Original research paper

Preschool speech intelligibility and vocabulary skills predict long-term speech and language outcomes following cochlear implantation in early childhood

Irina Castellanos[1], William G Kronenberger[2], Jessica Beer[1], Shirley C Henning[1], Bethany G Colson[1], David B Pisoni[1]

[1]DeVault Otologic Research Laboratory, Department of Otolaryngology Head and Neck Surgery, Indiana University School of Medicine, Indianapolis, IN, USA, [2]Riley Child and Adolescent Psychiatry Clinic, Department of Psychiatry, Indiana University School of Medicine, Indianapolis, IN, USA

Speech and language measures during grade school predict adolescent speech-language outcomes in children who receive cochlear implants (CIs), but no research has examined whether speech and language functioning at even younger ages is predictive of long-term outcomes in this population. The purpose of this study was to examine whether early preschool measures of speech and language performance predict speech-language functioning in long-term users of CIs. Early measures of speech intelligibility and receptive vocabulary (obtained during preschool ages of 3–6 years) in a sample of 35 prelingually deaf, early-implanted children predicted speech perception, language, and verbal working memory skills up to 18 years later. Age of onset of deafness and age at implantation added additional variance to preschool speech intelligibility in predicting some long-term outcome scores, but the relationship between preschool speech-language skills and later speech-language outcomes was not significantly attenuated by the addition of these hearing history variables. These findings suggest that speech and language development during the preschool years is predictive of long-term speech and language functioning in early-implanted, prelingually deaf children. As a result, measures of speech-language functioning at preschool ages can be used to identify and adjust interventions for very young CI users who may be at long-term risk for suboptimal speech and language outcomes.

Keywords: Cochlear implant, Preschool measures, Long-term outcomes, Speech and language, Speech intelligibility, Receptive language

Cochlear implants (CIs) provide profoundly deaf children with access to sound and spoken language during a period of dynamic brain plasticity, resulting in significant gains in speech and language skills (Niparko *et al.*, 2010). The FDA approved CIs as a treatment option for deaf children with profound hearing loss in 1990 and only recently has information about the long-term speech and language outcomes of prelingually deaf children who receive CIs during early childhood become available. Investigation of long-term outcomes following cochlear implantation is of considerable importance because the effectiveness of any treatment depends not only on short-term gains, but also on the degree to which those gains are maintained over time.

Data from long-term outcome studies have demonstrated that initial gains made in speech and language after cochlear implantation in early childhood continue to be maintained 10–15 years later and that a majority of these long-term CI users perform in the average range or better on several conventional measures of language outcomes (Davidson *et al.*, 2011; Geers and Hayes, 2011; Geers and Sedey, 2011; Spencer, 2004; Uziel *et al.*, 2007). In a recent study, users with 15–22 years of CI experience were found to have language skills similar to users with 7–14 years of CI experience (Ruffin *et al.*, under review). However, very long-term CI users (15–22 years of CI use) displayed lower performance on several measures of speech perception than other

Correspondence to: Irina Castellanos, DeVault Otologic Research Laboratory, Department of Otolaryngology Head and Neck Surgery, Indiana University School of Medicine, Riley Research 044, 699 Riley Hospital Drive, Indianapolis, IN 46202, USA. Email: icastell@indiana.edu

DOI 10.1179/1754762813Y.0000000043

long-term CI users (7–14 years). These outcome differences were explained by differences between the groups in etiology of hearing loss, pre-implant residual hearing, age of onset of deafness, and age of implantation. The very long-term CI users (15–22 years of CI use) had more meningitic etiology, poorer pre-implant pure-tone averages (PTAs), older age of onset of deafness, and older age of implantation (Ruffin *et al.*, under review).

Although studies of long-term speech-language outcomes following cochlear implantation in early childhood demonstrate positive outcomes for many users, considerable variability still remains in speech and language functioning in long-term CI users (Davidson *et al.*, 2011; Geers *et al.*, 2011; Ruffin *et al.*, under review). Understanding the source of this variability in long-term outcome and identifying predictors offers the potential for early identification and prevention of suboptimal speech and language outcomes. Long-term cochlear implantation outcome studies have shown, for example, that some conventional demographic and hearing history variables (e.g. earlier age at implantation, better pre-implant residual hearing, non-meningitic etiology of hearing loss, and shorter duration of deafness prior to cochlear implantation) predict positive long-term speech and language outcomes (Geers and Sedey, 2011; Ruffin *et al.*, under review; Uziel *et al.*, 2007). This information has been used to guide recommendations for treatment, as well as to identify children who might be at high risk for suboptimal speech-language outcomes (Copeland and Pillsbury, 2004).

Several recent longitudinal studies have also investigated the potential of early speech, language, and other neurocognitive skills to predict long-term speech-language outcomes following cochlear implantation. For example, Geers *et al.* (2011, 2008) examined children's speech perception and language skills following long-term CI use (an average of 13 years). Measures of speech perception and language were taken when the children entered grade school (aged 8–9 years) and then again during their high school years. The authors report that children made significant improvements during the grade school to high school period in terms of increases in speech perception, word recognition, and reading, and that performance during grade school was predictive of later long-term performance. Specifically, better language and reading scores during grade school predicted better scores during high school (Geers *et al.*, 2011). Additionally, measures of verbal short-term memory in children with CIs (as indexed by digit span) predicted long-term speech and language performance (Harris *et al.*, 2013; Pisoni *et al.*, 2011).

These studies demonstrate that behavioral measures of speech, language, and working memory development during the early grade school years are predictive of long-term speech-language outcomes into late childhood and adolescence in prelingually deaf, early implanted children. However, little is currently known about whether speech and language skills at even younger ages (e.g. preschool) can predict long-term outcomes. This knowledge is critically important for several reasons: first, it will enhance our understanding of the stability of speech and language development following cochlear implantation, and specifically whether speech-language functioning is stable even back to preschool years. Second, it will allow us to identify and treat children at-risk for poor outcomes during a period of rapid cognitive, linguistic, and social development, and third, this knowledge will inform the development of early, targeted interventions that are likely to impact long-term speech, language, and working memory outcomes. As the age of implantation continues to decrease, with many deaf children now receiving unilateral or bilateral implants prior to 24 months of age, the preschool years are likely to be a critical time for rapid development and early intervention in children with CIs.

