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Influence Of Implantation Age On School-Age Language Performance In Pediatric Cochlear Implant Users

Emily A. Tobey^{*}, Donna Thal[†], John K. Niparko[‡], Laurie S. Eisenberg[#], Alexandra L. Quittner[¶], Nae-Yuh Wang[‡], and the CDaCI Investigative Team

^{*}Dallas Cochlear Implant Program, University of Texas at Dallas, Dallas, Texas, USA

[†]San Diego State University and Center for Research on Language, University of California, San Diego, California, USA

[‡]Johns Hopkins School of Medicine, Baltimore, Maryland, USA

[#]House Research Institute, Los Angeles, California, USA

[¶]Department of Psychology, University of Miami, Miami, Florida, USA

Abstract

Objective—This study examined specific spoken language abilities of 160 children with severe-to-profound sensorineural hearing loss followed prospectively 4, 5, or 6 years after cochlear implantation.

Study sample—Ninety-eight children received implants before 2.5 years, and 62 children received implants between 2.5 and 5 years of age.

Design—Language was assessed using four subtests of the Comprehensive Assessment of Spoken Language (CASL). Standard scores were evaluated by contrasting age of implantation and follow-up test time.

Results—Children implanted under 2.5 years of age achieved higher standard scores than children with older ages of implantation for expressive vocabulary, expressive syntax, and pragmatic judgments. However, in both groups, some children performed more than two standard deviations below the standardization group mean, while some scored at or well above the mean.

Conclusions—Younger ages of implantation are associated with higher levels of performance, while later ages of implantation are associated with higher probabilities of continued language delays, particularly within subdomains of grammar and pragmatics. Longitudinal data from this cohort study demonstrate that after 6 years of implant experience, there is large variability in language outcomes associated with modifiers of rates of language learning that differ as children with implants age.

Keywords

Cochlear implants; language; pediatrics; syntax; vocabulary; pragmatics; comprehension

Correspondence: Emily A. Tobey, Dallas Cochlear Implant Program, University of Texas at Dallas, 800 West Campbell Road, Richardson, TX 75080, USA., etobey@utdallas.edu.

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Introduction

Many families with young children who have severe-to-profound sensorineural hearing losses (SNHL) choose intervention programs that incorporate cochlear implantation and emphasize spoken (rather than signed) language for their children. As research demonstrating the safety of cochlear implants (CIs) grows (e.g. Clark, 2009), professionals are motivated to provide implants to children at younger ages (e.g. Duchesne et al, 2009; Fink et al, 2007). The rationale for earlier implantation is to provide auditory signals during the sensitive developmental periods in which spoken language skills typically develop rapidly in children with normal hearing (NH) and during which the neural connections critical to normal language development occur. The auditory information delivered by CIs, although not the same as that provided by a normal human cochlea, appears to provide children with access to much of the critical and complex information necessary for learning spoken language (Svirsky et al, 2004; Tomblin et al, 2008; Wie et al, 2007; Yoshinaga-Itano et al, 2010). In this paper, we report on data further evaluating whether children with CIs routinely develop language skills within a range that allows success at school and explore what factors contribute to the levels of language performance.

Numerous studies demonstrate that, on average, language development in children with CIs remains behind that observed for same-aged children with NH (e.g. Archbold et al, 2008; Ching et al, 2009; Duchesne et al, 2009; Geers et al, 2009; Tomblin et al, 2008). There is, however, great variability in individual performance: some children with CIs acquire language skills within normal limits while other children remain far below average. Previous studies have identified a number of additional factors that appear to be directly related to spoken language development in children using CIs (e.g. Fink et al, 2007; Geers et al, 2008; Geers & Hayes, 2011; Niparko et al, 2010). These factors include the age at which the hearing loss is identified, the age at which cochlear implantation occurs, the amount of normal hearing experience a child has prior to losing their hearing, and the nature of interpersonal communication to which the child is exposed (Moog & Geers, 2010; Niparko et al, 2010). It is clear children with CIs generally develop better spoken-language abilities than children with profound SNHL who use hearing aids (Geers et al, 2008, 2009; Geers & Hayes, 2011; Hayes et al, 2009; Moog & Geers, 2010). Results consistently indicate better spoken language skills, on average, for children with CIs who were implanted at earlier ages (generally under 18 months of age) and who, therefore, experienced longer periods using auditory stimuli for communicative purposes and shorter periods of auditory deprivation (Nicholas & Geers, 2007; Tomblin et al, 2005). In addition, factors such as low caregiver sensitivity and long periods of hearing-aid experience in children with less profound hearing losses, in combination with later implantation ages, are strongly related to lower language abilities (Niparko et al, 2010).

Because of their earlier exposure to sound, it is logical to hypothesize children using CIs who are implanted at earlier ages will achieve higher levels of spoken language than later implanted children and will be closer to their peers with NH. Several previous studies indicate that this may be the case (Archbold et al, 2008; Desjardin et al, 2009; Hay-McCutcheon et al, 2008; Spencer & Oleson, 2008; Wang et al, 2008; Wie, 2010; Yoshinaga-Itano et al, 2010). More recently, in a study of spoken language outcomes of 188 children using CIs three years after implantation, children implanted prior to 18 months of age showed steeper rates of growth in language on the Reynell Developmental Language Scales (RDLS) (Reynell & Gruber, 1990) than children implanted between 18 months and 5 years of age (although on average, they did not reach age-appropriate levels) (Niparko et al, 2010). In this report, we extend these findings by describing the language skills of a subset of children from the same research project who completed language assessments at 4, 5, or 6 years after receiving their CIs. We assessed spoken language development using the core

composite standard score from the Comprehensive Assessment of Spoken Language (CASL) (Carrow-Woolfolk, 1999), a score that resembles the receptive and expressive language scores reported in previous studies using more global language comprehension and expressive measures, similar to the RDLs. We also expand on those studies by examining performance in three specific language domains (vocabulary, grammar (receptive and expressive syntax), and pragmatic judgments) using standardized subtests from the CASL. Use of these particular subtests allowed us to examine how children using CIs performed relative to children with NH on measures that are important to success at school ages. The CASL is normed on 1700 children and is representative of the United States population with respect to geographic area, gender, race, and socioeconomic status (SES). The CASL is intended to be used with a range of ages (3 to 21 years) and evaluates the Carrow-Woolfolk integrative language theory in which language knowledge and performance are measured in four linguistic categories (semantics, syntax, pragmatics, and supralinguistics) (Hoffman et al, 2011).

