) face challenges in developing spoken language because of an inability to detect acoustic-phonetic cues that are essential for speech recognition, even when fitted with traditional amplification devices (hearing aids). Over half the children identified with early, severe-to-profound SNHL are treated with cochlear implantation (CI).<sup>1</sup> CI systems are comprised of an externally worn microphone and a microprocessor programmed to extract intensity, frequency and timing cues from acoustic signals. The system transforms these acoustic cues into a physiologically useful electrical code. Internally, a surgically-placed receiver relays the transmitted code to an implanted array of contacts in the cochlea to stimulate surviving auditory neurons.<sup>2</sup> With experience, children understand speech, environmental sounds, and music with varying degrees of success after CI.<sup>3–6</sup>

Intervening at early ages with CI is predicated on behavioral data suggesting language performance is more accurate the earlier children can be implanted.<sup>3–8</sup> Early implantation may take advantage of neuronal flexibility inherent in critical periods of auditory-based learning.<sup>9</sup> Safety and technical concerns regarding early CI have been addressed with continued refinements of medical and surgical approaches.<sup>10</sup> Support for early implantation, however, must be tempered until sufficient longitudinal data are available. Behavioral studies supporting CI primarily use retrospective and case-series designs<sup>11</sup> and variance in observed results is notoriously high.<sup>3–7</sup> As a consequence, criteria remain unspecified regarding the timing of CI, especially those concerning the level of hearing loss and the associated delays in verbal language that should prompt CI.

To better understand the child, family, and clinical factors that promote verbal language growth after early CI, the Childhood Development after Cochlear Implantation (CDaCI) investigative group initiated a prospective study of spoken language outcomes in a cohort of children who underwent CI prior to the age of 5 years at 6 major US implant centers.<sup>12–15</sup>

## METHODS

### Study Design and Setting

Details of the study design have been published previously.<sup>14</sup> Participants were enrolled between 11/02 and 12/04; 3-year follow-up was completed between 11/05 and 5/08. Children with SNHL were enrolled prior to CI through 6 large implant centers situated in different regions of the US. Children with normal hearing were enrolled from 2 private preschools affiliated with each of 2 implant centers. The study was approved by the centers' Institutional Review Boards and written informed consent was obtained from the parent(s) of each enrolled child.

### Study Population

For the experimental group, children under the age of 5 years with severe-to-profound SNHL were consecutively screened for CI based on the absence of medical contraindications and an inability to amplify the acoustic-phonetic cues of speech to audible

levels. Children deemed candidates for CI were enrolled based on developmental criteria described below and a willingness of their parents to participate in a longitudinal study. Inclusion of a normal-hearing group served as a reference for longitudinal assessment of language development. Normal hearing children within the age range of children undergoing CI who met the same developmental criteria were enrolled based on family willingness to participate.

Inclusion criteria for both groups required that 1) parents speak English and either plan to, or had already enrolled their child in English-speaking schools, and 2) the child attained scores within 2 SDs of the norms on the *Bayley Scales of Infant Development Motor Scale (BSID II)*<sup>16</sup> or *Leiter International Performance Scale Revised (Leiter-R)*<sup>17</sup>. Performance within 2 SDs served as an indicator that the child demonstrated cognitive and motor skills appropriate for their chronological age<sup>16, 17</sup>. Children were also excluded if they had any condition that prevented testing with the *Reynell Developmental Language Scales (RDLS)*.<sup>18,19</sup>

### Data Collection and Testing

At baseline, parents completed questionnaires on family demographics, communication, educational services, and their child's hearing and medical history. Birth and medical records were used to determine periods of normal hearing, hearing loss without intervention, and amplification prior to implantation. Residual hearing was assessed in each ear as thresholds for pure-tones at 500, 1000, 2000 and 4000 Hz. Hearing was determined for each ear, and the average threshold for the better hearing ear served as a proxy for residual hearing.

A battery of tests (Table 1) was administered at each data point. In addition to the RDLS scores, measures of speech recognition and videotaped parent-child interactions were collected and coded to assess their co-variation with RDLS performance. All measures were administered pre-CI (baseline) and at follow-up visits scheduled for 6, 12, 24 and 36 months post-CI activation. For hearing children, follow-up visits were scheduled for the same time intervals as CI, but anchored at 6 weeks after baseline to correspond to the delay imposed by post-surgical healing and activation of the implant in children who underwent CI.

### Main Outcome Measures

The RDLS<sup>18,19</sup> comprehension and expressive language scales were administered as interdependent measures of spoken language performance at baseline (pre-CI for children undergoing CI). Age-equivalent language level (language age) was determined based on RDLS scores. If language level is "on par" with the mean established by normative data from hearing children of the same age, the gap between a child's chronologic and language ages would be 0; if scores match those expected of a child 1 year younger, the gap is 1 year.