In order to address these research questions, we investigated the relationship between speech intelligibility and receptive vocabulary skills in a group of 35 deaf children during preschool ages (after at least 1 year of CI use) and a broad set of speech, language, and verbal memory skills following long-term use (at least 7 years of CI use). Speech intelligibility and receptive vocabulary are a routine part of speech and language assessments of preschool children with CIs because of their functional importance for tracking language development, verbal intelligence, and quality of life (e.g. Moog and Geers, 1999). Unlike some more complex speech and language skills (e.g. reading, expression, language production, and comprehension of complex spoken language), these basic building blocks of speech and language can be reliably assessed at very young ages with simple behavioral tests and are fundamental for communication and language development (Robbins, 2003).

Children's speech intelligibility can be examined using assessments such as the Beginners Intelligibility Test (BIT; Osberger *et al.*, 1994), the McGarr Sentence Intelligibility Test (McGarr, 1983), and the Monsen sentences (Monsen, 1983). In these assessments, children repeat short and meaningful English sentences, and intelligibility is measured by having small groups of normal-hearing adults transcribe the children's speech. Studies using these assessment tools have shown that several hearing and device characteristics affect children's speech intelligibility. Deaf children acquire better speech intelligibility if they have better residual hearing prior to cochlear implantation (Svirsky *et al*., 2000), receive CIs earlier in development (Peng *et al*., 2004; Svirsky *et al*., 2007), use oral versus total communication strategies (Svirsky *et al*., 2000; Tobey *et al*., 2003, 2011), receive mainstream versus special education (Tobey *et al*., 2003), and use multipeak, as compared to spectral-peak, speech-coding strategies in their speech processor (Peng *et al*., 2004). Similarly, Tobey *et al*. (2003) report that additional device characteristics such as more active electrodes and wider dynamic ranges were predictive of better speech intelligibility in children who had 3–7 years of CI use. These findings underlie the important relationship between auditory signals delivered by a CI and phonological coding and the development of robust lexical representations of speech.

Compared to normal-hearing children, children with CIs have poorer speech intelligibility and show slower improvements in their intelligibility across development (Chin *et al*., 2003). The speech intelligibility of children with CIs continues to improve with development and increased device use (Chin *et al*., 2003; Osberger *et al*., 1994; Svirsky *et al*., 2007; Tobey *et al*., 2003, 2011). However, to date, no research has been carried out to assess whether measures of speech intelligibility obtained during early development (e.g. the preschool years) are predictive of long-term speech and language functioning.

Assessments such as the Reynell Developmental Language Scales (RDLS; Reynell and Gruber, 1990) and Peabody Picture Vocabulary Test (PPVT; Dunn and Dunn, 1997, 2007) have been extensively used to examine the vocabulary and language skills of very young children with CIs. Demographic, family environment, and hearing history factors have all been shown to contribute to the observed variability in children's language scores. Greater pre-implant residual hearing (El-Hakim *et al*., 2001; Geers *et al*., 2007), early age of cochlear implantation (Connor *et al*., 2006; Geers *et al*., 2007; Hayes *et al*., 2009), higher nonverbal intelligence (Geers *et al*., 2007), oral versus total communication strategies (El-Hakim *et al*., 2001), higher paternal education (Hayes *et al*., 2009), and family environments with higher levels of organization, but lower levels of control (Holt *et al*., 2012), have all been found to be associated with deaf children acquiring better language skills.

The receptive language skills of deaf children continue to improve following cochlear implantation and with development. However, compared to their normal-hearing peers, children with CIs are delayed in acquiring new words and have smaller vocabularies (Hayes *et al*., 2009). To our knowledge, only one study has investigated whether early measures of receptive language are predictive of long-term speech and language functioning. Hay-McCutcheon *et al*. (2008) examined whether language measures obtained from children under 7 years old, who received CIs, on average, at 4 years old, were predictive of long-term language development. Results indicated that measures of receptive (but not expressive) language obtained as early as 6 months following cochlear implantation and under 7 years of age were predictive of core language skills in adolescence (up to 17 years old). As children begin to receive CIs at earlier ages, it is important to understand how performance during these early periods of CI intervention affects long-term functioning. It is not yet known whether early measures of receptive language, obtained in early implanted children under 6 years old, are predictive of long-term speech and language functioning.

Taken together, Geers *et al*. (2008) and Hay-McCutcheon *et al*. (2008) were the first studies to report that early measures of children's speech and language are predictive of long-term performance. To extend these findings, we investigated whether measures obtained at even earlier ages of development would be predictive of long-term performance in a group of children who received CIs during a sensitive period for language development (implanted on average below 26 months; see Houston and Miyamoto, 2010). We asked whether preschool measures of speech intelligibility and receptive language (as assessed by the BIT and the Peabody Picture Vocabulary Test – 3, respectively) were predictive of speech, language, and verbal working memory functioning after at least 7 years of CI use ($M = 11.36$, range = 7.08–19.84). Continued research in this area is of critical importance at this time because the findings will allow for the early identification of children who may be at high risk for developing poor long-term outcomes, thereby allowing for interventions to occur at earlier stages in development in order to address not only speech-language deficits but also the cascading neurocognitive effects of speech-language delays (Pisoni *et al*., 2010).

Methods

Participants

Study participants were drawn from a sample of 58 CI users who volunteered to be contacted for research and who were evaluated as a part of a larger study of long-term outcome following cochlear implantation in childhood (see Ruffin *et al*., under review, for descriptive statistics on the demographics, hearing history, speech, and language variables of the full sample). Participants in the larger study were required to (1) have severe-to-profound prelingual hearing loss (>70 dB HL in the better hearing ear prior to age 3 years); (2) have received their CI prior to age 7 years; (3) have used their CI for 7 years or more; (4) use a currently available state-of-the-art multichannel

CI system; (5) live in a household with English as the primary language; and (6) have no additional developmental, neurological, or cognitive handicapping conditions other than hearing loss. In order to be include in this study, participants were also required to have completed either the PPVT-3 or the BIT between the ages of 3 and 6 years inclusive as a part of a prior, related study at our CI clinic. Of the 58 participants in the larger sample, 35 children had completed the PPVT-3 ($N = 12$), BIT ($N = 16$), or both PPVT-3 and BIT ($N = 7$) during the required 3–6-year age range, forming the sample used for analysis in this study.