Methods

The data for this report were collected as part of the Childhood Development after Cochlear Implantation (CDaCI) study. Specific details on the prospective, longitudinal design and language outcomes three years after implantation from the CDaCI study have been previously reported (Eisenberg et al, 2006; Fink et al, 2007; Niparko et al, 2010; Wang et al, 2008). Institutional Review Board approval was obtained at each of the test sites: the Johns Hopkins School of Medicine (Baltimore, USA), the House Research Institute (Los Angeles, USA), the University of Miami (Miami, USA), the University of Michigan (Ann Arbor, USA), the University of North Carolina (Chapel Hill, USA), and the Callier Advanced Hearing Research Center of the University of Texas at Dallas (Dallas, USA).

Participants

The participants in this report included 160 children who were implanted between the ages of 6 months and 4 years 11 months. At the time of enrollment, children were required to have a severe- to-profound SNHL and live with English-speaking families that were committed to enrolling their children in English-speaking schools once they reached school age. The exclusion criteria were: (1) chronological age of more than 5 years at baseline, (2) difficulty with post-surgical complications, and/or (3) English not the primary language spoken at home. Spoken language outcomes were assessed prior to implantation (at baseline), at 6 and 12 months post implantation, and annually thereafter. The RLDS were used for spoken language assessment from baseline to January 2007 when the CASL was introduced into the protocol. The children were required to pass a developmental screening with a score of at least 70 on the motor scales of the Bayley Scales of Infant Development (BSID II) (Bayley, 1993) if the children were enrolled in the study under the age of 2 years, or a score of at least 66 on the Leiter International Performance Scale - Revised (Leiter-R) (Roid & Miller, 2002) if the children were enrolled in the study between 2 years and 4 years 11 months prior to implantation. These two measures were selected to provide a general developmental screening that did not require spoken language responses, per se.

As shown in Table 1, the sample was 76% Caucasian, 18% Hispanic, and 53% female. Ninety-three percent of the families had a mother with at least a high school diploma, and 39% of the families reported annual incomes under \$50,000. The U.S. Census Bureau reported in 2010 that the median household income in the U.S. was \$49,445, and approximately 31.6% of the respondents to the census reported a two-month or greater period of poverty between 2004 and 2007 (DeNavas-Walt et al, 2011). Thirty-six percent of the children with CIs used Advanced Bionics Corporation devices, 15% used Med-El

Corporation devices, and 49% received Cochlear Corporation devices. Twenty-one percent of the sample received a second, sequential implant.

Auditory history is indicated for each group as a function of age of diagnosis and amount of residual hearing in the better ear prior to implantation (indicated as a 4-frequency threshold average). On average, the population was identified at 2.5 months, entered the study at 26.4 months, and was activated at 29 months. Average pre-implant unaided hearing thresholds were 104.7 dB HL. Three years following the first implantation, the speech recognition index in quiet (SRI-Q) averaged 21.2 points. Fifty-seven percent of the population reported congenital etiologies. In addition, 37% of parents reported they used a speech emphasis when communicating with their child. Communication performance before implantation was assessed with two measures: a standardized language test (the RDLS) and a measure of parent-child interactions when conversing with their child. Maternal interactions were coded using the National Institute of Child Health and Development (NICHD) Early Child-care Study codes, including the maternal sensitivity composite which consisted of four subscales: (1) Sensitivity/responsivity, (2) Respect for child's autonomy, (3) Positive regard, and (4) Hostility. Trained observers coded the videotaped interactions using a seven-point rating scale (1 = very low, 7 = very high). The sensitivity/responsivity scale reflected the degree to which the mother expressed positive regard and emotional support to the child. Respect for child's autonomy assessed whether the mother recognized and respected the child's individuality, motives, and perspectives during the session. Positive regard rated the amount of positive feelings directed toward his/her child (e.g. parent watches attentively, praises child). Hostility was reversed-coded and reflected the parent's expression of anger toward or rejection of the child.

The participants were divided into two age groups, < 2.5 years and 2.5 to 5 years of age, as described below under the *Statistical treatment* section. As shown in Table 1, 98 children were implanted under the age of 2.5 years, and 62 children were implanted between 2.5 and 5 years of age. Average age at baseline was 16.5 months for the younger group and 42.2 months for the older group. Hearing thresholds were similar (107.6 and 100.2 dB HL for the younger and older group, respectively). Higher average raw scores on the RDLS were observed for the older group (11.5 and 15.9 for baseline language comprehension and expression) than the younger group (1.6 and 4.8, respectively). Developmental screening scores were similar for the two age groups on the BSID-II and Leiter-R measures at baseline. Bilateral implantation occurred in 24% and 16% of the younger and older implanted groups, respectively. Sixty-nine percent of the younger group and 37% of the older group experienced congenital hearing losses. Additional breakdowns between the two groups for race, gender, maternal education level, ethnicity, income, and communication mode are similar and shown in Table 1.