Secondary measures addressed emergent speech recognition and characteristics of parent-child interactions. Speech recognition was represented by the Speech Recognition Index (SRI) (Table 1) summarizing the scores collected from a hierarchical battery of speech recognition tests and developed specifically for this study to track speech recognition performance via growth curve analysis.<sup>13,20</sup> Videos of parent-child interactions were coded for respect for child autonomy, positive regard, cognitive stimulation, shared visual attention, and bi-directional interaction (Table 1).<sup>12,28</sup>

### Sample Size Determination

Sample size was based upon 80% statistical power to detect a 1.3 points/year growth in RDLS raw score between 3 equal-sized (2 CI and 1 normal hearing) subgroups in a 3 year,

longitudinal analysis. The needed parameters for the calculation were adopted from reported RDLS scores of children undergoing CI.<sup>18, 29</sup>

## Statistical Analysis

Baseline demographic, socioeconomic and medical history factors were characterized as means and standard deviations for continuous variables and as frequency distributions for categorical variables. Baseline comparisons between the children undergoing CI and hearing controls were tested using analysis of variance (ANOVA) for continuous variables and *chi-square* tests for categorical variables. Stratified analyses based on age at implantation were conducted for the CI group to identify factors associated with age at implantation and to explore post-CI spoken language growth and language age gaps.

Developmental trajectories of verbal language in different sub-groups were explored using nonparametric regression with locally-weighted smoothing scatterplot (lowess)<sup>30</sup> to identify the mean trajectories without assuming *a priori* its parametric forms. The identified mean trajectories were then modeled using mixed-effects linear regression models, approximating any identified non-linear mean trajectories using segment-linear models for ease of interpretation. Child-specific intercepts and slopes over the period of follow-up were included as random effects, while other co-variates were modeled as fixed effects. Follow-up time, based on actual visit dates, provided the time variable.

Co-variates included child characteristics related to hearing thresholds and speech recognition performance; family characteristics related to caregiver sensitivity to communication and income; and intervention characteristics related to time spent in normal hearing, SNHL, length of traditional amplification, age at implantation, mode of communication and bilateral CI. Emergent speech recognition and bilateral CI were treated as time-dependent co-variates. Associations were adjusted for demographic characteristics and other baseline variables related to center, gender, race, ethnicity, maternal education (high school graduate or not), hearing onset, cognition, baseline RDLS scores, and time to CI activation. We performed sensitivity analyses evaluating the effect of censoring those children who underwent bilateral CI after their 2nd implant.

The *rate* of language growth, as modified by a given covariate, was modeled by including the cross-product of the follow-up time by co-variate as an interaction term. All longitudinal analyses were adjusted for centers. SAS version 9.1 (SAS Institute, Cary, NC) was used for all analyses, and all tests were two-sided ( $\alpha = 0.05$ ).

## RESULTS

### Study Population

For the experimental group, 425 children with severe-to-profound SNHL were consecutively screened; 268 were deemed appropriate candidates for CI and met inclusion criteria. Families of 188 children (70%) were willing to participate and were enrolled. Families of 80 (30%) of these children did not wish to participate. Children with SNHL enrolled in the study did not differ from non-participants in average age or socioeconomic status, although there was a difference in racial background, with African American families constituting 19% of non-participants and 9% of participants.<sup>14</sup> A total of 97 children with normal hearing who were within the age range of children undergoing CI were enrolled. The mean age at enrollment was 2.2 years for the CI group and 2.3 years for the normal hearing children. Thirty-one (17.4%) of the children undergoing CI received a second, contralateral ear implant after enrollment.

Table 2 shows baseline measures of hearing status, child and family demographics, and language status. Children undergoing CI and hearing controls differed in family income, scores of RDLs comprehension and expression, parents' perception of communication behaviors (IT-MAIS/MAIS)<sup>21,22</sup>, ratings of parent-child interactions, and cognition, as well as hearing thresholds.

Children undergoing CI were stratified into 3 groups by age of implantation: <18 months (N=72; 38%); 18–36 months (N=64; 34%), and >36 months (N=52; 28%). These 3 groups demonstrated significantly different lengths of time spent in hearing, SNHL and amplification (Table 1). There were significant differences in their baseline RDLs comprehension and expression and MAIS scores, gender, maternal education, family income, congenital onset of SNHL, and communication mode. They did not differ in mean baseline scores of parent-child interactions or cognition.

Figure 1 demonstrates the trajectories of raw score changes on the RDLs comprehension and expressive language scales over the 3 year follow-up. Children who underwent CI demonstrated slower and more variable language trajectories compared with hearing children. However, children undergoing CI produced steeper growth trajectories than those predicted by their baseline comprehension and expression scores. Significantly higher rates of comprehension and expression were noted in children undergoing CI at <18 months of age, compared with children undergoing CI between 18–36 months, and >36 months. The majority of children implanted prior to 18 months revealed trajectories of growth that paralleled those of hearing controls. CI after 18 months of age was associated with less favorable trajectories of growth in performance and greater variability in measures of both comprehension and expression.

