Spoken Language Development in Children Following Cochlear Implantation

John K. Niparko, MD
Emily A. Tobey, PhD
Donna J. Thal, PhD
Laurie S. Eisenberg, PhD
Nae-Yuh Wang, PhD
Alexandra L. Quittner, PhD
Nancy E. Fink, MPH
for the CDaCI Investigative Team

Context Cochlear implantation 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 cochlear implantation in young children.

Design, Setting, and Participants Prospective, longitudinal, and multidimensional assessment of spoken language development over a 3-year period in children who underwent cochlear implantation 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 (Reynell Developmental Language Scales).

Results Children undergoing cochlear implantation showed greater improvement in spoken language performance (10.4; 95% confidence interval [CI], 9.6-11.2 points per year in comprehension; 8.4; 95% CI, 7.8-9.0 in expression) than would be predicted by their preimplantation baseline scores (5.4; 95% CI, 4.1-6.7, comprehension; 5.8; 95% CI, 4.6-7.0, expression), although mean scores were not restored to age-appropriate levels after 3 years. Younger age at cochlear implantation was associated with significantly steeper rate increases in comprehension (1.1; 95% CI, 0.5-1.7 points per year younger) and expression (1.0; 95% CI, 0.6-1.5 points per year younger). Similarly, each 1-year shorter history of hearing deficit was associated with steeper rate increases in comprehension (0.8; 95% CI, 0.2-1.2 points per year shorter) and expression (0.6; 95% CI, 0.2-1.0 points per year shorter). In multivariable analyses, greater residual hearing prior to cochlear implantation, higher ratings of parent-child interactions, and higher socioeconomic status were associated with greater rates of improvement in comprehension and expression.

Conclusion The use of cochlear implants in young children was associated with better spoken language learning than would be predicted from their preimplantation scores.

JAMA. 2010;303(15):1498-1506 www.jama.com

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). More than half of such children are treated with cochlear implantation.[1] Cochlear implant systems comprise 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 an 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.[2] With experience, children understand speech, environmental sounds, and music with varying degrees of success.[3-6]

Intervening at early ages with cochlear implantation is predicated on behavioral data suggesting language performance is more accurate the earlier children receive implants.[3-8] Early implantation may take advantage of neuronal flexibility inherent in critical periods of auditory-based learning.[9] Safety and technical concerns regarding early implantation have been addressed with continued refinements of medical and surgical approaches.[10] Support for early implantation, however, must be tempered until sufficient longitudinal data are available. Behavioral studies supporting cochlear implantation primarily use retrospective and case-series designs[11] and variance in observed results is notoriously high.[3-7] As a consequence, the timing of cochlear implantation, especially the level of hearing

Author Affiliations: Departments of Otolaryngology (Dr Niparko) and Medicine (Dr Wang), Johns Hopkins University School of Medicine, Baltimore, Maryland; Callier Center for Communication Disorders, University of Texas at Dallas (Dr Tobey); School of Speech, Language, and Hearing Sciences, University of California, San Diego (Dr Thal); House Ear Institute, Los Angeles, California (Dr Eisenberg); Department of Psychology, University of Miami, Miami, Florida (Dr Quittner); and Departments of Epidemiology (Dr Niparko and Ms Fink) and Biostatistics (Dr Wang), Johns Hopkins Bloomberg School of Public Health, Baltimore.
The CDaCI Investigators are listed in the eAppendix at http://www.jama.com.
Corresponding Author: John K. Niparko, MD, The Johns Hopkins Hospital, 601 N Caroline St, JHOC-6161A, Baltimore, MD 21287 (jnipark1@jhmi.edu).

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loss and the associated delays in spoken language that should prompt implantation, remains unspecified.

To better understand the factors that promote spoken language development after early cochlear implantation, the Childhood Development after Cochlear Implantation (CDaCI) investigative team initiated a prospective study of spoken language outcomes in a cohort of children who underwent implantation prior to age 5 years.[12-15]

METHODS

Details of the study design have been published previously.[14] Participants were enrolled between November 2002 and December 2004; 3-year follow-up was completed between November 2005 and May 2008. Children with SNHL were enrolled prior to cochlear implantation through 6 large implant centers situated in different regions of the United States. 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 parents of each enrolled child.

Study Population

For the experimental group, children younger than 5 years with severe to profound SNHL were consecutively screened for cochlear implantation 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 cochlear implantation were enrolled based on developmental criteria. A normal-hearing group served as a reference for longitudinal assessment of language development. Normal-hearing children had to be within the age range of children undergoing cochlear implantation and meet the same developmental criteria.

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

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 mean threshold for the better-hearing ear served as a proxy for residual hearing.

A battery of tests (eTable, available at http://www.jama.com) was administered at each data point. In addition to RDLS scores, measures of speech recognition and videotaped parent-child interactions were collected and coded to assess their covariation with RDLS performance. All measures were administered before cochlear implantation (baseline) and at follow-up visits scheduled for 6, 12, 24, and 36 months after implant activation. For hearing children, follow-up visits were scheduled for the same time intervals but anchored at 6 weeks after baseline to correspond to the delay imposed by postsurgical healing and activation of the implant in children who underwent cochlear implantation.