Language measurement

At each test session, four subtests of the CASL were administered orally. Each of the subtests was considered a "core" test for the ages of the children in the study as shown in Table 2. The subtests (antonyms (vocabulary), syntax construction, paragraph comprehension of syntax, and pragmatic judgment) examine language skills appropriate for assessing these particular ages most reliably. The antonyms subtest assessed single-word retrieval and production. Thus, correct responses depended on both receptive and expressive vocabulary. The test requires a child to identify words that are opposite in meaning when asked in an open-ended format, "Tell me a word that is opposite of ____." The syntax construction test evaluated rules used to formulate and express spoken sentences (expressive syntax), while the paragraph comprehension of syntax test assessed understanding of those structures (receptive syntax). The pragmatic judgment test assessed knowledge of social interactional rules for using language in situations typical for children with NH in this age

range (pragmatics). The tests were administered at the CDaCI clinical centers by trained, certified professionals.

Dependent variables included age-based standard scores, with a mean of 100 points and standard deviation (SD) of 15 points. A core composite score, similar to the standard scores obtained from tests like the RDLS or the Preschool Language Scales (PLS) (Zimmerman et al, 1991) also was calculated. The composite and individual sub-test scores allowed us to evaluate how much additional information was provided using the domain-specific subtests. The tests were administered to the children 4, 5, and 6 years after they began using their CIs, when they were 6.5 (range 4.8–9.3), 7.4 (range 5.6–10.3), and 8.4 (range 6.7–11.5) years of age, on average.

Statistical treatment

The distributions of standard scores were first examined for the core composite and then for each domain-specific subtest at the three follow-up test times for the entire group of children. In addition, we also stratified the data by quintiles of age at implantation. Results suggested a nonlinear trend of means for the CASL standard scores over age at implantation and showed wide but relatively homoscedastic variability across the implantation age range. Nonparametric regressions employing locally weighted, smoothed scatter (lowess) plots were used to further explore CASL standard scores as a function of age at implantation, stratified by year of follow-up.

Children were stratified into two age groups based on the results of an exploratory analysis examining their performance on the core composite of the CASL as a function of age of implantation: children implanted under the age of 2.5 years and children implanted between 2.5 and 5 years of age. As shown in the upper panel of Figure 1, standard scores decrease as a function of age of implantation until around 2.5 years of age and then appear to plateau as a function of age of implantation between 2.5 and 5 years of age. Figure 1 illustrates the 95% confidence band for the implant-age related outcome trajectories of the core composite standard scores at the 6-year follow-up. The 6-year confidence bands enveloped both the 4- and 5-year trajectories, indicating no statistically significant differences between the trajectories by age of implantation among the three annual follow-up visits studied. These exploratory analyses guided our main modeling approach, a mixed-effects model, which treated ages at implantation as continuous variables and used unstructured correlations to account for within-child correlations among the 4-, 5- and 6-year values obtained for each child. A linear spline term of 2.5 years of age was introduced in the model to allow for the different regression slopes associated with ages of implantation occurring before and after that point in age as suggested by the exploratory analyses. The multivariable model adjusted for race, gender, ethnicity, and baseline characteristics of study site, family income, maternal education, congenital onset, residual hearing (4-frequency pure-tone average), communication mode, RDLS verbal comprehension score, speech recognition index SRI-Q score, and cognitive screening score. Given the existence of certain sparse cells, Fisher's exact test was used to compare differences in frequency distributions of standard scores between the two groups for each follow-up visit as a conservative way to evaluate associations between the categorized language outcomes and age at implantation. All tests were two-sided and a p-value of < 0.05 was considered statistically significant. Analyses were carried out using SAS version 9.2.

Results

Core composite scores

The top panel of Figure 1 displays the trajectories of the core composite standard scores based on age at implantation. Trajectories indicating the mean standard score performance at 4-, 5- and 6-years follow-up are indicated by solid lines, and the dashed lines represent the 95% confidence limits bounding performances acquired at 6-years post implantation. The bottom panel of Figure 1 illustrates the data divided into quintiles as a function of age of implantation. Means and SDs are shown for each of the three follow-up test sessions as a function of age of implantation quintile. Across the range of ages at implantation and across all three follow-up visits, the variability of the core composite scores was large, with SD estimates consistently greater than 15, the SD reported for the CASL. At the 4-year follow-up, the mean chronological age of the participants was 6.5 years, and the overall mean core composite standard score was 76.4 (SD 22.6), ranging from 89.3 (SD 25.9) for children in the youngest age of implantation quintile to 69.6 (SD 22.5) for children in the oldest age of implantation quintile. At the 6-year follow-up, the mean chronological age was 8.4 years, and the overall mean core composite standard score was 77.6 (SD 24.3), ranging from 94.7 (SD 19.9) for children in the youngest age of implantation quintile to 71.3 (SD 28.4) for children in the oldest age of implantation quintile.

The trajectory of mean core composite standard scores over the range of ages at implantation was not linear and did not show statistically significant differences in trajectory patterns at 4-, 5-, and 6-year follow-ups (see top panel of Figure 1). For children implanted under 2.5 years of age, each additional year later that implantation occurred was associated with an average of 12.8 (95% confidence interval: 6.8 to 18.8, $p < 0.0001$) points lower in core composite standard scores over the 4- to 6-year period. Implantation age was not associated with significant change in core composite standard scores (Mean = -1.3 , 95% confidence interval: -4.3 to 7.1 , $p = 0.65$) during the same period for children implanted between 2.5 and 5 years of age (as shown in unadjusted model detailed in Table 3).

In the multivariable-adjusted analysis, the core composite trajectory for children implanted before 2.5 years of age was attenuated but remained statistically significant. Each additional year later in implantation age was associated with an average of 7.3 (95% confidence interval: 1.8 to 12.7, $p = 0.0088$) points lower in core composite standard score over the 4- to 6-year follow-up (as depicted in the adjusted model detailed in Table 3). A significant change in trajectory pattern was observed for children who were implanted between 2.5 to 5 years of age after multivariable adjustment, with each additional year in implantation age associated with an average decline of 10.6 (95% confidence interval: 5.5 to 15.8, $p < 0.0001$) points in core composite standard scores after 4- to 6-year implant use, similar to the trajectories observed for the younger implant recipients.