Demographic variables coded for each participant included age, sex, family income (coded by income ranges on a 1 (under $5500) to 10 ($95 000 and over) scale, with values of 4, 6, and 8 corresponding to income values of $15 000–$24 999, $35 000–$49 999, and $65 000–$79 999, respectively), and race/ethnicity. Additional hearing history variables included age at onset of deafness (defined as the age at which deafness was identified or age at the time of a known event causing deafness), age at time of implantation, duration between preschool and long-term follow-up testing, duration of CI use at preschool testing, duration of CI use at long-term follow-up testing, pre-implant residual hearing (mean unaided PTA in the better-hearing ear for the frequencies 500, 1000, and 2000 Hz in dB HL), communication mode (coded 1 for total communication and 2 for oral communication), and etiology of deafness.

Characteristics of the study sample are summarized in Table 1. Etiology of deafness included unknown ($N = 22$), familial (at least one immediate family member also had deafness of unknown etiology) ($N = 5$), meningitis ($N = 3$), Mondini malformation ($N = 3$), and auditory neuropathy ($N = 2$). On average, children were implanted with a CI prior to 26-month old. All participants ($N = 35$) were fitted with unilateral CIs prior to preschool testing and 31% ($N = 11$) of participants were fitted with bilateral CIs by the time of the long-term follow-up testing. At the time of preschool testing, participants ranged in age from 3 to 6 years old and all had at least 1 year

Table 1 Participant demographics and hearing history

N	At preschool visit: BIT 23 M (SD) (range)	At preschool visit: PPVT-3 19 M (SD) (range)	At long-term follow-up visit: All 35 M (SD) (range)
Onset of deafness (months)	2.61 (*7.01*) (0.00–25.00)	0.00 (*0.00*) (0.00–0.00)	1.71 (*5.78*) (0.00–25.00)
Age at implantation (months)	25.77 (*10.16*) (9.92–43.47)	22.37 (*10.75*) (8.28–47.70)	25.47 (*10.77*) (8.28–47.70)
Age at testing (years)	3.92 (*0.86*) (3.00–5.59)	4.10 (*0.86*) (3.15–5.79)	13.48 (*3.77*) (7.80–23.36)
Duration of CI use (years)	1.78 (*0.76*) (1.00–3.99)	2.23 (*0.70*) (1.04–3.53)	11.36 (*3.40*) (7.08–19.84)
Pre-implant PTA*	112.44 (*6.20*) (100.00–118.43)	108.55 (*10.49*) (85.00–118.43)	110.67 (*8.93*) (85.00–118.43)
Income level**	NA	NA	7 (*2.24*) (2.00–10.00)
		Count (% of sample)	
Bilateral/unilateral CI			
Bilateral CI	0 (0.00)	0 (0.00)	11 (0.31)
Unilateral CI	23 (1.00)	19 (1.00)	24 (0.69)
Communication mode***			
Sign/total	7 (0.30)	6 (0.32)	5 (0.14)
Oral/cued	16 (0.70)	13 (0.68)	30 (0.86)
Gender			
Female	10 (0.43)	8 (0.42)	14 (0.40)
Male	13 (0.57)	11 (0.58)	21 (0.60)
Race			
Asian	1 (0.04)	1 (0.08)	1 (0.03)
Black	0 (0.00)	0 (0.00)	0 (0.00)
White	22 (0.96)	18 (0.92)	34 (0.97)
Ethnicity			
Hispanic	0 (0.00)	1 (0.05)	1 (0.03)
Not Hispanic	23 (1.0)	18 (0.95)	34 (0.97)

*Unaided pure-tone average in the better ear for the frequencies 500, 1000, and 2000 Hz in dB HL.
**Income level (not available for the preschool visit) is coded on a scale from under $5000 (coded 1) to $95 000 and over (coded 10) with a code of 7 = $50 000–$64 999 and a code of 8 = $65 000–$79 999.
***Communication mode is coded as sign/total communication (coded 1) or oral/cued (coded 2).
CI, cochlear implant.

of CI use. At the time of long-term follow-up testing, participants averaged 13.48 (SD = 3.77, range = 7.80–23.36) years old and averaged 11.36 (SD = 3.40, range = 7.08–19.84) years of CI use.

Procedure

All study procedures were reviewed and approved by the local institutional review board, and written informed consent was obtained for all participants or parents prior to initiation of any study procedures. Data were obtained from research visits to a large, university-hospital-based CI clinic conducted as a part of two related studies: (1) a long-running, longitudinal speech perception and production study that spanned preschool through adolescence; and (2) a cross-sectional long-term neurocognitive outcome study described earlier. The long-term outcome study provided the initial pool of 58 potential participants, for whom preschool visit data were sought from the longitudinal study database. If participants were tested more than once during preschool ages, the earliest testing session after age 3 years and 1 year of CI use was selected for analysis. Long-term follow-up visits took place an average of 9.58 (SD = 3.68, range = 4.56–18.83) years after the preschool visit. During the research visits, licensed speech-language pathologists administered tests of speech perception, speech production, language, and/or working memory in the child's preferred mode of communication (see Oral versus Total Communication Mode in Table 1). Speech perception test stimuli were presented from digital recordings over a loudspeaker at 65 dB SPL in a sound field within a sound-treated audio booth at 0° azimuth approximately 3 feet from the participant.

Measures

At the preschool visit, measures of *speech production* (BIT) and single word receptive *vocabulary* (Peabody Picture Vocabulary Test-3) were obtained to measure components of early speech and language development. At the long-term follow-up visit, measures of *speech perception* (Hearing in Noise Test for Children and Lexical Neighborhood Test), *language* (Peabody Picture Vocabulary Test-4 and Clinical Evaluation of Language Fundamentals), and *verbal working memory capacity* (Digit Span Forward and Digit Span Backward) were obtained to provide a broad characterization of long-term outcomes related to speech and language skills. Because of ability, attention, fatigue, or time constraints, not all children received every test at long-term follow-up assessment; see assessment descriptions below for group sample sizes.

Beginners Intelligibility Test (Osberger *et al.*, 1994). The BIT is a measure of speech intelligibility, which requires participants to repeat a list of 10 short and meaningful English sentences (sentences contain two to six words) spoken live-voice by an examiner. Audio recordings of the children's speech were made and played back to three normal hearing adult judges (naïve to deaf speech), who were instructed to transcribe each sentence. BIT scores expressed as percent of correct words transcribed, averaged across the three judges, were used as a measure of speech intelligibility during preschool ages. All children who completed the BIT during the preschool visit (N = 23) also completed all speech and language, and verbal short-term memory assessments during the long-term follow-up visit.