Unadjusted, mixed-effects modeling analyses revealed that after 3 years, children who underwent CI had a mean deficit of 22.3;(95% confidence interval:19.4–25.2) points in comprehension and 19.8;(17.3–22.3) points in expression compared with hearing peers after 3 years. When stratified by age, the average deficit in comprehension scores for children undergoing CI was 8.1;(6.2–9.9) for those implanted at <18 months, 27.0;(23.8–30.1) for those implanted between 18–36 months, and 38.7;(34.2–43.2) for those implanted >36 months. The average deficit in expression scores for children undergoing CI was 8.2;(6.4–9.9) for those implanted <18 months, 21.7;(19.3–24.1) for those implanted between 18–36 months, and 29.4;(24.1–34.7) for those implanted after 36 months.

Trajectories of change in speech recognition capacity are shown in eFigure 1. Children undergoing CI showed mean rates of progress through the SRI hierarchy of speech recognition measures that were parallel to that of normal hearing children. Figure 2 demonstrates the chronologic age at which RDLs raw scores of 30.1 and 27.6 were obtained for comprehension and expression, respectively, representing the mean scores for the normal hearing children at baseline (at their enrollment age of 2.3 years). For children undergoing CI, this reference comprehension score was attained at 3.4, 4.7, and 5.3 years for children undergoing CI at <18 months, 18–36 months, and >36 months, respectively. For verbal expression, the reference score was attained at 3.4, 4.5, and 5.2 years for children undergoing CI at <18 months, 18–36 months, and >36 months, respectively.

eFigure 2 demonstrates the mean trajectories of growth in comprehension and expression scores. Faster average rates of growth in verbal comprehension and language expression trajectories were associated with earlier age of CI (age at implant <18 months).

Table 3 compares the child's chronologic age with their language age equivalent. Whereas the gap in language growth between children undergoing CI at <18 months and normal

hearing children did not widen during follow-up, larger gaps accrued in children implanted at older ages.

Independent associations of child-, family- and treatment variables with growth in comprehension and expressive skills are shown in Table 4. RDLS comprehension growth was positively associated with the amount of pre-CI residual hearing. Better baseline hearing thresholds (e.g. 85dB HL versus 105dB HL) were associated with greater growth in comprehension and expression. Although higher RDLS comprehension scores at baseline were associated with higher comprehension scores over the course of follow-up ( $P < 0.0001$ ), baseline comprehension was not significantly related to the *rate* of comprehension development.

Comprehension growth was not associated gender, congenital onset of SNHL, or baseline cognition level (Table 4). Growth in speech recognition was significantly associated with improvements in verbal language ( $P < 0.0001$ ).

Table 4 summarizes associations of language growth with characteristics of the child's family. Higher parent-child interaction scores were significantly associated with higher growth rates of comprehension and expressive language. Although lower family income was strongly associated with reduced rates of growth in comprehension and expression using bi-variable analyses (both  $P < 0.0001$ ), these negative associations were either attenuated (comprehension,  $P < .05$ ) or diminished (expression,  $P = .329$ ) after adjustment (Table 4).

Longer periods of normal hearing (prior to onset of hearing loss) were associated with higher language scores at baseline after accounting for duration of hearing loss and pre-implant amplification. A reduced rate of language development after CI was associated with longer periods of hearing loss, without or with pre-CI amplification. Prolonged periods of hearing loss without and with amplification contributed to an older age at implantation and were associated with slower growth in language comprehension. eFigure 3 highlights the slow growth in comprehension and expression associated with extended hearing aid use in children who underwent CI at a later age.

Exclusive use of spoken communication at baseline did not significantly associate with RDLS growth in children undergoing CI (Table 4). Centers were found to be significantly associated with different rates of growth in comprehension scores. Bilateral implantation was associated with a statistically insignificant tendency for positive growth in RDLS comprehension and expression scores compared to unilateral implantation after adjusting for other variables. Results from the multivariable model analyses present in Table 4 (*exc.* Bilateral Status) were unchanged for 3-year comprehension and expression.

## COMMENT

Parents commonly seek CI because they want their children with SNHL to hear and speak like children with normal hearing.<sup>14,31</sup> Language learning through listening and speaking serves as an effective marker of later school performance in children with normal hearing.<sup>32,33</sup>

CI associated with significant improvement in comprehension and expression of spoken language over the first 3 years. Growth of spoken language was positively associated with earlier, as opposed to later, ages of CI and greater residual hearing prior to CI. Unfortunately, results also revealed that gaps in spoken language growth between normal hearing children and those who underwent CI were not eliminated in the first 3 years of implant use.

Consistent with the critical period concept for language learning, early implantation in infants and toddlers associates with significantly accelerated spoken language learning. Two indices of spoken language growth revealed the association between age of CI and spoken language outcome: Performance scores in children implanted younger are closer to scores of normal hearing controls (Figures 1 & 2) and older age at CI associated with greater gaps between chronological and language ages (Table 3).