Modeling outcome trajectories as a continuous linear function of age at implantation indicated a consistent decline of mean core composite standard scores as implantation age increased across 4-, 5-, and 6-year follow-up, as shown in Table 4. At the 4-year test session, each additional year later in implantation resulted in a decrease of 9.5 in core composite standard scores (95% confidence intervals: -12.6 to -6.5 , $t = -6.16$, $p < .0001$). The adjusted analyses also indicated baseline measures of developmental screening scores, parent-child interaction scores, SRI-Q scores, and RDLS comprehension scores were associated significantly with the core composite scores. The core composite standard score was higher by 0.9 points for every point higher achieved on the RDLS comprehension score at baseline ($t = 5.15$, $p < .0001$), and was higher by 7.1 points with every point higher on the mean parent-child interaction score at baseline ($t = 3.93$, $p = .0002$). Significantly higher core composite standard scores were independently associated with higher developmental

screening scores ($t = 3.23$, $p = .0016$) and SRI-Q scores ($t = 2.22$, $p = .0287$) at baseline after adjustment. The estimated linear decline of core composite standard scores over age at implantation improved significantly at the 5-year (average points 1.1, $t = 2.00$, $p = 0.0475$) and 6-year (average points 2.0, $t = 2.71$, $p = 0.0079$) follow-up over the slope at 4-year.

Domain-specific language scores

An identical pattern was found for paragraph comprehension (reception), syntactic construction (expression), and pragmatic judgments (pragmatics) shown in Figures 2, 3, and 4, respectively. At the 4-year follow-up, each additional year later that implantation occurred resulted in a decrease of 9.5 in receptive syntax standard scores ($t = -5.16$, $p < .0001$), a decrease of 11.9 in expressive syntax standard scores ($t = -6.72$, $p < .0001$), and a decrease of 10.1 in pragmatics standard scores ($t = -6.50$, $p < .0001$). Developmental screening scores, SRI-Q, parent-child interaction scores, and RDLS vocabulary comprehension scores at baseline were also significantly associated with receptive syntax, expressive syntax, and pragmatic judgment language domains. The receptive syntax standard scores were 1.0 points higher for each point higher in vocabulary comprehension scores at baseline on the RDLS ($t = 4.50$, $p < .0001$), 6.9 points higher for each point higher in mean parent-child interaction scores at baseline ($t = 3.67$, $p = .0004$), 6.1 points higher per 100 points higher in the SRI-Q scores at baseline ($t = 2.56$, $p = .0118$), and 4.1 points higher per 20 points higher in the developmental screening scores at baseline ($t = 2.55$, $p = .0121$). The expressive syntax standard scores were 1.0 points higher for each point increase in vocabulary comprehension score at baseline on the RDLS ($t = 5.14$, $p < .0001$), 6.2 points higher for 1 point higher mean parent-child interaction scores at baseline ($t = 3.27$, $p = .0014$), 6.2 points higher per 100 points higher in the SRI-Q scores at baseline ($t = 2.54$, $p = .0125$), and 3.7 points higher per 20 points higher in the cognitive screening score at baseline ($t = 2.89$, $p = .0047$).

The pragmatics standard scores were 0.7 points higher for each point increase in vocabulary comprehension score at baseline on the RDLS ($t = 3.94$, $p = .0001$), 6.8 points higher for 1 point higher mean parent-child interaction scores at baseline ($t = 4.38$, $p < .0001$), 5.4 points higher per 100 points higher in the SRI-Q scores at baseline ($t = 2.59$, $p = .0111$), and 3.9 points higher per 20 points higher in the developmental screening scores at baseline ($t = 3.33$, $p = .0012$). The estimated slopes of the linear function based on age at implantation significantly improved for expressive syntax (2.8-point gain, $t = 3.44$, $p = .0008$) and pragmatics (1.8-point gain, $t = 2.40$, $p = .0180$) at the 5-year, and for receptive syntax (2.5-point gain, $t = 2.03$, $p = .0447$), expressive syntax (4.4-point gain, $t = 3.56$, $p = .0006$) and pragmatics (2.9-point gain, $t = 3.21$, $p = .0018$) at the 6-year follow-up.

The pattern was similar for vocabulary (antonyms, Figure 5) with one exception; the SRI-Q scores at baseline were not significantly associated with this language sub-domain. At the 4-year follow-up, each additional year later in implantation resulted in a decrease of 5.7 in standard scores ($t = -3.31$, $p = .0013$). Parent-child interaction scores, developmental screening scores, and paragraph comprehension on the RDLS at baseline vocabulary scores significantly. The vocabulary standard scores were 0.9 points for higher vocabulary comprehension score at baseline on the RDLS ($t = 5.60$, $p < .0001$), 5.6 for every point higher in mean maternal sensitivity scores at baseline ($t = 3.20$, $p = .0018$), and 3.6 points higher per 20 points higher in the developmental screening score at baseline ($t = 2.87$, $p = .0049$). The slope of the function based on age at implantation did not change significantly at the 5- and 6-year follow-ups for vocabulary standard scores.

Discussion

Our data support and extend observations indicating better spoken language outcomes occur in children who receive CIs at younger ages (Nicholas & Geers, 2007; Niparko et al, 2010;

Yoshinaga-Itano et al, 2010). Mean scores for children implanted after 2.5 years were significantly lower than children implanted at younger ages (Fagan & Pisoni, 2010; Holt & Svirsky, 2008; Nicholas & Geers, 2007; Niparko et al, 2010). A critical corollary of the present observations is that baseline gaps in language performance with later age at implantation are not effectively mitigated, and language delays persist. The spoken language advantage for the children implanted prior to 2.5 years was seen in each of the specific language domains. Although early implantation, on average, appears to provide an advantage for spoken language development, it did not assure the development of spoken language in the normal range for all children by school age, nor did implantation up to 5 years of age obviate the opportunity to develop spoken language skills within the normal range 4 to 6 years after implantation. In part, these persistent delays may reflect the poor psychometric properties of the developmental screening tools (BSID-II and Leiter-R). Further exploration will need to be conducted to determine how early developmental screenings influence group characteristics.