Peabody Picture Vocabulary Test – 3rd and 4th editions (PPVT-3, PPVT-4; Dunn and Dunn, 1997, 2007). The PPVT is a one-word receptive vocabulary test, which requires participants to choose one of four pictures matching a spoken word. For children using total communication, Signed Exact English accompanied the spoken word. PPVT standard scores were used as a measure of word knowledge at both preschool and long-term follow-up. The PPVT-3 was given to participants during their preschool visit and the PPVT-4 was given during the long-term follow-up visit. Dunn and Dunn (2007) report high correlations between scores obtained using the 3rd and 4th editions of the PPVT. All children who completed the PPVT-3 during the preschool visit (N = 19) also completed the PPVT-4 during the long-term follow-up visit.

Lexical Neighborhood Test (LNT; Kirk *et al.*, 1995). The LNT is an open-set test of speech perception, which requires participants to repeat monosyllabic words. The LNT contains 50 lexically easy (LNT-E) and lexically hard (LNT-H) words. The percent correct responses to LNT-E and LNT-H words were used as measures of speech perception skills at long-term follow-up. All children who completed the PPVT-3 during the preschool visit (N = 19) also completed the LNT during the long-term follow-up visit.

Hearing in Noise Test for Children (HINT-C; Nilsson *et al.*, 1996). The HINT-C is an open-set sentence recognition test, which requires participants to repeat 10 sentences in quiet and 10 sentences in +5-dB background noise. The percent of correctly repeated words were used to assess speech perception skills at long-term follow-up. Eighteen children completed the PPVT-3 during the preschool visit and the HINT-C during the long-term follow-up visit.

Clinical Evaluation of Language Fundamentals Fourth Edition (CELF-4; Semel *et al.*, 2003). The CELF-4 is a measure of simple and complex receptive and expressive language skills, which assesses participants on understanding concepts and following directions, recalling sentences, formulating sentences, and vocabulary knowledge. For children using total communication, Signed Exact English accompanied spoken words. CELF-4 Core Language standard scores were used to measure global language functioning at long-term follow-up. Seventeen children completed the PPVT-3 during the preschool visit and the CELF during the long-term follow-up visit.

Digit Span. Verbal working memory capacity was assessed using the Digit Span subtest of the Wechsler Intelligence Scale for Children, Third Edition (WISC-III; Wechsler, 1991) and the Visual Digit Span subtest of the Wechsler Intelligence Scale for Children, Fourth Edition – Integrated (WISC-IV-I; Wechsler *et al*., 2004). The Digit Span subtest requires participants to reproduce a sequence of spoken digits presented in forward (Digit Span Forward) or backward (Digit Span Backward) order, whereas the Visual Digit Span subtest requires repetition of a series of visually presented digits in forward order. Hence, Digit Span Forward and Visual Digit Span differ in presentation modality but involve the same rote short-term verbal memory task. Digit Span Backward, on the other hand, includes an additional concurrent component of cognitive processing (digit reversal) during memory; therefore, relative to Digit Span Forward, Digit Span Backward requires additional processing effort in the form of controlled attention (executive control) in addition to rote short-term memory encoding and retrieval. Scaled scores for Digit Span Forward, Digit Span Backward, and Visual Digit Span were used as measures of verbal working memory capacity at long-term follow-up. All children who completed the PPVT-3 during the preschool visit ($N = 19$) also completed Visual Digit Span during the long-term follow-up visit. Eighteen children who completed the PPVT-3 during the preschool visit also completed Digit Span Forward and Digit Span Backward during the long-term follow-up visit.

Data analysis

First, in order to examine the relationship between early speech-language skills and long-term outcomes, scores from the BIT and PPVT-3 completed at preschool were correlated with long-term follow-up scores on the speech perception (LNT, HINT-C), language (PPVT-4, CELF-4), and verbal working memory capacity (Digit Span Forward, Digit Span Backward, Visual Digit Span) measures. Next, in order to evaluate the independent contribution of preschool speech-language scores to long-term outcome while also accounting for demographic and hearing history variables, hierarchical blockwise regression analyses were conducted with each long-term follow-up speech-language score as the criterion variable and blocks of variables entered sequentially as follows: Block 1 (Preschool Speech-Language Functioning) consisted of either preschool BIT or preschool PPVT-3 as a measure of baseline early speech-language skills. Since preschool speech-language skills were the focus of this research, the variable from this block was retained in the regression analysis regardless of its statistical significance. Variables for Blocks 2 and 3 were entered using a stepwise method in which only variables significant at the $P < 0.05$ level were entered into the equation; this method was necessary due to the large number of variables in these blocks. Block 2 (Demographic and Hearing History) consisted of demographic and hearing history variables: gender, age of onset of deafness, age at implantation, best unaided PTA pre-implantation, age at long-term follow-up testing, duration of CI use at long-term follow-up testing, and duration of time between the preschool and long-term follow-up visits. Communication mode was not included in these analyses because only 12 participants used a total communication mode at preschool visit, and only 5 participants used a total communication mode at long-term follow-up visit.

Finally, Block 3 (Moderator Effects) consisted of the interaction (product term) of the variables in Block 1 (Preschool BIT or PPVT-3) and Block 2 (any demographic or hearing history variables retained in the stepwise analyses). These regression equations test the extent to which conventional demographic or hearing history variables might add to or attenuate the effects of early (Preschool) speech-language skills (BIT or PPVT-3) on later (Long-Term Follow-Up) speech-language abilities. If the regression coefficient of preschool BIT or PPVT-3 is attenuated (substantially reduced) by the addition of demographic and hearing history variables in later blocks, the more basic demographic and hearing history variables may be considered to account for the relation observed between preschool and long-term follow-up speech-language skills (for example, in a mediating relationship; see Holmbeck, 1997). Furthermore, a statistically significant product term in Block 3 would demonstrate that the effect of preschool BIT or PPVT-3 on long-term follow-up speech-language outcome is moderated by the demographic or hearing history variable (Holmbeck, 1997).