Main Outcome Measures

The RDLS[18,19] comprehension and expressive language scales were administered as interdependent measures of spoken language performance. Age-equivalent language level (language age) was determined based on RDLS scores. If language level was "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 was 0; if scores matched those expected of a child 1 year younger, the gap was 1 year.

Secondary exploratory measures addressed emergent speech recognition and characteristics of parent-child interactions. Speech recognition was represented by the Speech Recognition Index (SRI), which summarizes scores collected from a hierarchical battery of speech recognition tests and was developed for this study to track speech recognition performance via growth curve analysis.[13,20] Videos of parent-child interactions were coded for child autonomy, positive regard, cognitive stimulation, shared visual attention, and bidirectional interaction (eTable).[12,21]

Statistical Analysis

A sample size of 90 participants per group was based on 80% statistical power to detect a 1.3-point-per-year increase in RDLS raw score between 3 equal-sized (2 cochlear implantation and 1 normal-hearing) subgroups in a 3-year, longitudinal analysis. The parameters for the calculation were adopted from reported RDLS scores of children undergoing cochlear implantation.[14,18,22]

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 cochlear implantation and those serving as hearing controls were tested using analysis of variance for continuous variables and χ^2 tests for categorical variables. Stratified analyses based on age at implantation were conducted for the cochlear implantation group to identify factors associated with age at implantation and to explore post–cochlear implantation spoken language development and language age gaps.

Developmental trajectories of spoken language in different subgroups

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were explored using nonparametric regression with locally weighted smoothing scatterplot (lowess)[23] 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 nonlinear 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 covariates were modeled as fixed effects. Follow-up time, based on actual visit dates, provided the time variable.

Covariates 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 with normal hearing, SNHL, length of traditional amplification, age at implantation, mode of communication, and bilateral cochlear implantation. Emergent speech recognition and bilateral cochlear implantation were treated as time-dependent covariates. Associations were adjusted for demographic characteristics and other baseline variables related to center, sex, race, ethnicity, maternal education, hearing onset, cognition, baseline RDLS scores, and time to implant activation. We performed sensitivity analyses evaluating the effect of censoring those children who underwent bilateral cochlear implantation after their second implant.

The rate of language development, as modified by a given covariate, was modeled by including the cross-product of the follow-up time × covariate as an interaction term. All longitudinal analyses were adjusted for center. SAS version 9.1 (SAS Institute, Cary, North Carolina) was used for all analyses, and all tests were 2-sided ($\alpha = .05$).

RESULTS

Study Population

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

TABLE 1 shows baseline measures of hearing status, child and family demographics, and language status. Children undergoing cochlear implantation and hearing controls differed in family income, scores of RDLS comprehension and expression, parents' perception of communication behaviors (measured by Meaningful Auditory Integration Scale, or MAIS),[24,25] ratings of parent-child interactions, and cognition, as well as hearing thresholds.

Children undergoing cochlear implantation were stratified into 3 groups by age at implantation: younger than 18 months (n=72; 38%); 18 to 36 months (n=64; 34%), and older than 36 months (n=52; 28%). These 3 groups demonstrated significantly different lengths of time spent with hearing, SNHL, and amplification (Table 1). There were significant differences in their baseline RDLS comprehension and expression and MAIS scores, sex, 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.

Spoken Language: Comprehension and Expression

FIGURE 1 and interactive graphs (available at http://www.jama.com) demonstrate the trajectories of raw score changes on the RDLS comprehension and expressive language scales over the 3-year follow-up. Children who underwent cochlear implantation demonstrated slower and more variable language trajectories compared with hearing children. However, children undergoing implantation produced steeper trajectories (10.4; 95% confidence interval [CI], 9.6-11.2, points per year in comprehension; 8.4; 95% CI, 7.8-9.0, in expression) than those predicted by their baseline comprehension and expression scores (5.4; 95% CI, 4.1-6.7, comprehension; 5.8; 95% CI, 4.6-7.0, expression). Significantly higher rates of comprehension and expression were noted in children undergoing implantation at younger than 18 months compared with children undergoing implantation at ages between 18 and 36 months and at older than 36 months. The majority of children who received implants prior to age 18 months revealed trajectories of improvement that paralleled those of hearing controls. Cochlear implantation after 18 months of age was associated with less favorable trajectories of improvement 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 cochlear implantation had a mean deficit of 22.3 (95% CI, 19.4-25.2) points in comprehension and 19.8 (95% CI, 17.3-22.3) points in expression compared with hearing peers after 3 years. Younger age at cochlear implantation was associated with significantly steeper rate increases in comprehension (1.1; 95% CI, 0.5-1.7 points per year younger) and expression (1.0; 95% CI, 0.6-1.5 points per year younger). When stratified by age, the mean deficit in comprehension scores for children undergoing implantation was 8.1 (95% CI, 6.2-9.9) for those receiving implants at younger than 18 months, 27.0 (95% CI, 23.8-30.1) for those receiving implants between ages 18 and 36 months, and 38.7 (95% CI, 34.2-43.2) for those receiving implants at older than 36 months. The mean deficit in expression scores for children undergoing cochlear im-

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plantation was 8.2 (95% CI, 6.4-9.9) for those receiving implants at younger than 18 months, 21.7 (95% CI, 19.3-24.1) for those receiving implants between ages 18 and 36 months, and 29.4 (95% CI, 24.1-34.7) for those receiving implants after age 36 months.

eFigure 1 (available at http://www.jama.com) demonstrates the mean trajectories of improvement in comprehension and expression scores. Faster mean rates of improvement in verbal comprehension and language expression trajectories were associated with earlier age at cochlear implantation (age at implant, <18 months).

