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Attention to speech and spoken language development in deaf children with cochlear implants: A ten-year longitudinal study

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Abstract

Early auditory/language experience plays an important role in language development. In this study, we examined the effects of severe-to-profound hearing loss and subsequent cochlear implantation on the development of attention to speech in children with cochlear implants (CIs). In addition, we investigated the extent to which attention to speech may predict spoken language development in children with CIs. We tested children with CIs and compared them to chronologically age-matched peers with normal hearing (NH) on their attention to speech at four time points post implantation; specifically, less than 1 month, 3 to 6 months, 12 months, and 18 months post implantation. We also collected a variety of well-established speech perception and spoken language measures from the children with CIs in a 10-year longitudinal study. Children with CIs showed reduced attention to speech as compared to their peers with NH at less than 1 month post implantation, but a similar degree of attention to speech as their NH peers during later time points. In addition, attention to speech at 3 to 6 months post implantation predicts speech perception in children with CIs. These results inform language acquisition theories and bring insights into our understanding of early severe-to-profound hearing loss on infants' attention to speech skills. In addition, the findings have significant clinical implications for early intervention on hearing loss, which emphasizes the importance of developing strong listening skills.

Keywords

attention to speech; language development; children with cochlear implants

Cochlear implants (CIs) provide children who have severe-to-profound sensorineural hearing loss access to sound, which has permitted deaf children to attain unprecedented levels of spoken language abilities (Kirk, 2000; Tomblin, Spencer, Flock, Tyler, & Gantz, 1999). However, challenges remain because CIs deliver only degraded and impoverished representations of the acoustic signal to their users (Geers, Tobey, Moog, & Brenner, 2008; Houston, Beer, et al., 2012; Houston & Bergeson, 2014). While some children with CIs develop age-appropriate speech and spoken language skills and appear to be well on their

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way to acquire spoken language through their implants, many others who receive CIs, even at very early ages, often lag behind their peers with normal hearing (NH) and never reach the critical milestones in speech and language development (Conway, Pisoni, Anaya, Karpicke, & Henning, 2011; Geers, Strube, Tobey, Pisoni, & Moog, 2011; Holt, Beer, Kronenberger, Pisoni, & Lalonde, 2012; Houston & Bergeson, 2014). Several demographic variables have been found to be related to speech and language outcomes after cochlear implantation, including age at implantation (Connor, Craig, Raudenbush, Heavner, & Zwolan, 2006; Holt & Svirsky, 2008; Kirk, Miyamoto, Ying, Perdew, & Zuganelis, 2002), amount of residual hearing before implantation (Niparko et al., 2010), communication mode (Nittrouer, 2010), and duration of CI use (Fryauf-Bertschy, Tyler, Kelsay, & Gantz, 1997). However, these factors do not explain all the variance in outcome measures in children with CIs (Geers et al., 2011). More important, they do not address the fundamental linguistic and/or cognitive processes that allow for successful spoken language acquisition. Given the pervasiveness of language delay and variability of spoken language outcomes in children with CIs, studies examining early intrinsic precursors to language development in children with CIs are critical for both theoretical and clinical purposes.

Although it is common to consider deafness as affecting hearing alone, there is a growing body of evidence suggesting that early auditory/language deprivation due to hearing loss[2] has an impact on many cognitive skills, including memory, attention, learning, and information processing, that are essential for speech and spoken language development (Bharadwaj & Mehta, 2016; Conway et al., 2011; Horn, Davis, Pisoni, & Miyamoto, 2005; Pisoni & Geers, 2000; Smith, Quittner, Osberger, & Miyamoto, 1998). For example, using an auditory digit span test, Pisoni and Geers (2000) compared the working memory of 8- and 9-year-old prelingually deaf children who had used CIs for a period of at least 4 years to age-matched peers with NH. They found that children with CIs had poorer working memory capacity, which was related to their speech perception, speech production, language comprehension, and reading abilities (Moossavi, Etemadi, Javanbakht, Bakhshi, & Sharafi, 2016; Pisoni & Cleary, 2003; Pisoni & Geers, 2000; Willstedt-Svensson, Löfqvist, Almqvist, & Sahlén, 2004). Smith et al. (1998) also reported a poorer visual selection attention in children with CIs. They suggested that the deficit was due to poor multimodal sensory integration. In a recent study, Hall et al. (2017) compared executive function between Deaf native signers and their age-matched children with NH and found that Deaf native signers achieved similar scores as their peers with NH. This finding raised the possibility that early language exposure serves as a protective role in the development of executive function in Deaf children. Taken together, this body of research suggests that early auditory/language exposure is crucial for the development of general cognitive skills that would contribute to speech and spoken language development.