The rate of growth in performance on spoken language measures was less steep in children undergoing CI at later ages. At birth, normal hearing infants discriminate speech sounds used in all languages.<sup>34</sup> Hearing infants lose the ability to discriminate the sounds of other languages as they develop more precision with their native language, typically between 7 and 10 months of age.<sup>35, 36</sup> Accurate native language discrimination at 7.5 months of age is associated with accelerated language abilities; conversely, continued non-native language discrimination is related to reduced language abilities later.<sup>37</sup> Infants between 8 and 18 months of age use the statistical distributional properties of speech to identify the patterns contributing to the words of their language which, in turn, makes learning word meanings possible.<sup>38</sup> Models of verbal language development stress that sound pattern learning requires a neural commitment to the acoustic properties of a native language.<sup>39</sup> Children with early severe-to-profound SNHL do not experience similar neural commitments and, as such, spoken language growth is altered.

The decision to pursue CI must weigh the potential benefit of CI versus continued amplification of residual hearing. Here we observed that higher language scores at baseline were associated with greater residual hearing prior to CI. However, significantly reduced language learning was associated with the prolonged use of hearing aids prior to CI. These findings suggest that delaying implantation in order to extend hearing aid use for children with severe-to-profound hearing loss may be detrimental to language growth following CI. Spoken language learning relies on *effective* hearing, characterized as a young infant's ability to perceive the acoustic-phonetic cues of speech. Close monitoring of performance with hearing aids can determine whether speech is effectively amplified to allow spoken language acquisition to progress without imposing cumulative delays.

Although spoken language outcomes were significantly associated with the age of implantation and residual hearing, associations with environmental factors were also evident. Maternal engagement in early communication reflected in greater scores of parent-child interactions associated with growth in spoken language skills. Language comprehension and expression are influenced by parent-child interactions in bidirectional spoken communication.<sup>28, 40, 41</sup> Language exposure and caregivers' mentoring provide the context for language learning. Neuro-developmental mechanisms that support early language learning rely on interactional cues available almost exclusively in social settings.<sup>42</sup>

Family income above \$50,000 was associated with better language performance at baseline (pre-CI). This factor also favorably associated with growth rates in language comprehension. These findings are consistent with studies of the persistent problem of lower language growth experienced by children of poor families.<sup>32</sup> The notion that children reared in disadvantaged environments may have fewer early language experiences that are associated with optimal language development may extend to children undergoing CI. However, multivariable adjustment attenuated associations with income. Higher family income was associated with higher maternal education and greater maternal engagement in communication.

Some limitations of our study deserve comment. The observational design of this study and the absence of a control group of SNHL children without CI preclude causal conclusions. For ethical reasons, we could not randomize or match children with similar levels of SNHL who continue to use hearing aids. Such trials could formally test the efficacy of CI in children who are implanted at different ages or stages of linguistic development. Instead, baseline, pre-CI performance across candidates, provided an estimate of language learning trajectory of children with severe-to-profound SNHL and without CI. Although we employed rigorous adjustment procedures to mitigate the impact of potential confounds, residual confounding cannot be ruled out in this observational study.

Although bilateral CI associated with greater spoken language growth, this observation must be viewed cautiously given the brief period between the 2nd CI and the end of follow-up. Continued assessment will allow us to more carefully determine how such factors associate with longterm trends in the acquisition of spoken language skills.

The generalizability of our results beyond major implant centers is uncertain. The representativeness of the children with SNHL was likely influenced by access to participating centers and our inclusion criteria. Outcomes may have been influenced by the expertise of participating clinicians and other caregivers. Regional and other characteristics specific to participating implant centers were not directly evaluated.

## CONCLUSIONS

Results from this study carry implications for the clinical management of children with severe-to-profound SNHL. Though not determinative, age at implantation and residual hearing are variables associated with growth rates for the acquisition of spoken language in children with cochlear implants. These findings underscore the need to develop objective tools that can monitor the benefit of amplification in supporting the emergence of early skills that support spoken language acquisition and guide timely intervention with CI. Environmental factors significantly associated with performance on measures of spoken language after CI and may further account for variation in observed outcomes.

## Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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All authors are without financial interests and relationships and affiliations relevant to the subject of this manuscript. Dr. Niparko serves on advisory boards without remuneration for two cochlear implant manufacturers: Advanced Bionics Corporations and the Cochlear Corporation. The terms of this arrangement are being managed by the Johns Hopkins University in accordance with its conflict of interest policies.

External Advisors received honoraria for their review of the study protocol and progress reports. Dr. Niparko had full access to the entirety of the data in this study and takes responsibility for the integrity of the data and the accuracy of the data analysis.

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## CDaCI Investigative Team

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## Resource Centers

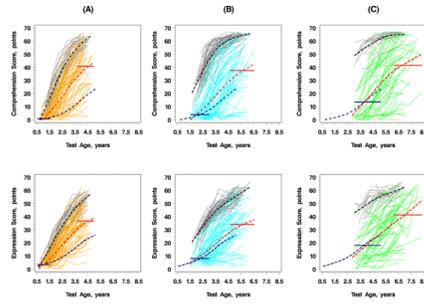
Data Coordinating Center, Johns Hopkins University, Welch Center for Prevention, Epidemiology & Clinical Research, Baltimore: Nancy E. Fink, MPH (PI); Patricia Bayton (data assembly); Daniel Habtemarian (data assembly); Neil R. Powe, MD, MPH, MBA (Senior Epidemiologist); Thelma Vilche (data assembly); Nae-Yuh Wang, PhD (Co-PI, Biostatistician, data assembly and analysis).