TABLE 2 compares the children's chronologic ages with their language age equivalents. Whereas the gap in language development between children undergoing implantation at younger than 18 months and normal-hearing children did not widen during follow-up, larger gaps accrued in children who received implants at older ages.

Speech Recognition

Trajectories of change in speech recognition capacity are shown in eFigure 2. Children undergoing cochlear implan-

Table 1. Baseline Characteristics of Participants in the CDaCI Study

	Children Undergoing Cochlear Implantation				
	Age at Implantation, mo				
Characteristic	<18 (n=72)	18-36 (n=64)	>36-60 (n=52)	All (n = 188)[a]	Children With Normal Hearing (n=97)[a]
Age at baseline, mean (SD), mo[b]	12.6 (3.1)	27.1 (5.5)	46.1 (7.3)	26.7 (14.5)	27.5 (13.1)
Age at SNHL diagnosis, mean (SD), mo[b]	3.8 (4.0)	12.2 (8.5)	18.0 (13.1)	10.6 (10.5)	NA
Time with hearing before implantation, mean (SD), mo[b]	0.7 (2.4)	1.6 (4.1)	7.4 (12.2)	2.8 (7.4)	NA
Median (IQR)	0.0 (0.0-0.0)	0.0 (0.0-0.0)	0.0 (0.0-12.0)	0.0 (0.0-0.0)	NA
Percentage of time with hearing before implantation, mean (SD)	4.9 (15.8)	6.1 (15.6)	16.0 (26.8)	8.2 (19.8)	NA
Time with hearing loss, mean (SD), mo[b]	5.0 (3.4)	13.4 (7.8)	13.3 (10.3)	10.1 (8.4)	NA
Median (IQR)	3.0 (4.0-6.0)	13.0 (6.0-20.0)	12.0 (4.0-21.5)	6.0 (3.0-15.5)	NA
Percentage of time with hearing loss, mean (SD)	39.1 (21.6)	50.4 (27.9)	29.8 (24.0)	40.4 (25.7)	NA
Time with amplification, mean (SD), mo[b]	6.8 (3.4)	11.9 (8.9)	24.9 (15.3)	13.5 (12.1)	NA
Median (IQR)	7.0 (4.4-9.3)	5.3 (2.2-10.0)	23.3 (12.9-38.9)	9.5 (5.3-17.8)	NA
Percentage of time with amplification, mean (SD)	55.7 (25.3)	43.0 (29.5)	52.3 (28.9)	50.4 (28.2)	NA
Age at activation, mean (SD), mo[b]	15.5 (3.2)	29.4 (5.6)	48.5 (7.4)	29.4 (14.4)	NA
4-Tone hearing threshold in better ear, mean (SD), dB[b]	108.9 (15.6)	105.1 (15.6)	100.0 (17.4)	105.1 (16.4)	14.2 (7.1)[d]
RDLS comprehension score, mean (SD)[b]	0.8 (1.7)	3.9 (6.4)	13.5 (13.4)	5.8 (9.8)	30.1 (18.3)[d]
RDLS expression score, mean (SD)[b]	3.4 (1.8)	8.0 (6.2)	17.9 (12.9)	9.5 (10.0)	27.6 (15.2)[d]
MAIS score, mean (SD)[b]	12.4 (15.0)	22.7 (23.0)	38.0 (31.4)	23.0 (25.3)	97.1 (7.7)[d]
Parent-child interaction score, mean (SD)	5.4 (0.7)	5.2 (0.8)	5.1 (0.6)	5.2 (0.7)	5.7 (0.6)[d]
Cognitive status score, mean (SD)					
Bayley PDI (<2 y)[b]	94.7 (18.3) [n=61]	95.7 (19.4) [n=51]	79.5 (18.3) [n=18]	93.0 (19.4)	108.2 (14.7)[d] [n=41]
Leiter-R brief IQ (>2 y)	102.5 (21.7) [n=10]	100.3 (13.4) [n=12]	106.3 (21.0) [n=33]	104.3 (19.5)	109.4 (11.5) [n=53]
Combined[c]	97.6 (19.9)	97.7 (19.5)	93.4 (24.9)	96.5 (21.2)	106.9 (15.1)[d]
White, No. (%)[e]	58 (81)	42 (66)	40 (77)	140 (74)	77 (79)
Female, No. (%)[b]	32 (44)	31 (48)	35 (67)	98 (52)	60 (62)
Maternal education, HS graduate, No. (%)[b]	70 (97)	60 (94)	43 (83)	173 (93)	91 (95)
Hispanic, No. (%)[e]	9 (13)	17 (27)	11 (22)	37 (20)	9 (10)
Household income ≥$50 000, No. (%)[b]	43 (60)	24 (38)	21 (40)	88 (46)	76 (78)[d]
Congenital SNHL, No. (%)[b]	56 (78)	31 (51)	18 (38)	105 (56)	NA
Communication mode, No. (%)[b]					
Speech only	19 (26)	8 (13)	10 (19)	37 (20)	NA
Speech/sign: speech emphasis[b]	14 (19)	13 (20)	8 (15)	35 (19)	NA
Speech/sign: sign emphasis[b]	1 (1)	4 (6)	7 (13)	12 (6)	NA
Sign only	12 (17)	19 (30)	4 (8)	35 (19)	NA
Other/no response	26 (36)	20 (31)	23 (44)	69 (37)	NA

Abbreviations: Bayley PDI, Bayley Psychomotor Developmental Index; CDaCI, Childhood Development after Cochlear Implantation; HS, high school; IQR, interquartile range; Leiter-R, Leiter International Performance Scale–Revised; MAIS, Meaningful Auditory Integration Scale; NA, not applicable; RDLS, Reynell Developmental Language Scales; SNHL, sensorineural hearing loss.