Results

Correlational analyses

Correlations of preschool PPVT-3 and BIT scores with scores on the long-term follow-up speech-language measures are summarized in Table 2. PPVT-3 scores obtained during the preschool visit were significantly correlated with long-term performance on PPVT-4 ($r = 0.55$) and HINT-C in Noise scores ($r = 0.49$). Non-significant trends were found for correlations of preschool PPVT-3 with long-term follow-up CELF-Core, HINT-C in Quiet, LNT-H, and Digit Span

Table 2 Correlations between preschool speech intelligibility and receptive vocabulary and long-term speech and language outcomes

	Preschool measures	
	PPVT-3	BIT
Long-term follow-up outcomes		
Speech perception:		
HINT-C in Quiet	0.45†	0.30
HINT-C in Noise	0.49*	0.53*
LNT-E	0.32	0.30
LNT-H	0.39†	0.29
Language:		
PPVT-4	0.55*	0.58**
CELF-Core	0.48†	0.62**
Verbal working memory capacity:		
Digit span forward	0.42†	0.46*
Digit span backward	−0.14	0.17
Visual digit span	0.15	0.47*

PPVT, Peabody Picture Vocabulary Test given during the Preschool and the Long-term Follow-up visits; Preschool BIT, Beginners Intelligibility Test given during the Preschool visit; CELF Core, Clinical Evaluation of Language Fundamentals Fourth Edition Core Language Score; HINT-C, Hearing in Noise Test for Children (In Noise = +5dB); LNT, Lexical Neighborhood Test – Easy Words (E), Hard Words (H).
†$P < 0.10$; *$P < 0.05$; **$P < 0.01$.

Forward. Preschool BIT scores were significantly correlated with long-term follow-up performance on the PPVT-4 ($r = 0.58$), CELF-Core ($r = 0.62$), HINT-C in Noise ($r = 0.53$), Digit Span Forward ($r = 0.46$), and Visual Digit Span ($r = 0.47$) tests. Additionally, preschool PPVT-3 and preschool BIT scores were highly correlated ($r = 0.85$, $P < 0.05$) with each other for the seven participants who received both assessments during the preschool visit.

Regression models predicting long-term outcomes

Table 3 displays a summary of the results of the regression analyses using preschool BIT as a predictor of long-term follow-up speech and language scores. None of the preschool or long-term follow-up demographic or hearing history variables significantly added to or attenuated the relation obtained between preschool BIT scores long-term follow-up language outcomes: Preschool BIT accounted for 34–39% of the variance in long-term follow-up PPVT-4 and CELF-Core scores. Of the long-term follow-up speech perception scores, preschool BIT significantly predicted only HINT-C in Noise, and this relation remained significant (although somewhat attenuated) following the entry of age of onset of deafness into the regression equation predicting HINT-C in Noise (older age of onset of deafness was found to be significantly related to poorer HINT-C in Noise scores). The overall equation with preschool BIT and onset age of deafness as predictors accounted for 59% of the variance in HINT-C in Noise scores at long-term follow-up. Higher preschool BIT and earlier age of implantation significantly predicted better performance on the other long-term follow-up speech perception scores (HINT-C in Quiet, LNT-Easy, and LNT-Hard), accounting for 34–49% of the variance in those scores. None of the product terms (Block 3) were significant for these equations, indicating that the relations between preschool BIT and long-term follow-up speech perception scores were not moderated by age of onset of deafness or age of implantation. For verbal working memory scores at long-term follow-up, none of the demographic or hearing history variables added to the predictive value of preschool BIT in the regression equations (see Table 2 for correlations of BIT with verbal working memory scores; bivariate regression results for preschool BIT predicting each verbal working memory variable are not reported because they are equivalent to correlational results).

Table 3 Regressions predicting long-term speech and language outcomes

	Long-term follow-up speech-language					
	PPVT-4 N = 23	CELF-Core N = 23	HINT-C in Quiet N = 23	HINT-C in Noise N = 23	LNT-Easy N = 23	LNT-Hard N = 23
Block 1:						
Preschool BIT	0.58**	0.62**	0.30	0.53*	0.30	0.29
R^2	0.34**	0.39**	0.09	0.28*	0.09	0.09
Block 2:						
Preschool BIT			0.34	0.38*	0.35*	0.33
Onset of Deafness				−0.57**		
Age at Implantation			−0.50*		−0.64**	−0.51*
R^2			0.34*	0.59**	0.49**	0.34*

Note: Because we analyzed the data using a stepwise regression, only demographic and hearing history variables significant at the $P < 0.05$ level were entered into the equation and displayed in this table in Block 2. Values are standardized regression coefficients. Preschool BIT, Beginners Intelligibility Test given during the Preschool visit; PPVT-4, Peabody Picture Vocabulary Test Fourth Edition; CELF Core, Clinical Evaluation of Language Fundamentals Fourth Edition Core Language Score; HINT-C, Hearing in Noise Test for Children (In Noise = +5dB); LNT, Lexical Neighborhood Test – Easy Words (E), Hard Words (H).
*$P < 0.05$; **$P < 0.01$.

In contrast to preschool BIT results, none of the demographic or hearing history variables added to the predictive value of preschool PPVT-3 scores for any of the long-term follow-up speech, language, or working memory variables (see Table 2 for correlations of preschool PPVT-3 with speech and language, and verbal working memory scores, which are equivalent to bivariate regression results).

Discussion

This study demonstrated that early measures of preschool receptive vocabulary and speech intelligibility skills predict later speech and language outcomes in long-term users of CIs. Receptive vocabulary (PPVT-3) during preschool was found to be highly predictive of long-term receptive vocabulary (PPVT-4) and speech perception (HINT-C in Noise), on average, 9 years later. Traditional demographic and hearing history variables did not add to this predictive relationship in hierarchical regression analyses. Speech intelligibility (BIT) during preschool was even more strongly related to long-term speech and language outcomes, significantly predicting language (PPVT-4 and CELF-Core) and forward short-term verbal memory capacity (Digit Span Forward and Visual Digit Span). In the regression analysis including age at implantation and onset of deafness (the only two demographic/hearing history variables adding significantly to the predictiveness of preschool speech intelligibility for later speech and language outcomes), speech intelligibility (BIT) during preschool also significantly predicted long-term speech perception (HINT-C and LNT); none of the product terms (between preschool BIT and the demographic/hearing history variables in Block 2) significantly predicted long-term speech perception. Thus, preschool PPVT and preschool BIT scores predicted later speech-language functioning, and these predictive values were not attenuated or moderated by including the conventional demographic and hearing history variables that add to the relations with long-term speech-language outcomes.