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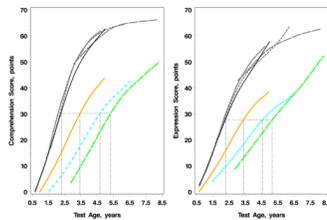
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**Figure 1.**

Developmental Trajectories of RDLs Raw Scores of Comprehension and Expression Grouped by Age At Baseline

Hearing children in gray (A) <18 months, (B) 18–36 months, and (C) >36 months; mean trajectories of sub-groups by age of enrollment in black. Children undergoing CI: (A) Implant age <18 months in orange, (B) 18–36 months in blue, (C) >36 months in green. Mean trajectories for respective age group in red dash. Mean score after 3 years designated by horizontal line, the width of which spans the age range at time of testing. Mean scores established by baseline (pre-CI) scores of children undergoing CI are shown in navy blue dash.



**Figure 2.** Nonparametric Fit of RDLs Raw Scores of Comprehension and Expression Stratified by Age at Baseline and Test Age  
Children with normal hearing in gray lines (categorized by age at baseline: solid gray line < 18 months; short dashed gray line 18–36 months; and long dashed gray line between >36 months). Children undergoing CI: <18 months at implant age in orange, 18–36 months in cyan dash, and >36 months in green dash. Mean trajectories established by baseline scores of all normal hearing children in black. For comparative assessment, the horizontal, gray line projects the chronological age at which the mean scores of normal hearing children at baseline (30.1 for comprehension and 27.6 for expression) were obtained by sub-group of children undergoing CI at different age.  
The vertical drop-lines indicate ages at which this score was obtained for each group of children. On the comprehension scale, the ages were 2.3 years for hearing children; 3.4 years for CI <18 months at implant age, 4.7 years for CI 18–36 months at implant age, 5.3 years for CI >36 months at implant age. On the expression scale, the ages were 2.3 years for hearing children; 3.4 years for CI <18 months at implant age, 4.5 years for CI 18–36 months at implant age, and 5.2 years for CI >36 months at implant age.

**Table 1**  
Measures: Domain Probed, Age Administered, Respondent, Construct(s) Measured and Interpretation

Domain/Use	Test	Age	Respondent	Construct(s) Measured	Interpretation
<b>Cognition/Screening</b>	Bayley Scales of Infant Development <sup>16</sup>	<2 years	Child	Physical and mental development	Exclusion from study if >2 SDs from age-normed scores established in typical childhood populations
	Leiter Brief Form <sup>17</sup>	>2 years	Child	Nonverbal cognitive abilities	
<b>Language/1° Outcome</b>	Reynell Developmental Language Scales <sup>18,19</sup>	1 year – 6 years 11 months	Child	Comprehension and expressive skills of spoken language	Score relative to age-normed scores established in typical childhood populations
	Infant-toddler meaningful auditory integration scale (IT-MAIS) <sup>21</sup>	1–3 years	Parent	Parent perception of everyday communication behaviors	
	Meaningful auditory integration scale (MAIS) <sup>22</sup>	>4 years	Parent	Parent perception of everyday communication behaviors	
	Early speech perception (low verbal) <sup>23</sup>	>2 years	Child	Word perception using toys	
	Early speech perception (standard) <sup>23</sup>	3–7 years	Child	Word perception requiring a picture-pointing response	
	Pediatric Speech Intelligibility (PSI-Format II) (standard) <sup>24</sup>	3–7 years	Child	Word/sentence stimuli requiring a picture-pointing response	
	Multisyllabic/lexical Neighborhood Test (MLNT) <sup>25</sup>	>3 years	Child	Word recognition ability requiring verbal response	
	Lexical Neighborhood Test (LNT) <sup>25</sup>	>4 years	Child	Word recognition ability requiring verbal response	
	Phonetically balanced word list (PBK) <sup>26</sup>	>5 years	Child	Word recognition ability requiring verbal response	
	Hearing in Noise Test (HINT-C) <sup>27</sup>	>5 years	Child	Word and sentence recognition requiring verbal response	
<b>Speech Recognition Index 1.3.20/2° Outcome</b>	Parent-child interactions videotaped and coded <sup>28</sup> :				Hierarchical battery of tests of speech recognition The SRI generates scores between 0–600 based on: IT-MAIS/MAIS: 0–100 ESP: 101–200 PSI: 201–300 MLNT/LNT: 301–400 PBK: 401–500 HINT-C: 501–600
	• Free play, Puzzles, Art gallery, Symbolic play				
<b>Video analysis of child/provider interaction/2° Outcome</b>		Toy sets selected for age of child	Parent-Child interaction	Respect for child autonomy, positive regard, cognitive stimulation, shared visual attention, bi-directional interaction	Mean scaled scores of 2 observers rating parental sensitivity, time in joint attention, and communicative competence