[a] Age range of all children in implantation and control groups is 9 months to 5 years.

[b] Differences among children undergoing cochlear implantation at <18 months; 18-36 months; and >36 months ($P<.05$).

[c] Cognitive status measured by Bayley PDI score for children <24 months of age and by Leiter-R brief IQ score for children ≥24 months of age.

[d] Differences between children undergoing cochlear implantation and children with normal hearing ($P<.05$).

[e] Race/ethnicity classifications were based on parental reporting in a written registration form using categories determined by National Institutes of Health policy.

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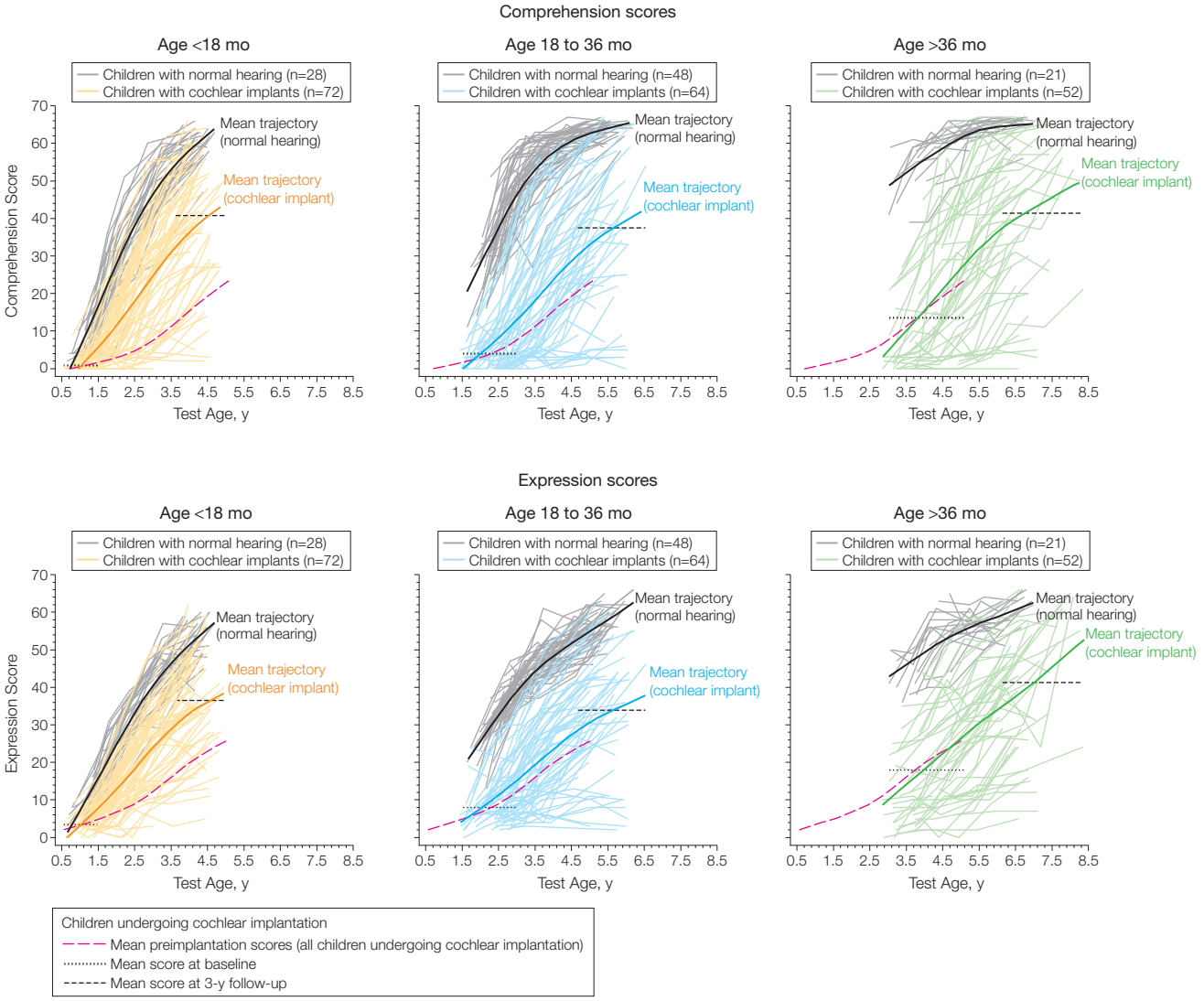
tation showed mean rates of progress through the SRI hierarchy of speech recognition measures that were parallel to that of normal-hearing children.