The integration of auditory and visual cues carries important implications for developmental learning in sensitive periods. Although auditory sensation is restored after cochlear implantation, these co-activated processes in deaf children demonstrate a bias toward visual rather than auditory stimuli (e.g. Bergeson et al, 2010). The persistence of a visual bias suggests multisensory integration may not develop normally when a single sense dominates during early development. Sustained auditory stimulation during early sensitive periods may, therefore, be necessary to ensure adequate influence on multisensory integration circuits. Cochlear implantation within the first year of life may redeem the functional capacity of these circuits and enable matching of auditory and visual cues (Bergeson et al, 2010). Detection and learning of grammar requires these multisensory interactions due to the low perceptual salience of grammatical and pragmatic cues. Unlike vocabulary and semantic judgments, grammar substantially improved for children who received implants before 18 months, perhaps because early activation of auditory cortex served to rescue multisensory development that ultimately amplified the salience of grammatical cues.

At least two lines of research are relevant to this consistent implant-age finding, both related to developmental neurobiology. The first demonstrates the effects of lack of auditory input on the cortical neuronal connections essential to development of normal auditory processing abilities (Giraud & Lee, 2007; Kral & Eggermont, 2007; Sharma et al, 2007, 2009). One effect of auditory deprivation is to reconfigure cortical circuits. The auditory system, similar to the visual system, requires stimulation during early periods of maximal receptiveness (Sharma et al, 2007, 2009). These sensitive periods in the development of auditory cortex terminate with reduction in synaptic activity and are associated with an inability to completely restore hearing function (Kral & Eggermont, 2007). Because neuronal structures typically used for auditory processing may become dedicated to other sensory domains through experience, competition occurs from the other sensory inputs (e.g. the visual domain) and seriously compromises the ability to learn spoken language (Kral & Eggermont, 2007). In essence, children with CIs learn with brains that are no longer optimally suited to processing the sensory information essential to auditory/oral language learning. Further clinical data support this notion.

Electroencephalography studies of auditory evoked potentials demonstrated normal latencies of cortical responses in children using CIs only if they received an implant before 3.5 years of age, suggesting age of implantation has effects on cortical processing (Sharma et al, 2009). In this study, lower vocabulary scores were associated with lower speech recognition scores at baseline.

The second line of research arises from the field of developmental psycholinguistics. Language learning is experience dependent, with early perceptual experiences shaping the language learning mechanism. Early proliferation of synaptic connections in the neonate creates a brain that is optimally primed for learning. Postnatal changes in these connections that come through learning make learning more or less efficient, depending on the nature of the input (Bates et al, 2003). In the case of a child with NH, language learning is driven by sound and social interactions (Pressman et al, 1999). In the 8- to 10-month period after birth, language comprehension, communicative interactions, imitation, and reasoning are well documented and correlated with changes in frontal lobe activity (Bates et al, 2003). This is also the period in which children with normal hearing begin to become aware of the statistical regularities in their language that allow them to identify the patterns that form words and to ignore the phonemic contrasts of sounds that are not part of their native language. Changes in synaptic density also coincide with bursts in vocabulary and grammar between 16 and 36 months of age. This growth in spoken language abilities appears to be directly related to the quantity of speech heard by children. The number of words heard from birth to one year has a major effect on language skills at three years of age and on language and academic performance at school age (Pressman et al, 1999; Bates et al, 2003).

Early language experience not only has an effect on spoken language comprehension and production, but also on processing abilities that enhance language learning. Children who experience richer input from their caregivers develop more efficient processing skills in addition to developing vocabulary more quickly, so that they become even better able to learn more language more quickly (Fernald et al, 2010; Fernald, 2010; Fernald & Marchman, 2012). This observation is supported in our data by the higher scores associated with average parent-child interaction scores resulting in a range of 5.6 (vocabulary) to 7.1 (core composite) scores in the children using CIs. Individual differences in the speed and accuracy of recognizing spoken words at 18- and 25-months of age predict differences in cognitive and language outcomes at 8 years of age (Fernald & Marchman, 2011). Higher speech recognition scores result in higher standard scores for expressive and receptive syntax, as well as pragmatic judgments. These critical early language experiences are denied a young deaf child and place such children at serious risk for continued language impairment even after cochlear implantation. The absence of these early experiences is a potential explanation for the continued lag of children using CIs, even when they are implanted prior to 30 months of age. Based on early clinical observations, there are those who argue strongly for implantation during the first year (Sharma et al, 2007) and evidence of striking success is apparent in at least one study in which children were implanted as early as 5 months of age (Wie, 2010). This is an area in which more carefully controlled studies are needed.

Our findings of positive relationships between spoken language outcomes and developmental functioning, language comprehension, and speech reception prior to implantation are consistent with prior studies (Fagan & Pisoni, 2010; Geers et al, 2011; Niparko et al, 2010; Yoshinaga-Itano et al, 2010). After multivariable adjustment, we failed to find a relation between family income or residual hearing and spoken language outcomes 4, 5, and 6 years after implantation, unlike the 3-year follow-up when there was a positive relationship (Niparko et al, 2010). These differences may be the result of experiences, such as schooling, and/or as yet undiscovered intervening variables that mitigated the SES and early hearing effects observed at the 3-year follow-up.