Domain/Use	Test	Age	Respondent	Construct(s) Measured	Interpretation
	Noun learning •				

Table 2

Baseline Characteristics of CDaCI Participants

Characteristic	Children Undergoing Cochlear Implantation Age at Implantation			Normal Hearing Children 9 months-5 years (n=97)
	<18 months (n=72)	18-36 months (n=64)	>36-60 months (n=52)	
	Means (SD)			
Age at baseline, <i>months</i> <sup>+</sup>	12.6 (3.1)	27.1 (5.5)	46.1 (7.3)	27.5 (13.1)
Age at SNHL dx, <i>months</i> <sup>+</sup>	3.8 (4.0)	12.2 (8.5)	18.0 (13.1)	n/a
Time in hearing pre-op, <i>months</i> <sup>+</sup>	0.7 (2.4)	1.6 (4.1)	7.4 (12.2)	n/a
Median {25 <sup>th</sup> , 75 <sup>th</sup> percentile}	0.0 {0.0, 0.0}	0.0 {0.0, 0.0}	0.0 {0.0, 0.0}	
Time in hearing loss, <i>months</i> <sup>+</sup>	5.0 (3.4)	13.4 (7.8)	13.3 (10.3)	n/a
Median {25 <sup>th</sup> , 75 <sup>th</sup> percentile}	3.0 {4.0, 6.0}	13.0 {6.0, 20.0}	12.0 {4.0, 21.5}	
Time in amplification, <i>months</i> <sup>+</sup>	6.8 (3.4)	11.9 (8.9)	24.9 (15.3)	n/a
Median {25 <sup>th</sup> , 75 <sup>th</sup> percentile}	7.0 {4.4, 9.3}	5.3 {2.2, 10.0}	23.3 {12.9, 38.9}	
Age at activation, <i>months</i> <sup>+</sup>	15.5 (3.2)	29.4 (5.6)	48.5 (7.4)	n/a
% time in hearing pre-op <sup>+</sup>	4.9 (15.8)	6.1 (15.6)	16.0 (26.8)	n/a
% time in hearing loss <sup>+</sup>	39.1 (21.6)	50.4 (27.9)	29.8 (24.0)	n/a
% time in amplification <sup>+</sup>	55.7 (25.3)	43.0 (29.5)	52.3 (28.9)	n/a
4 tone hearing threshold average, dB, better ear <sup>+</sup>	108.9 (15.6)	105.1 (15.6)	100.0 (17.4)	14.2 (7.1)*
RDLS Verbal Comprehension <sup>+</sup>	0.8 (1.7)	3.9 (6.4)	13.5 (13.4)	30.1 (18.3)*
RDLS Expressive Language <sup>+</sup>	3.4 (1.8)	8.0 (6.2)	17.9 (12.9)	27.6 (15.2)*
MAIS <sup>+</sup>	12.4 (15.0)	22.7 (23.0)	38.0 (31.4)	97.1 (7.7)*
Average Parent-Child Interaction score	5.4 (0.7)	5.2 (0.8)	5.1 (0.6)	5.7 (0.6)*
Cognitive Status				
Bayley PDI (< 2 years) <sup>+</sup>	94.7 (18.3) [n=61]	95.7 (19.4) [n=51]	79.5 (18.3) [n=18]	108.2 (14.7)* [n=41]
Leiter Brief IQ (> 2 years)	102.5 (21.7) [n=10]	100.3 (13.4) [n=12]	106.3 (21.0) [n=33]	109.4 (11.5) [n=53]
Combined <sup>†</sup>	97.6 (19.9)	97.7 (19.5)	93.4 (24.9)	106.9 (15.1)*
	N (%)			

Characteristic	Children Undergoing Cochlear Implantation Age at Implantation			Normal Hearing Children 9 months–5 years (n=97)
	<18 months (n=72)	18–36 months (n=64)	>36–60 months (n=52)	
Race, white	58 (81)	42 (66)	40 (77)	77 (79)
Gender, female <sup>+</sup>	32 (44)	31 (48)	35 (67)	60 (62)
Maternal Education, HS graduate <sup>+</sup>	70 (97)	60 (94)	43 (83)	91 (95)
Hispanic	9 (13)	17 (27)	11 (22)	9 (10)
Household income, \$50,000 <sup>+</sup>	43 (60)	24 (38)	21 (40)	76 (78)*
Congenital <sup>+</sup>	56 (78)	31 (51)	18 (38)	n/a
Communication Mode <sup>+</sup>				
Speech Only	19 (26)	8 (13)	10 (19)	n/a
Speech/Sign: Speech emphasis <sup>+</sup>	14 (19)	13 (20)	8 (15)	n/a
Speech/Sign: Sign emphasis <sup>+</sup>	1 (1)	4 (6)	7 (13)	n/a
Sign Only	12 (17)	19 (30)	4 (8)	n/a
Other/NR	26 (36)	20 (31)	23 (44)	n/a

<sup>†</sup> Cognitive Status measured Bayley Psychomotor Developmental Index score for children < 24 months of age and by Letter Brief IQ score for children > 24 months of age.