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 mean scores for the normal-hearing children at baseline (at their enrollment age of 2.3 years). This reference comprehension score was attained at 3.4, 4.7, and 5.3 years for children undergoing implantation at younger than 18 months, 18 to 36 months, and older than 36 months, respectively, and for verbal expression, at 3.4, 4.5, and 5.2 years, respectively.

Multivariable Model

Independent associations of child, family, and treatment variables with improvement in comprehension and expressive skills are shown in TABLE 3. Increase in RDLS comprehension score was positively associated with the amount of preimplantation residual hearing. Better baseline hearing thresholds (eg, 85-dB vs 105-dB loss) were associated with greater improvement in comprehension (2.23; 95% CI, 0.25-4.31; $P = .03$) and expression (2.24; 95% CI, 0.27-4.20; $P = .03$). Similarly, each 1-year shorter history of hearing deficit was associated with steeper increases in comprehension (0.8; 95% CI, 0.2-1.2 points per year) and expression (0.6; 95% CI, 0.2-1.0 points per year). In multivariable analyses, greater residual hearing prior to implantation, higher

Figure 1. Developmental Trajectories of RDLS Raw Scores of Comprehension and Expression Grouped by Age at Baseline



Widths of the horizontal dashed and dotted lines representing Reynell Developmental Language Scales (RDLS) scores span the age ranges at time of testing. Interactive graphs are available at http://www.jama.com. Preimplant trajectories were nonparametrically estimated based on cross-sectional data from baseline. Other trajectories were nonparametrically estimated based on longitudinal data over 3 years of follow-up.

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ratings of parent-child interactions, and higher socioeconomic status were associated with greater rates of improvement in comprehension and expression. Although higher RDLS comprehension scores at baseline were associated with higher comprehension scores over the course of follow-up ($P<.001$), baseline comprehension was not significantly related to the rate of comprehension development.

Improvement in comprehension was not associated with sex, congenital onset of SNHL, or baseline cognition level (Table 3). Improvement in speech recognition was significantly associated with improvements in verbal language (3.76; 95% CI, 3.07-4.45 points in comprehension per 100-point increase in SRI score; $P<.001$; 1.98; 95% CI, 1.41-2.45 points in expression; $P<.001$).

Higher parent-child interaction scores were significantly associated with higher rate increases of comprehension (3.75 points; 95% CI, 1.77-5.73; $P<.001$) and expression (3.45 points; 95% CI, 1.60-5.29; $P<.001$). Although lower family income was strongly associated with reduced rates of improvement in comprehension and expression using bivariable analyses (both $P<.001$), these negative asso-

Table 2. Estimates of Language Age for Children Undergoing Cochlear Implantation and Children With Normal Hearing at Baseline and 3-Year Follow-up Based on Comprehension and Expression[a]