Prior research has shown strong relations between early speech and language skills and the development of long-term speech and language skills in samples of children with CIs. Hay-McCutcheon *et al.* (2008) reported that a measure of early receptive language using the RDLS, obtained before 7 years of age in a group of children who, on average, received their CIs at 4 years of age, was predictive of complex language functioning in adolescence. Davidson *et al.* (2011), Geers *et al.* (2008), and Tobey *et al.* (2011) also reported that measures of speech intelligibility, speech perception, and language obtained during grade school were predictive of speech and language performance approximately 8–10 years later. They examined several factors associated with the variance of individual scores and showed that overall, speech intelligibility, speech perception, and language skills of children with CIs continue to improve across the grade school to high school years.

This study extends these earlier research findings by demonstrating relationships between speech and language skills at even earlier (preschool) ages and long-term speech and language outcomes in users of CIs for an average of over 11 years (range = 7.08–19.84 years). Our findings demonstrate that speech intelligibility and receptive vocabulary measures obtained from a sample of children implanted within an early age range (8.28–47.70 months, $M = 25.47$) are predictive of a comprehensive set of speech, language, and verbal working memory long-term outcome measures. We also accounted for several factors that typically contribute a large amount of predicted variance in speech and language outcomes by selecting a homogenous sample of children with respect to the onset of deafness (below 2 years old on average, range = 0.00–25.00 months), age at cochlear implantation (below 26 months old on average, range = 8.28–47.70 months), chronological age at the time of preschool testing (3–6 years old), and duration of CI use at time of preschool testing (at least 1 year, range = 1.00–3.99 years). This is the first study to document that measures of very early speech-language functioning during the preschool years (3–6 years old) can be used to identify children with CIs who are at risk for poor outcomes or who are likely to experience positive speech-language outcomes after many years of long-term CI use.

In our regression analysis, relatively few of the demographic and hearing history variables added significantly to the variance contributed by preschool PPVT-3 and preschool BIT in predicting long-term speech and language outcomes. Furthermore, only age of onset of deafness (for preschool BIT and long-term follow-up HINT-C in Noise) and age at implantation (for preschool BIT and long-term follow-up HINT-C in Quiet and LNT) added to the relationship between preschool and long-term follow-up speech and language functioning. This finding suggests that the predictive value of early measures of speech-language functioning for later speech-language functioning is minimally augmented by the addition of conventional demographic and hearing history variables. However, in line with previous findings, a small set of hearing history variables were predictive of long-term outcomes (Nikolopoulos and Vlastarakos, 2010; Niparko *et al.*, 2010; Peterson *et al.*, 2010; Wheeler *et al.*, 2009) even after early speech-language performance was taken into account in regression equations. Not surprisingly, children who were implanted earlier in childhood or who had later onset of deafness had better word and sentence recognition skills after an average of 11 years of CI use (range = 7.08–19.84).

The finding that better speech intelligibility (BIT) and receptive vocabulary (PPVT-3) skills during the preschool ages is also predictive of verbal short-term memory performance (Digit Span Forward and Visual Digit Span) at long-term follow-up is a novel finding that is consistent with prior research demonstrating close links between speech production and spoken word recognition and the rehearsal of phonological and lexical representations in immediate verbal memory (Pisoni *et al.*, 2011). Several studies have found similar links between core measures of neurocognitive functioning (using digit span as an index for working memory capacity) and speech and language performance in children with CIs (Harris *et al.*, 2013; Pisoni *et al.*, 2011; Pisoni and Geers, 2000), but this is the first study to demonstrate a predictive relationship between preschool speech and language measures and later verbal short-term memory performance.

Speech intelligibility assessments such as the BIT are extremely useful because they not only provide information about children's speech production skills, but they also provide some insights into children's verbal working memory and control processes. In carrying out the BIT, children are read sentences and are instructed to repeat sentences to the examiner, which requires encoding, storage, and retrieval of phonological, lexical and syntactic information. Working memory is central to complex language processing skills and numerous past studies have shown that children with delays and deficits in language also have disturbances in working memory (Adams and Gathercole, 2000; Riches, 2012). Because both speech production and language comprehension draw on the same core foundational information processing operations (encoding, storage, and retrieval of phonological and lexical representations in working memory), it is not entirely surprising that better speech intelligibility skills were predictive of better long-term speech and language functioning in this sample. Speech intelligibility measures like the BIT might, in fact, be a proxy for the speed and efficiency of verbal working memory and may provide a global measure of how the entire system is functioning together in an integrated fashion. Relations have been reported between speech intelligibility and short-term memory dynamics: Children with CIs who produce more intelligible speech have longer memory spans for forward digits (Pisoni and Geers, 2000; Pisoni *et al.*, 1999), and children who speak faster display longer memory spans for forward digits (Burkholder and Pisoni, 2003; Pisoni and Cleary, 2003), and better word recognition (Pisoni and Cleary, 2003). These findings suggest close links between early auditory experience and activity-dependent learning after cochlear implantation and the long-term development of verbal working memory skills.

Our findings that preschool speech intelligibility and receptive vocabulary are predictive of long-term speech and language and verbal short-term memory performance also demonstrate that preschool assessments of speech intelligibility and vocabulary skills are clinically relevant beyond just short-term functioning. A viable starting point for identifying at-risk CI users and initiating interventions to maximize long-term adjustment should begin as early as possible during the preschool years. Recently, we also showed that significant developmental delays in executive functioning also begin to emerge during the preschool years in children with CIs (Beer *et al.*, under review), suggesting that long-term adjustment risks may be identified at younger ages than previously recognized. Auditory-oral interventions that selectively focus and target improving the speech intelligibility of children with CIs and that emphasize articulation and phonological awareness that transfer from clinical to real-world settings (see Ertmer and Ertmer, 1998, for specific intervention techniques) may have long-term as well as short-term benefits for poorly performing children. Additionally, children's speech intelligibility may be assessed periodically beginning intensively in preschool as an approach to individualize educational programs (Ertmer, 2011).