Given the important linkages between experience and language learning, these data suggest that as children with implants mature, their changing environment should be considered. That is, the language demands of their everyday setting likely manifest effects, and children of elementary school age increasingly attain language exposure and language models outside

of their homes. As such, the immediacy of parent-child interaction is likely attenuated. As the language learning environment evolves, subdomains are likely differentially affected by the environment in which an implant is used, with emergent “peer effects” (Mashburn et al, 2009). Pragmatics are likely to be improved in settings where typical, hearing language models are present. Such model access, along with the active teacher control (e.g. avoidance of stringent didactic teaching models as in strictly rote self-contained classroom) and optimized behavioral management, better-managed classrooms can support quality peer-to-peer interactions to associate with an outcome of a higher rate of spoken language acquisition in typically developing children.

The significant amount of variation within the children using CIs remains another significant challenge, particularly for clinicians who are in the position of helping parents decide whether to choose a CI or an alternative hearing aid for their child. Studies focusing on individual differences are needed to understand how environmental factors influence CI use and resultant developmental outcomes. Our data suggest an even stronger statement: a study of individual differences within both of the age groups is of critical importance to understand why some younger-implanted children fail to acquire age-appropriate language skills and why some older-implanted children adopt trajectories that diminish gaps. Studies that aim to detect individual differences in perception, cognition, learning styles, and experience may supplement data from clinical trials to better elucidate significant associations with outcome; thus, contributing to better clinical guidance.

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Acronyms

CI(s)	Cochlear implant(s)
NH	Normal hearing
RDLS	Reynell Developmental Language Scales
CASL	Comprehensive Assessment of Spoken Language
CDaCI	Childhood Development after Cochlear Implantation
BSID II	Bayley Scales of Infant Development
Leiter-R	Leiter International Performance Scale-Revised
SD	Standard Deviation
SRI-Q	Speech Recognition Index in Quiet
WISC-IV	Wechsler Intelligence Scale for Children-IV
SNHL	Sensorineural Hearing Losses
NICHD	National Institute of Child Health and Development

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CDaCI Investigative Team Members

Data collection sites

1. House Research Institute, Los Angeles: Laurie S. Eisenberg, PhD, CCC-A (PI); Karen Johnson, PhD, CCC-A (coordinator); William Luxford, MD (surgeon); Leslie Visser-Dumont, MA, CCC-A (data collection); Amy Martinez, MA, CCC-A (data collection); Dianne Hammes Ganguly, MA (data collection); Jennifer Still, MHS (data collection); and Carren J. Stika, PhD (data collection).
2. Johns Hopkins University, Listening Center, Baltimore: John K. Niparko, MD (PI); Steve Bowditch, MS, CCC-A (data collection); Jill Chinnici, MA, CCC-A (data collection); James Clark, MD (data assembly); Howard Francis, MD (surgeon); Rick Ostrander, EdD (data collection); Dawn Marsiglia, MA, CCC-A/SLP (data collection); and Jennifer Yeagle, MEd, CCC-A (data collection).
3. Johns Hopkins University, The River School, Washington, DC: Nancy Mellon, MS (administration); Meredith Ouellette, MS (coordinator); Meredith Dougherty, MS (data collection); Patricia Gates-Ulanet, PhD (data collection); and Julie Verhoff, AuD, CCC-A (data collection).
4. University of Miami, Miami: Annette Hodges, PhD, CCC-A (PI); Thomas Balkany, MD (surgeon); Alina Lopez, MA, CCC-SLP/A (coordinator); Leslie Goodwin, MSN, CCRC (data collection).
5. University of Michigan, Ann Arbor: Teresa Zwolan, PhD, CCC-A (PI); Caroline Arnedt, MA, CCC-A (clinic coordinator); Hussam El-Kashlam, MD (surgeon); Kelly Starr, MA, CCC-SLP (data collection); and Ellen Thomas, MA, CCC-SLP, Cert AVT (data collection).
6. University of North Carolina, Carolina Children's Communicative Disorders Program, Chapel Hill: Holly F.B. Teagle, AuD, CCC-A (PI); Craig A. Buchman, MD (surgeon); Carlton Zdanski, MD (surgeon); Hannah Eskridge, MSP (data collection); Harold C. Pillsbury, MD (surgeon); and Jennifer Woodard (coordinator).
7. University of Texas at Dallas, Dallas Cochlear Implant Program, Callier Advanced Hearing Research Center, Dallas: Emily A. Tobey, PhD, CCC-SLP (PI); Lana Britt, AuD, CCC-A (Co-coordinator); Janet Lane, MS, CCC-SLP (data collection); Peter Roland, MD (surgeon); Sujin Shin (data collection); Madhu Sundarajan, MS, CCC-SLP (data collection); and Andrea Warner-

Czyz PhD CCC-A (cocoordinator)

Resource centers

1. Data Coordinating Center, Johns Hopkins University, Welch Center for Prevention, Epidemiology & Clinical Research, Baltimore: Nae-Yuh Wang, PhD (PI, biostatistician); Christine Carson (study manager, data analysis); Thelma Grace (data assembly); Nancy E. Fink, MPH (former PI); and Patricia Bayton (data assembly).
2. Psychometrics Center, University of Miami, Department of Psychology, Coral Gables: Alexandra Quittner, PhD (PI); Ivette Cruz, PhD (data analysis); Cara Kimberg (data assembly); Sandy Romero (data assembly); and Mary Beth Grimley (data assembly).

Study oversight committees

1. Executive committee: John K. Niparko, MD (chair); Laurie S. Eisenberg, PhD; Nancy E. Fink, MPH (former member); Alexandra L. Quittner, PhD; Donna Thal, PhD; Emily A. Tobey, PhD; Nae-Yuh Wang, PhD.
2. External advisors: Noel Cohen, MD; Julia Evans, PhD; Ann Geers, PhD; Karen Iler Kirk, PhD.