	Children Undergoing Cochlear Implantation						
	<12 mo[b]	12-18 mo	<18 mo	18-36 mo	>36-60 mo	All	Children With Normal Hearing
Baseline	(n=33)	(n=39)	(n=72)	(n=64)	(n=52)	(n=188)	(n=97)
Chronologic age	9.8 (6.8 to 12.8)	14.9 (12.2 to 17.7)	12.6 (10.5 to 14.6)	27.1 (24.9 to 29.3)	46.1 (43.7 to 48.5)	26.2 (24.2 to 28.1)	27.5 (24.7 to 30.3)
Comprehension, No.	21	34	55	59	50	164	97
Language age, mo		<13.0[c]		13.7 (11.5 to 16.0)	18.3 (15.8 to 20.7)	14.8 (13.5 to 16.2)	28.6 (26.8 to 30.4)
CDaCI comprehension age, mo[d]		9.6 (6.2 to 12.9)		11.5 (9.0 to 13.9)	17.6 (14.9 to 20.3)	12.7 (11.1 to 14.2)	29.2 (27.2 to 31.2)
Age gap, chronologic – language				12.8 (11.4 to 14.3)	27.2 (25.6 to 28.7)	12.9 (11.3 to 14.5)	–0.3 (–2.4 to 1.7)
Age gap, chronologic – CDaCI		5.6 (3.7 to 7.5)		15.1 (13.7 to 16.6)	27.8 (26.2 to 29.3)	15.1 (13.7 to 16.6)	–1.0 (–2.9 to 0.9)
Expression, No.	21	34	55	59	50	164	97
Language age, mo		<16.0[c]		16.5 (14.5 to 18.6)	21.2 (19.0 to 23.4)	17.7 (16.5 to 19.0)	27.6 (25.9 to 29.2)
CDaCI expressive age, mo[d]		8.2 (4.5 to 11.9)		11.9 (9.1 to 14.7)	20.4 (17.4 to 23.4)	13.2 (11.5 to 15.0)	29.8 (27.5 to 32.1)
Age gap, chronologic – language				10.0 (8.6 to 11.5)	24.2 (22.6 to 25.8)	10.0 (8.4 to 11.6)	0.7 (–1.4 to 2.8)
Age gap, chronologic – CDaCI		7.0 (4.7 to 9.3)		14.7 (13.0 to 16.4)	25.0 (23.2 to 26.9)	14.6 (13.2 to 16.0)	–1.6 (–3.4 to 0.3)
36-Month follow-up	(n=32)	(n=37)	(n=69)	(n=61)	(n=50)	(n=180)	(n=84)
Chronologic age	49.5 (46.5 to 52.5)	53.5 (50.7 to 56.4)	51.6 (49.5 to 53.7)	65.7 (63.5 to 67.9)	85.0 (82.6 to 87.4)	65.8 (63.7 to 67.9)	64.1 (61.0 to 67.2)
Comprehension, No.	31	37	68	59	41	168	84
Language age, mo	35.5 (31.2 to 39.9)	35.2 (31.3 to 39.1)	35.4 (32.5 to 38.3)	33.9 (30.7 to 37.0)	37.2 (33.6 to 40.9)	35.3 (33.5 to 37.2)	58.0 (55.4 to 60.5)
CDaCI comprehension age, mo[d]	35.7 (31.8 to 39.6)	35.0 (31.4 to 38.6)	35.3 (32.7 to 38.0)	33.5 (30.7 to 36.4)	35.7 (32.3 to 39.1)	34.8 (33.1 to 36.5)	59.3 (56.9 to 61.7)
Age gap, chronologic – language	13.6 (8.4 to 18.8)	17.2 (12.4 to 22.0)	15.5 (12.0 to 19.1)	32.0 (28.2 to 35.8)	46.3 (41.9 to 50.7)	29.2 (26.4 to 31.9)	5.9 (2.1 to 9.8)
Age gap, chronologic – CDaCI	13.7 (9.8 to 17.6)	17.6 (13.9 to 21.3)	15.8 (13.1 to 18.5)	32.2 (29.4 to 35.1)	47.8 (44.4 to 51.3)	29.5 (27.2 to 31.9)	4.5 (1.2 to 7.9)
Expression, No.	31	36	67	59	41	167	84
Language age, mo	34.4 (30.1 to 38.8)	33.5 (29.5 to 37.5)	33.9 (31.0 to 36.9)	33.4 (30.2 to 36.6)	40.4 (36.7 to 44.0)	35.4 (33.5 to 37.3)	56.2 (53.6 to 58.9)
CDaCI expressive age, mo[d]	35.8 (31.6 to 40.0)	34.2 (32.0 to 39.8)	35.9 (33.0 to 38.7)	34.2 (31.2 to 37.3)	40.2 (36.6 to 43.8)	36.3 (34.5 to 38.2)	59.4 (56.8 to 62.0)
Age gap, chronologic – language	14.7 (9.7 to 19.7)	19.2 (14.5 to 23.9)	17.1 (13.7 to 20.5)	32.5 (28.8 to 36.1)	43.4 (39.2 to 47.6)	29.3 (26.7 to 31.8)	7.5 (3.9 to 11.1)
Age gap, chronologic – CDaCI	13.6 (9.5 to 17.8)	16.9 (12.9 to 20.9)	15.3 (12.5 to 18.2)	31.5 (28.5 to 34.5)	43.6 (40.0 to 47.2)	28.1 (25.8 to 30.4)	4.5 (1.2 to 7.8)

Abbreviations: CDaCI, Childhood Development after Cochlear Implantation; RDLS, Reynell Developmental Language Scales.
[a] Values are mean (95% confidence interval) unless otherwise indicated.
[b] The RDLS is not validated for ages younger than 12 months; hence, spoken language development is numerically not ascertainable through RDLS for the youngest group at baseline.
[c] 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.
[d] The CDaCI comprehension age and CDaCI expression age are scored according to the estimated mean ages when children with normal hearing in the CDaCI study attained these respective RDLS comprehension and expression scores.

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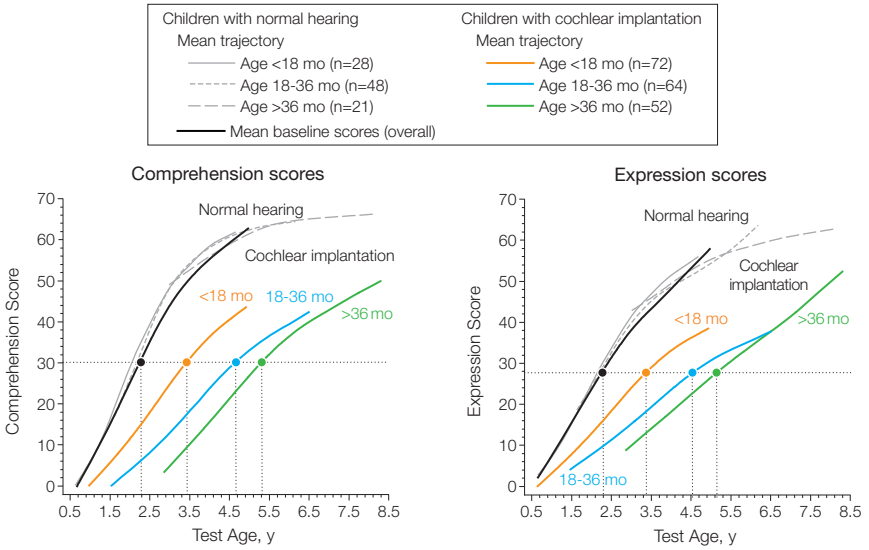
Figure 2. Nonparametric Fit of Reynell Developmental Language Scales Raw Scores of Comprehension and Expression Stratified by Age at Baseline and Test Age



For comparative assessment, the horizontal dotted 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 subgroups of children undergoing cochlear implantation at different ages. 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 normal-hearing children and among children undergoing cochlear implantation, 3.4 years for children younger than 18 months at implant, 4.7 years for those aged 18 to 36 months at implant, and 5.3 years for those older than 36 months at implant. On the expression scale, the ages were 2.3 years for hearing children and among children undergoing cochlear implantation, 3.4 years for children younger than 18 months at implant, 4.5 years for those aged 18 to 36 months at implant, and 5.2 years for those older than 36 months at implant.

ciations were either attenuated (comprehension, −2.2 points; 95% CI, −3.93 to −0.47; $P=.01$) or diminished (expression, $P=.33$) after adjustment.