<sup>+</sup> Differences among children undergoing CI at <18 months; 18–36 months; and >36 months (P<0.05).

\* Differences between children undergoing CI and normal hearing children (P<0.05).

**Table 3**

Mean [95% confidence Interval] Estimates of Language Age for Children Undergoing CI and Normal Hearing Children at Baseline and 3Year Follow-up Based on Comprehension and Expression

	Children undergoing Cochlear Implantation							Normal Hearing
	CI < 12 <sup>†</sup> months	CI 12-18 months	<18 months	18-36 months	36-60 months	All	All	
<b>BASELINE</b>	<b>N=33</b>	<b>N=39</b>	<b>N=72</b>	<b>N=64</b>	<b>N=52</b>	<b>N=188</b>	<b>N=97</b>	
Chronologic Age, months	9.8 [6.8-12.8]	14.9 [12.2-17.7]	12.6 [10.5-14.6]	27.1 [24.9-29.3]	46.1 [43.7-48.5]	26.2 [24.2-28.1]	27.5 [24.7-30.3]	
Comprehension								
N	---	34	---	59	50	163	97	
Language Age, months	---	< 13.0*	---	13.7 [11.5-16.0]	18.3 [15.8-20.7]	14.8 [13.5-16.2]	28.6 [26.8-30.4]	
‡ CDaCI Comprehension Age, months	---	9.6 [6.2-12.9]	---	11.5 [9.0-13.9]	17.6 [14.9-20.3]	12.7 [11.1-14.2]	29.2 [27.2-31.2]	
Gap Chronologic Age less Language Age	---	---	---	12.8 [11.4-14.3]	27.2 [25.6-28.7]	12.9 [11.3-14.5]	-0.3 [-2.4-1.7]	
Gap Chronologic Age less CDaCI Age	---	5.6 [3.7-7.5]	---	15.1 [13.7-16.6]	27.8 [26.2-29.3]	15.1 [13.7-16.6]	-1.0 [-2.9-0.9]	
Expression								
Language Age, months	---	< 16.0*	---	16.5 [14.5-18.6]	21.2 [19.0-23.4]	17.7 [16.5-19.0]	27.6 [25.9-29.2]	
‡ CDaCI Expressive Age, months	---	8.2 [4.5-11.9]	---	11.9 [9.1-14.7]	20.4 [17.4-23.4]	13.2 [11.5-15.0]	29.8 [27.5-32.1]	
Gap Chronologic Age less Language Age	---	---	---	10.0 [8.6-11.5]	24.2 [22.6-25.8]	10.0 [8.4-11.6]	0.7 [-1.4-2.8]	
Gap Chronologic Age less CDaCI Age	---	7.0 [4.7-9.3]	---	14.7 [13.0-16.4]	25.0 [23.2-26.9]	14.6 [13.2-16.0]	-1.6 [-3.4-0.3]	
<b>36 MONTH FOLLOW-UP</b>	<b>N=32</b>	<b>N=37</b>	<b>N=69</b>	<b>N=61</b>	<b>N=50</b>	<b>N=180</b>	<b>N=84</b>	
Chronologic Age, months	49.5 [46.5-52.5]	53.5 [50.7-56.4]	51.6 [49.5-53.7]	65.7 [63.5-67.9]	85.0 [82.6-87.4]	65.8 [63.7-67.9]	64.1 [61.0-67.2]	
Comprehension								
N	31	37	68	59	41	167	84	
Language Age, months	35.5 [31.2-39.9]	35.2 [31.3-39.1]	35.4 [32.5-38.3]	33.9 [30.7-37.0]	37.2 [33.6-40.9]	35.3 [33.5-37.2]	58.0 [55.4-60.5]	
‡ CDaCI Comprehension Age, months	35.7 [31.8-39.6]	35.0 [31.4-38.6]	35.3 [32.7-38.0]	33.5 [30.7-36.4]	35.7 [32.3-39.1]	34.8 [33.1-36.5]	59.3 [56.9-61.7]	
Gap Chronologic Age less Language Age	13.6 [8.4-18.8]	17.2 [12.4-22.0]	15.5 [12.0-19.1]	32.0 [28.2-35.8]	46.3 [41.9-50.7]	29.2 [26.4-31.9]	5.9 [2.1-9.8]	
Gap Chronologic Age less CDaCI Age	13.7 [9.8-17.6]	17.6 [13.9-21.3]	15.8 [13.1-18.5]	32.2 [29.4-35.1]	47.8 [44.4-51.3]	29.5 [27.2-31.9]	4.5 [1.2-7.9]	
Expression								
Language Age, months	34.4 [30.1-38.8]	33.5 [29.5-37.5]	33.9 [31.0-36.9]	33.4 [30.2-36.6]	40.4 [36.7-44.0]	35.4 [33.5-37.3]	56.2 [53.6-58.9]	
‡ CDaCI Expressive Age, months	35.8 [31.6-40.0]	34.2 [32.0-39.8]	35.9 [33.0-38.7]	34.2 [31.2-37.3]	40.2 [36.6-43.8]	36.3 [34.5-38.2]	59.4 [56.8-62.0]	