Figure 1.

In the top panel, the mean trajectories for CASL core composite scores as a function of age of implantation are shown for the 4-, 5- or 6-year follow-up visits after implantation. The dashed lines represent the 95% confidence limits for the trajectories acquired at the 6-year follow-up. The bottom panel depicts the means and standard deviations for the five quintiles for each test period.

Figure 2.

In the top panel, the mean trajectories for CASL receptive syntax standard scores as a function of age of implantation are shown for the 4-, 5- and 6-year follow-up visits after implantation. The dashed lines represent the 95% confidence limits for the trajectories acquired at the 6-year follow-up. The bottom panel depicts the mean and standard deviations for the five quintiles for each test period.

Figure 3.

In the top panel, the mean trajectories for CASL expressive syntax standard scores as a function of age of implantation are shown for the 4-, 5- and 6-year follow-up visits after implantation. The dashed lines represent the 95% confidence limits for the trajectories acquired at the 6-year follow-up. The bottom panel depicts the mean and standard deviations for the five quintiles for each test period.

Figure 4.

In the top panel, the mean trajectories for CASL pragmatics standard scores as a function of age of implantation are shown for the 4-, 5- and 6-year follow-up visits after implantation. The dashed lines represent the 95% confidence limits for the trajectories acquired at the 6-year follow-up. The bottom panel depicts the mean and standard deviations for the five quintiles for each test period.

Figure 5.

In the top panel, the mean trajectories for CASL antonym scores as a function of age of implantation are shown for the 4-, 5- and 6-year follow-up visits after implantation. The dashed lines represent the 95% confidence limits for the trajectories acquired at the 6-year follow-up. The bottom panel depicts the mean and standard deviations for the five quintiles for each test period.

Table 1

Characteristics of 160 CDaCI participants with spoken language assessments at 4, 5, or 6 years after their first cochlear implantation using Comprehensive Assessment of Spoken Language (CASL).

Characteristic	Implanted < 2.5 years (n = 98)	Implanted 2.5–4.9 years (n = 62)	Overall 9 months – 5 years (n = 160)
<i>Hearing history</i>		<i>Means (SD)</i>	
Age at baseline, <i>months</i>	16.5 (6.2)	42.2 (9.2)	26.4 (14.6)
Age at SNHL diagnosis, <i>months</i> ¹	1.1 (3.1)	4.9 (9.9)	2.5 (6.7)
Age at activation, <i>months</i>	19.2 (6.2)	44.6 (9.4)	29.0 (14.6)
4-frequency hearing threshold average, dB, better ear ^{1 1}	107.6 (15.9)	100.2 (16.3)	104.7 (16.4)
SRI-Q at 3 years post 1st implant ^{1 1 1}	13.1 (15.1)	19.4 (24.0)	21.1 (24.6)
<i>Early communication</i> [#]			
RDLS verbal comprehension [#]	1.6 (2.8)	11.5 (12.9)	5.9 (10.0)
RDLS language expression [#]	4.8 (3.7)	15.9 (12.0)	9.6 (10.0)
Average parent-child interaction score	5.3 (0.7)	5.2 (0.7)	5.3 (0.7)
<i>Developmental screening score</i>			
Bayley Scales of Infant Development (< 2 years)	96.9 (18.0) [<i>n</i> = 81]	87.0 (18.5) [<i>n</i> = 29]	94.3 (18.5) [<i>n</i> = 110]
Leiter International Performance Scale - Revised (> 2 years)	101.9 (20.3) [<i>n</i> = 15]	106.7 (20.0) [<i>n</i> = 32]	105.1 (20.0) [<i>n</i> = 47]
		<i>Demographics N (%)</i>	
Race, Caucasian	78 (80)	43 (69)	121 (76)
Gender, female	46 (47)	39 (63)	85 (53)
Maternal education, High school graduate	97 (99)	52 (84)	149 (93)
Hispanic	15 (15)	13 (21)	28 (18)
Household income, < \$50,000	34 (35)	29 (47)	63 (39)
Congenital onset	68 (69)	23 (37)	91 (57)
<i>Communication mode</i>			
Speech only or speech emphasis	40 (41)	19 (31)	59 (37)
Other	58 (59)	43 (69)	101 (63)
Bilateral cochlear implant status at 3 years post first implant	24 (24)	10 (16)	34 (21)

¹ SNHL, sensorineural hearing loss;

^{1 1} average 4-frequency hearing threshold (average of 0.5, 1, 2, and 4 kHz);

^{1 1 1} SRI-Q, speech recognition index in quiet;

[#] RDLS, Reynell Developmental Language Scale.

Table 2

Description of the key component features of the four subtests administered from the Comprehensive Assessment of Spoken Language (CASL).

Test	Ages (years)	What the test assesses	Instructions	Interpretation
Antonyms	Core 5–12 Supplemental 13–21	Word knowledge and language expression	Tell me a word that is opposite of _____.	Child needs to know the opposite of the stimulus word and demonstrate ability to retrieve and produce the opposite
Syntax construction	Core 3–10 Supplemental 11–21	Generation of sentence	Imitate, complete sentences, answer questions, formulate sentences in a narrative	Expressive ability in using morphosyntactic structures
Paragraph comprehension of syntax	Core 5–10 Supplemental 3–4 11–12	Comprehension of sentences varying in syntactic difficulty	Point to one of four pictures that represents the correct response	Comprehend narratives that have successive levels of syntactic difficulty
Pragmatic judgment	Core All ages	Judging the appropriateness of language used or using actual language in a situation	Series of vignettes describing every day events read out loud by the examiner	Difficulties relate to lack of experience in the use of pragmatically correct language or the ability to generalize from one situation to another

Table 3

Mean (95% confidence interval) standard scores on core composite and four subtests of the Comprehensive Assessment of Spoken Language (CASL) test associated with every year older in age at implantation. Mixed-effects modeling results of CASL data collected at 4-, 5-, or 6-years after cochlear implantation in 160 SNHL children who received their first cochlear implant before age of 5 years.