Several limitations must be taken into account in interpreting the results of this study. First, the sample size was relatively small ($N = 23$ for the preschool BIT and $N = 19$ for the preschool PPVT-3), placing limits on statistical power, particularly for the regression analyses. Small sample size also affected the ability to test relations between variables, particularly the effects of communication mode, which was highly skewed toward the auditory–oral communication modality. Second, because of behavioral and time constraints, both preschool assessments were not given to every child (i.e. only 7 of 35 children received both the preschool BIT and preschool PPVT-3), thus limiting our ability to fully correlate performance across both preschool measures. Finally, data were obtained from only two (preschool and long-term follow-up) time points. Longitudinal data with more time points may provide information about more subtle changes in developmental trajectory over time.

In summary, this study investigated the ability of preschool speech-language measures to predict long-term speech-language functioning in users of CIs. Measures of receptive vocabulary (PPVT-3) and speech intelligibility (BIT) obtained during the preschool years were found to be predictive of long-term speech, language, and verbal working memory performance after 7–19 years of CI use in early-implanted deaf children with CIs. These findings suggest the clinical utility of very early measurements of speech and language skills in identifying children who may be at high risk for poor long-term speech and language outcomes following cochlear implantation.

Acknowledgment

This research was supported by grants from the National Institutes of Health-National Institute on Deafness and Other Communication Disorders: R01 DC009581, R01 DC000111, and T32 DC00012.

References

Adams A.M., Gathercole S.E. 2000. Limitations in working memory: implications for language development. *International Journal of Language & Communication Disorders*, 35: 95–116. doi:10.1080/136828200247278

Beer J., Kronenberger W.G., Castellanos I., Colson B.G., Henning S.C., Pisoni D.B. (under review). Executive functioning skills in preschool-age children with cochlear implants.

Burkholder R.A., Pisoni D.B. 2003. Speech timing and working memory in profoundly deaf children after cochlear implantation. *Journal of Experimental Child Psychology*, 85: 63–88. doi:10.1016/S0022-0965(03)00033-X

Chin S.B., Tsai P.L., Gao S. 2003. Connected speech intelligibility of children with cochlear implants and children with normal hearing. *American Journal of Speech-Language Pathology*, 12: 440–451.

Connor C.M., Craig H.K., Raudenbush S.W., Heavner K., Zwolan T.A. 2006. The age at which young deaf children receive cochlear implants and their vocabulary and speech-production growth: is there an added value for early implantation? *Ear & Hearing*, 27: 628–644.

Copeland B.J., Pillsbury H.C. 2004. Cochlear implantation for the treatment of deafness. *Annual Review of Medicine*, 55: 157–167. doi:10.1146/annurev.med.55.091902.105251

Davidson L.S., Geers A.E., Blamey P.J., Tobey E., Brenner C. 2011. Factors contributing to speech perception scores in long-term pediatric CI users. *Ear & Hearing*, 32: 19S–26S.

Dunn L.M., Dunn D.M. 2007. *Peabody picture vocabulary test, fourth edition manual.* Bloomington, MN: Pearson.

Dunn L.M., Dunn L.M. 1997. *Peabody picture vocabulary test, third edition manual.* Circle Pines, MN: American Guidance Service.

El-Hakim H., Levasseur J., Papsin B.C., Panesar J., Mount R.J., Stevens D., *et al.* 2001. Assessment of vocabulary development in children after cochlear implantation. *Archives of Otolaryngology—Head & Neck Surgery*, 127: 1053–1059.

Ertmer D.J. 2011. Assessing speech intelligibility in children with hearing loss: toward revitalizing a valuable clinical tool. *Language, Speech, and Hearing Services in Schools*, 42: 52–58.

Ertmer D.J., Ertmer P.A. 1998. Constructivist strategies in phonological intervention: facilitating self-regulation for carryover. *Language, Speech, and Hearing Services in Schools*, 29: 67–75.

Geers A., Tobey E., Moog J., Brenner C. 2008. Long-term outcomes of cochlear implantation in the preschool years: from elementary grades to high school. *International Journal of Audiology*, 47: S21–S30. doi:10.1080/14992020802339167

Geers A.E., Hayes H. 2011. Reading, writing, and phonological processing skills of adolescents with 10 or more years of cochlear implant experience. *Ear & Hearing*, 32: 49S–59S. doi:10.1097/AUD.0b013e3181fa41fa

Geers A.E., Sedey A.L. 2011. Language and verbal reasoning skills in adolescents with 10 or more years of cochlear implant experience. *Ear & Hearing*, 32: 39S–48S.

Geers A.E., Nicholas J.G., Moog J.S. 2007. Estimating the influence of cochlear implantation on language development in children. *Audiological Medicine*, 5: 262–273. doi:10.1080/16513860701659404

Geers A.E., Strube M.J., Tobey E.A., Moog J.S. 2011. Epilogue: factors contributing to long-term outcomes of cochlear implantation in early childhood. *Ear & Hearing*, 32: 84S–92S. doi:10.1097/AUD.0b013e3181ffd5b5

Harris M.S., Kronenberger W.G., Gao S., Hoen H.M., Miyamoto R.T., Pisoni D.B. 2013. Verbal short-term memory development and spoken language outcomes in deaf children with cochlear implants. *Ear & Hearing*, 34: 179–192. doi:10.1097/AUD.0b013e318269ce50

Hay-McCutcheon M.J., Kirk K.I., Henning S.C., Gao S., Qi R. 2008. Using early language outcomes to predict later language ability in children with cochlear implants. *Audiology and Neurotology*, 13: 370–378. doi:10.1159/000148200

Hayes H., Geers A.E., Treiman R., Moog J.S. 2009. Receptive vocabulary development in deaf children with cochlear implants: achievement in an intensive auditory-oral educational setting. *Ear & Hearing*, 30: 128–135. doi:10.1097/AUD.0b013e3181926524

Holmbeck G.N. 1997. Toward terminological, conceptual, and statistical clarity in the study of mediators and moderators: examples from the child-clinical and pediatric psychology literatures. *Journal of Consulting and Clinical Psychology*, 65: 599–610.

Holt R.F., Beer J., Kronenberger W.G., Pisoni D.B. 2013. Developmental effects of family environment on outcomes in pediatric cochlear implant recipients. *Otology & Neurotology*, 34, 388–395. doi:10.1097/MAO.0b013e318277a0af

Houston D.M., Miyamoto R.T. 2010. Effects of early auditory experience on word learning and speech perception in deaf children with cochlear implants: implications for sensitive periods of language development. *Otology & Neurotology*, 31: 1248–1253.