	Children undergoing Cochlear Implantation						Normal Hearing
	CI < 12 <sup>†</sup> months	CI 12–18 months	<18 months	18–36 months	36–60 months	All	
<b>BASELINE</b>	N=33	N=39	N=72	N=64	N=52	N=188	All N=97
<b>Gap Chronologic Age less Language Age</b>	14.7 [9.7–19.7]	19.2 [14.5–23.9]	17.1 [13.7–20.5]	32.5 [28.8–36.1]	43.4 [39.2–47.6]	29.3 [26.7–31.8]	7.5 [3.9–11.1]
<b>Gap Chronologic Age less CDaCI Age</b>	13.6 [9.5–17.8]	16.9 [12.9–20.9]	15.3 [12.5–18.2]	31.5 [28.5–34.5]	43.6 [40.0–47.2]	28.1 [25.8–30.4]	4.5 [1.2–7.8]

<sup>†</sup>The RDLS is not validated for age < 12 months, hence the oral language development is numerically not ascertainable through RDLS for the group “CI < 12” at Baseline.

<sup>\*</sup>The floor of RDLS language age measure is coded as “< 13 months” for comprehension, and “< 16 months” for expression. Language age measures below the RDLS floor are numerically not ascertainable through RDLS.

<sup>‡</sup>The CDaCI Comprehension Age and CDaCI Expression Age are scored according to the estimated mean ages when normal hearing children in the CDaCI Study attained these respective RDLS comprehension and expression scores

Table 4

Multivariable Adjusted<sup>†</sup> Mixed-Effects Modeling Analyses for Children Undergoing CI: Associations with RDL S Raw Scores of Comprehension and Expression at Baseline and Rate of Gain over 3 Years

	Comprehension			Expression		
	Estimate	95% Confidence Interval	P-value	Estimate	95% Confidence Interval	P-value
<b>Factors associated with RDL S raw scores at Baseline</b>						
<b>Child Characteristics</b>						
Hearing threshold average, per 20 dB ↑	-0.31	-1.11–0.49	0.4491	-0.17	-0.75–0.41	0.5668
<b>Family Characteristics</b>						
Mean Rating of Parent-Child Interactions, per point ↑	0.51	-0.15–1.16	0.1306	0.59	0.06–1.12	0.0299
Self-reported Income < \$50k @ BL	0.28	-0.41–0.96	0.4251	-0.24	-0.72–0.24	0.3202
<b>Intervention Characteristics</b>						
Length Normal Hearing, per 6 months ↑	0.71	0.05–1.38	0.0360	0.62	0.24–0.99	0.0014
Length Hearing Loss (prior to amplification), per 6 months ↑	0.01	-0.42–0.44	0.9713	0.07	-0.25–0.39	0.6800
Length Amplification, per 6 months ↑	-0.14	-0.49–0.22	0.4432	-0.02	-0.32–0.28	0.8928
Oral Communication Mode @ BL	0.08	-0.52–0.69	0.7839	-0.14	-0.53–0.26	0.5031
<b>Factors associated with differential rate of gain in RDL S raw scores over 3 Years</b>						
<b>Child Characteristics</b>						
Hearing threshold average, per 20 dB ↑	-2.28	-4.31–0.25	0.0280	-2.24	-4.20–0.27	0.0257
Speech Recognition Index, Time Dependent <sup>*</sup> , per 100 points ↑	3.76	3.07–4.45	<.0001	1.98	1.41–2.54	<.0001
<b>Family Characteristics</b>						
Mean Rating of Parent-Child Interactions, per point ↑	3.75	1.77–5.73	0.0002	3.45	1.60–5.29	0.0003
Self-reported Income < \$50k @ BL	-2.20	-3.93–0.47	0.0126	-0.82	-2.45–0.82	0.3290
<b>Intervention Characteristics</b>						
Length Normal Hearing, per 6 months ↑	-1.45	-2.70–0.19	0.0236	-2.20	-3.60–0.81	0.0021
Length Hearing Loss (prior to amplification), per 6 months ↑	-0.74	-1.91–0.43	0.2120	-0.51	-1.63–0.60	0.3663
Length Amplification, per 6 months ↑	-1.31	-2.11–0.52	0.0013	-1.12	-1.79–0.44	0.0012
Oral Communication Mode @ BL	1.57	-0.22–3.35	0.0848	1.69	-0.02–3.41	0.0523
Bilateral CI Status, time-dependent	2.19	-0.86–2.55	0.1049	1.67	-0.94–4.27	0.2087

<sup>†</sup> Each variable is adjusted for study centers, gender, race, ethnicity, maternal education (high school graduate or not), hearing onset, cognition, baseline RDL S raw scores, time to CI activation, time dependent bilateral implant status, and all other variables listed in the Table.

\* SRI scores were included as a time-dependent variable. The average gain in SRI scores over the 3-year follow-up period was 349 points moving from an average of 45 point at Baseline to 395 points after 3 years.