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 preimplant amplification. A reduced rate of language development after cochlear implantation was associated with longer periods of hearing loss, without or with preimplantation amplification. Prolonged periods of hearing loss without and with amplification contributed to an older age at implantation and were associated with slower improvement in language comprehension. eFigure 3 highlights the slower improvement in comprehension and expression associated with extended hearing aid use in children who underwent cochlear implantation at a later age.

Exclusive use of spoken communication at baseline was not significantly associated with RDLS score increase in children undergoing implantation (Table 3). Center was found to be significantly associated with different rates of increase in comprehension scores. Bilateral implantation was not associated with an increase in RDLS comprehension and expression scores compared with unilateral implantation after adjusting for other variables. Results from the multivariable analyses in Table 3 (except bilateral status) were unchanged for 3-year comprehension and expression after censoring outcome measures after the second implantation.

COMMENT

Parents commonly seek cochlear implantation because they want their children with SNHL to hear and speak like children with normal hearing.[14,26] Language learning through listening and speaking serves as an effective marker of later school performance in children with normal hearing.[4,27]

Cochlear implantation is associated with significant improvement in comprehension and expression of spoken language over the first 3 years of implant use. Development of spoken language was positively associated with younger, as opposed to older, age at implantation and greater residual hearing prior to implantation. On average, results also revealed that gaps in spoken language development between normal-hearing children and those who underwent implantation were not eliminated in the 3 years.

Consistent with the critical period concept for language learning, early implantation in infants and toddlers was associated with significantly accelerated spoken language learning. Two indices of spoken language development revealed the association between age at cochlear implantation and spoken language outcome: performance scores in children who received implants younger were closer to scores of normal-hearing controls and older age at implantation was associated with greater gaps between chronological and language ages.

The rate of improvement in performance on spoken language measures was less steep in children undergoing cochlear implantation at later ages. At birth, normal-hearing infants discriminate speech sounds used in all languages.[28] 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.[29,30] Infants between 8 and 18 months of age begin to identify the patterns contributing to the words of their language, which in turn makes learning word meanings possible.[31] Sound pattern learning requires a neural commitment to the acoustic properties of a native language.[32] Children with early severe to profound SNHL do not experience similar neural commitments, so spoken language development is altered.

The decision to pursue cochlear implantation must weigh the potential benefit of implantation vs continued

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amplification of residual hearing. We observed that higher language scores at baseline were associated with greater residual hearing prior to implantation. However, significantly reduced language learning was associated with the prolonged use of hearing aids prior to implantation. These findings suggest that delaying implantation to extend hearing aid use for children with severe to profound hearing loss may be detrimental to language development following cochlear implantation. Spoken language learning relies on effective hearing. Close monitoring of performance with hearing aids can determine whether speech is effectively amplified to allow spoken language acquisition to progress.

Although spoken language outcomes were significantly associated with age at implantation and residual hearing, associations with environmental factors were also evident. Maternal engagement in early communication reflected in greater scores of parent-child interactions was associated with increased development in spoken language skills. Language comprehension and expression are influenced by parent-child interactions in bidirectional spoken communication.[21,33,34] 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.[35]

Family income above $50 000 was associated with better language performance at baseline. This factor was also favorably associated with accelerated language comprehension. These findings are consistent with studies of poorer language development experienced by children of low-income families.[27] The notion that children reared in disadvantaged environments may have fewer early experiences that are associated with optimal language development may extend to children undergoing cochlear implantation. 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 the study deserve comment. The observational design and the absence of a control group of SNHL children without cochlear implantation preclude causal conclusions. For ethical reasons, we could not randomize or match children with similar levels of SNHL who continued to use hearing aids. Such trials could formally test the efficacy of cochlear implantation in children who receive implants at different ages or stages of linguistic development. Instead, baseline, preimplantation performance across candidates provided an estimate of language learning trajectory for children with severe to profound SNHL and without cochlear implants. Although we used rigorous adjust-

Table 3. Multivariable-Adjusted Mixed-Effects Modeling Analyses for Children Undergoing Cochlear Implantation[a]