Kirk K.I., Pisoni D.B., Osberger M.J. 1995. Lexical effects on spoken word recognition by pediatric cochlear implant users. *Ear & Hearing*, 16: 470–481.

McGarr N.S. 1983. The intelligibility of deaf speech to experienced and inexperienced listeners. *Journal of Speech and Hearing Research*, 26: 451–458.

Monsen R.B. 1983. The oral speech intelligibility of hearing-impaired talkers. *Journal of Speech and Hearing Disorders*, 48: 286–296.

Moog J.S., Geers A.E. 1999. Speech and language acquisition in young children after cochlear implantation. *Otolaryngologic Clinics of North America*, 32: 1127–1141. doi:10.1016/S0030-6665(05)70199-7

Nikolopoulos T.P., Vlastarakos P.V. 2010. Treating options for deaf children. *Early Human Development*, 86: 669–674. doi:10.1016/j.earlhumdev.2010.10.001

Nilsson M., Soli S.D., Gelnett D. 1996. *Development and norming of a hearing in noise test for children*, Vol. 5. House Ear Institute Internal Report. pp. 115–128.

Niparko J.K., Tobey E.A., Thal D.J., Eisenberg L.S., Wang N.Y., Quittner A.L., *et al.* 2010. Spoken language development in children following cochlear implantation. *JAMA: The Journal of the American Medical Association*, 303: 1498–1506. doi:10.1001/jama.2010.451

Osberger M.J., Robbins A.M., Todd S.L., Riley A.I. 1994. Speech intelligibility of children with cochlear implants. *The Volta Review*, 96: 169–180.

Peng S.C., Spencer L.J., Tomblin J.B. 2004. Speech intelligibility of pediatric cochlear implant recipients with 7 years of device experience. *Journal of Speech, Language, and Hearing Research*, 47: 1227–1236.

Peterson N.R., Pisoni D.B., Miyamoto R.T. 2010. Cochlear implants and spoken language processing abilities: review and assessment of the literature. *Restorative Neurology and Neuroscience*, 28: 237–250. doi:10.3233/RNN-2010-0535

Pisoni D.B., Cleary M. 2003. Measures of working memory span and verbal rehearsal speed in deaf children after cochlear implantation. *Ear & Hearing*, 24: 106S–120S. doi:10.1097/01.AUD.0000051692.05140.8E

Pisoni D.B., Cleary M., Geers A.E., Tobey E.A. 1999. Individual differences in effectiveness of cochlear implants in children who are prelingually deaf: new process measures of performance. *The Volta Review*, 101: 111–164.

Pisoni D.B., Conway C.M., Kronenberger W., Henning S., Anaya E. 2010. Executive function, cognitive control and sequence learning in deaf children with cochlear implants. In: Marschark

M.S., Spencer P.E. (eds.) *Oxford handbook of deaf studies, language, and education.* 2nd ed., Vol. 1. New York: Oxford University Press. pp. 439–457.

Pisoni D.B., Geers A.E. 2000. Working memory in deaf children with cochlear implants: Correlations between digit span and measures of spoken language processing. *The Annals of Otology, Rhinology & Laryngology Supplement*, 185: 92–93.

Pisoni D.B., Kronenberger W., Roman A., Geers A. 2011. Measures of digit span and verbal rehearsal speed in deaf children following more than 10 years of cochlear implantation. *Ear & Hearing*, 32: 60S–74S. doi:10.1097/AUD.0b013e3181ffd58e

Reynell J., Gruber C.P. 1990. *Reynell developmental language scales: manual.* Los Angeles, CA: Western Psychological Services.

Riches N.G. 2012. Sentence repetition in children with specific language impairment: an investigation of underlying mechanisms. *International Journal of Language & Communication Disorders*, 47: 499–510. doi:10.1111/j.1460-6984.2012.00158.x

Robbins A.M. 2003. Communication intervention for infants and toddlers with cochlear implants. *Topics in Language Disorders*, 23: 16–33.

Ruffin C.V., Kronenberger W.G., Colson B.G., Henning S.C., Pisoni D.B. (in press). Long-term speech and language outcomes in prelingually deaf children, adolescents and young adults who received cochlear implants in childhood. *Audiology & Neurotology*.

Semel E.M., Wiig E.H., Secord W. 2003. *Clinical evaluation of language fundamentals (CELF-4)*. San Antonio, TX: The Psychological Corporation.

Spencer P.E. 2004. Individual differences in language performance after cochlear implantation at one to three years of age: Child, family, and linguistic factors. *Journal of Deaf Studies and Deaf Education*, 9: 395–412. doi:10.1093/deafed/enh033

Svirsky M.A., Chin S.B., Jester A. 2007. The effects of age at implantation on speech intelligibility in pediatric cochlear implant users: clinical outcomes and sensitive periods. *Audiological Medicine*, 5: 293–306. doi:10.1080/16513860701727847

Svirsky M.A., Chin S.B., Miyamoto R.T., Sloan R.B., Caldwell M.D. 2000. Speech intelligibility of profoundly deaf pediatric hearing aid users. *The Volta Review*, 102: 175–198.

Tobey E.A., Geers A.E., Brenner C., Altuna D., Gabbert G. 2003. Factors associated with development of speech production skills in children implanted by age five. *Ear & Hearing*, 24: 36S–45S. doi:10.1097/01.AUD.0000051688.48224.A6

Tobey E.A., Geers A.E., Sundarrajan M., Lane J. 2011. Factors influencing elementary and high-school aged cochlear implant users. *Ear & Hearing*, 32: 27S–38S.

Uziel A.S., Sillon M., Vieu A., Artieres F., Piron J.P., Daures J.-P., *et al.* 2007. Ten-year follow-up of a consecutive series of children with multichannel cochlear implants. *Otology & Neurotology*, 28: 615–628. doi:10.1097/01.mao.0000281802.59444.02

Wechsler D. 1991. *Wechsler intelligence scale for children.* 3rd ed. San Antonio, TX: The Psychological Corporation.

Wechsler D., Kaplan E., Fein D., Kramer J., Morris R., Delis D., *et al.* 2004. *Wechsler intelligence scale for children – fourth edition – integrated.* San Antonio, TX: Harcourt Assessment.

Wheeler A., Archbold S.M., Hardie T., Watson L.M. 2009. Children with cochlear implants: the communication journey. *Cochlear Implants International*, 10: 41–62.