CASL	Age at implant			
	Unadjusted		Adjusted [†]	
	< 2.5 years	2.5–5 years	< 2.5 years	2.5–5 years
Core composite	–12.8 (–18.8, –6.8)	1.3 (–4.3, 7.1)	–7.3 (–12.7, –1.9)	–10.6 (–15.8, –5.5)
Subtest				
Antonyms	–8.1 (–13.3, –2.9)	1.8 (–3.5, 7.2)	–3.4 (–8.6, 1.7)	–7.8 (–13.0, –2.6)
Syntax construction	–14.0 (–20.0, –8.1)	1.5 (–4.2, 7.2)	–9.4 (–15.0, –3.8)	–10.5 (–16.6, –4.3)
Paragraph comprehension of syntax	–10.4 (–17.0, –3.7)	1.6 (–5.6, 8.8)	–6.3 (–12.9, 0.2)	–10.8 (–16.2, –5.4)
Pragmatic judgment	–14.2 (–19.8, –8.6)	0.2 (–5.0, 5.4)	–8.2 (–13.2, –3.1)	–10.1 (–15.6, –4.7)

[†] Adjusted for race, gender, ethnicity, baseline characteristics of study site, family income, maternal education, maternal sensitivity score, etiology of hearing loss, residual hearing (4-frequency pure-tone average), communication mode, verbal comprehension score of the Reynell Developmental Language Scales, speech recognition index score, and cognitive screening score, and bilateral implant status at 3 years after receiving the first implant.

Table 4

Multivariable-adjusted[†] mixed effects modeling analyses for the four subtests and core composite of the comprehensive Assessment of Spoken Language (CASL) acquired after 4-, 5- and 6-years experience with cochlear implant in 160 children who received their first implant at younger than 5 years of age.

Factors associated with differential change in standard scores	Antonyms Vocabulary			Syntax Construction Expression			Syntax Comprehension Reception			Pragmatic Judgment Pragmatics			Core Composite		
	Estimate	95% CI	P value	Estimate	95% CI	P value	Estimate	95% CI	P value	Estimate	95% CI	P value	Estimate	95% CI	P value
<i>Implantation age, per year</i>															
with 4 years CI experience	-5.7	-9.2 to -2.3	.0013	-11.9	-15.4 to -8.4	<.0001	-9.5	-13.2 to -5.9	<.0001	-10.1	-13.2 to -7.0	<.0001	-9.5	-12.6 to -6.5	<.0001
with 5 years CI experience	-6.0	-9.2 to -2.8	.0003	-9.1	-12.5 to -5.7	<.0001	-7.8	-11.7 to -3.8	.0002	-8.3	-11.4 to -5.3	<.0001	-8.4	-11.6 to -5.2	<.0001
with 6 years CI experience	-5.6	-8.6 to -2.7	.0003	-7.4	-11.2 to -3.7	.0002	-7.0	-11.0 to -3.1	.0006	-7.2	-10.2 to -4.1	<.0001	-7.6	-10.7 to -4.4	<.0001
Residual hearing, 4-frequency pure-tone average, per 20 dB	-0.5	-3.9 to 2.8	.75	-1.7	-5.1 to 1.7	.32	-4.0	-8.1 to 0.1	.06	-2.1	-5.4 to 1.3	.23	-1.6	-5.1 to 1.9	.38
Congenital onset	3.5	-1.9 to 8.9	.20	3.2	-2.0 to 8.3	.23	3.5	-2.3 to 9.3	.24	3.3	-1.6 to 8.2	.19	3.6	-1.4 to 8.6	.16
Developmental screening score for consistency, per 20 points	3.6	1.1 to 6.1	.0049	3.7	1.1 to 6.2	.0047	4.1	0.9 to 7.3	.01	3.9	1.6 to 6.2	.0012	4.3	1.7 to 6.9	.0016
Female gender	2.7	-2.7 to 7.9	.33	2.4	3.2 to 8.0	.39	6.0	-0.3 to 12.3	.06	3.8	-1.3 to 9.0	.14	4.1	-1.2 to 9.5	.13
Non-Caucasian	-0.9	-7.8 to 6.0	.79	-2.6	-8.7 to 3.6	.41	-0.5	-9.3 to 8.2	.90	-3.1	-9.1 to 2.9	.31	-3.4	-10.1 to 3.3	.31
Language comprehension [†]	0.9	0.6 to 1.3	<.0001	1.0	0.6 to 1.4	<.0001	1.0	0.5 to 1.4	<.0001	0.7	0.3 to 1.1	.0001	0.9	.6 to 1.3	<.0001
Parent-child interaction	5.6	2.1 to 9.1	.0018	6.2	2.5 to 10.0	.0014	6.9	3.2 to 10.6	.0004	6.8	3.7 to 9.9	<.0001	7.1	3.5 to 10.7	.0002
Speech versus other communication modes	2.1	-3.9 to 8.2	.049	4.5	-1.9 to 11.0	.17	2.5	-4.9 to 9.9	.50	4.0	-1.5 to 9.6	.15	3.5	-2.8 to 9.7	.28
Speech recognition index scores, per 100 pts	1.4	-2.7 to 5.6	.050	6.2	1.4 to 11.1	.01	6.1	1.4 to 10.8	.01	5.5	1.3 to 9.7	.01	5.2	0.5 to 9.8	.003

[†] Adjusted for all other variables in the table and further adjusted for ethnicity, family income, maternal education, type of hearing loss and clinical centers at baseline, and bilateral implant status at end of 3 years follow-up.

Comprehension score from the Reynell Developmental Language Scales (RDLS) at baseline.

+/−