	Comprehension		Expression	
	Estimate (95% CI)	*P* Value	Estimate (95% CI)	*P* Value
Factors Associated With RDLS Raw Scores at Baseline				
Child characteristic				
Mean hearing threshold, per 20-dB increase	–0.31 (–1.11 to 0.49)	.45	–0.17 (–0.75 to –0.41)	.57
Family characteristics				
Mean rating of parent-child interactions, per point increase	0.51 (–0.15 to 1.16)	.13	0.59 (0.06 to 1.12)	.03
Self-reported income <$50 000[b]	0.28 (–0.41 to 0.96)	.43	–0.24 (–0.72 to 0.24)	.32
Intervention characteristics				
Time with hearing, per 6-mo increase	0.71 (0.05 to 1.38)	.04	0.62 (0.24 to 0.99)	.001
Time with hearing loss (prior to amplification), per 6-mo increase	0.01 (–0.42 to 0.44)	.97	0.07 (–0.25 to 0.39)	.68
Time with amplification, per 6-mo increase	–0.14 (–0.49 to 0.22)	.44	–0.02 (–0.32 to 0.28)	.89
Spoken communication mode[b]	0.08 (–0.52 to 0.69)	.78	–0.14 (–0.53 to 0.26)	.50
Factors Associated With Differential Rate Increase in RDLS Raw Scores Over 3 Years				
Child characteristics				
Mean hearing threshold, per 20-dB increase	–2.28 (–4.31 to –0.25)	.03	–2.24 (–4.20 to –0.27)	.03
SRI score, per 100-point increase[c]	3.76 (3.07 to 4.45)	<.001	1.98 (1.41 to 2.54)	<.001
Family characteristics				
Mean rating of parent-child interactions, per point increase	3.75 (1.77 to 5.73)	<.001	3.45 (1.60 to 5.29)	<.001
Self-reported income <$50 000[b]	–2.20 (–3.93 to –0.47)	.01	–0.82 (–2.45 to 0.82)	.33
Intervention characteristics				
Time with hearing, per 6-mo increase	–1.45 (–2.70 to –0.19)	.02	–2.20 (–3.60 to –0.81)	.002
Time with hearing loss (prior to amplification), per 6-mo increase	–0.74 (–1.91 to 0.43)	.21	–0.51 (–1.63 to 0.60)	.37
Time with amplification, per 6-mo increase	–1.31 (–2.11 to –0.52)	.001	–1.12 (–1.79 to –0.44)	.001
Spoken communication mode[b]	1.57 (–0.22 to 3.35)	.09	1.69 (–0.02 to 3.41)	.05
Bilateral cochlear implantation status, time-dependent	2.19 (–0.86 to 2.55)	.11	1.67 (–0.94 to 4.27)	.21

Abbreviations: CI, confidence interval; RDLS, Reynell Developmental Language Scales; SRI, Speech Recognition Index.
[a] Each variable is adjusted for study center, sex, race, ethnicity, maternal education (high school graduate or not), hearing onset, cognition, RDLS raw scores at baseline, time to implant activation, time-dependent bilateral implant status, and all other variables listed in the table.
[b] As measured at baseline.
[c] Speech Recognition Index score was included as a time-dependent variable. The mean increase in SRI scores over the 3-year follow-up period was 349 points, moving from a mean of 45 points at baseline to 395 points after 3 years.

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ment procedures to mitigate the impact of potential confounders, residual confounding cannot be ruled out.

Although bilateral cochlear implantation was associated with nonsignificantly improved rates of spoken language development statistically, this observation must be viewed cautiously given the brief period between the second implantation and the end of follow-up. Continued assessment will determine how such factors are related to long-term trends in the acquisition of spoken language skills.

The generalizability of results beyond major implant centers is uncertain. The representativeness of the children with SNHL was likely influenced by access to participating centers and the 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. Although not determinative, age at implantation and residual hearing were associated with rate increases in 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 spoken language acquisition and guide timely intervention with cochlear implantation.

Author Contributions: Dr Niparko had full access to all of the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analysis.
Study concept and design: Niparko, Tobey, Thal, Eisenberg, Wang, Quittner, Fink.
Acquisition of data: Tobey, Eisenberg, Quittner, Fink.
Analysis and interpretation of data: Niparko, Tobey, Thal, Eisenberg, Wang, Quittner, Fink.
Drafting of the manuscript: Niparko, Tobey, Wang, Quittner, Fink.
Critical revision of the manuscript for important intellectual content: Niparko, Tobey, Thal, Eisenberg, Wang, Fink.
Statistical analysis: Wang.
Obtained funding: Niparko, Tobey, Wang, Quittner, Fink.
Administrative, technical, or material support: Niparko, Tobey, Eisenberg, Quittner, Fink.
Study supervision: Niparko, Tobey, Thal, Quittner, Fink.
Financial Disclosures: Dr Niparko reported serving on advisory boards without remuneration for 2 cochlear implant manufacturers, Advanced Bionics Corporations and the Cochlear Corporation, and serving on the board of directors for a school for children with hearing loss that has received gifts from cochlear implant manufacturers. The terms of these arrangements 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. No other disclosures were reported.
Funding/Support: The CDaCI was supported by grant R01 DC004797 from the National Institute on Deafness and Other Communication Disorders, the CityBridge Foundation, and the Sidgmore Family Foundation. Warranties on the implant devices used by children with implants in this study were discounted by 50% by the Advanced Bionics Corporation, Cochlear Corporation, and the MedEl Corporation.
Role of the Sponsor: The sponsors had no role in the design and conduct of the study; in the collection, analysis, and interpretation of the data; or in the preparation, review, or approval of the manuscript.
Online-Only Material: The eAppendix; eTable; eFigures 1, 2, and 3; and interactive graphs are available at http://www.jama.com.

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