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[2] There are two major hypotheses regarding the underlying cause of differences in cognitive skills between deaf and hearing children: the auditory scaffolding hypothesis (Conway, Pisoni, & Kronenberger, 2009) and the language deprivation hypothesis (Hall, Eigsti, Bortfeld, & Lillo-Martin, 2017). The focus of the current study is not to tease apart the two hypotheses, as our participants with CIs had very limited or no sign language input, thus lacking both auditory and language input. Throughout the paper, we do not make assumptions about whether it is auditory deprivation or language deprivation that leads to those differences. We consider both hypotheses in the Discussion.

Attention to speech

For infants with CIs who learn *spoken* language, one of the most important neurocognitive processes that is critical for speech perception and language development may be a child's sustained attention to speech. To become a successful language learner, the infant must be able to distinguish and attend to communicatively meaningful signals–speech in particular–among a range of sounds in the environment. To date, research has shown that typically-developing infants with NH prefer speech over: filtered speech (Spence & DeCasper, 1987), noise (Butterfield & Siperstein, 1970), synthetic sine-waves (Vouloumanos & Werker, 2004, 2007), silence (Houston, Pisoni, Kirk, Ying, & Miyamoto, 2003), other naturally occurring sounds (Shultz & Vouloumanos, 2010), and even a whistled surrogate form of language (May, Gervain, Carreiras, & Werker, 2017 Online advance). For example, infants from 1 day to 7 months old show a preference for natural speech over sinewave, as measured by sucking rate and looking time (Vouloumanos & Werker, 2004, 2007). In addition, Houston et al. (2003) found that 6- and 9-month-old NH infants attend longer to speech sounds such as [hɑp] than to silence. Moreover, a recent study showed that the temporal and frontal areas of the brain are activated in newborns in response to familiar and unfamiliar spoken languages, but not to a whistled surrogate form (May et al., 2017 Online advance). These findings suggest that attention to speech, as well as the neural specificity for spoken language, is innate or developed from in utero auditory experience.

Although numerous studies have assessed attention to speech in children with NH, there has been very little work investigating attention to speech in children who are profoundly deaf. It is possible that a period of severe-to-profound hearing loss early in life may lead to decreased attention to speech. Only one study has directly examined attention to speech in children with CIs relative to children with NH (Houston et al., 2003). Using the Visual Habituation paradigm (VHP) (Best, McRoberts, & Sithole, 1988), Houston et al. (2003) presented infants with a checkerboard pattern and repetitions of a sound, such as [hɑp] or [ɑ] on half the trials and silence on the other half. They found that infants with CIs at 6 months post implantation showed a significantly shorter looking time to the sound versus silent trials, as compared to their hearing-age matched peers with NH. These findings suggest that implanted infants' attention to speech was reduced. Importantly, in the Houston et al. (2003) study, children with CIs and children with NH were matched based on their hearing experience; therefore, the children with NH were much younger. It is possible that the differences in attention to speech vs. silence between these two groups may be simply due to age differences, as children's preferences for different types of sounds may change throughout development. As a result, the ability to attend to speech, as well as the specific developmental change of attention to speech, in children with CIs as compared to their peers with NH remains unknown. Therefore, the first goal of the present study was to expand the findings of Houston et al. (2003) by examining attention to speech between deaf children who later received CIs and their *chronological* age-matched peers with NH at four different time periods post cochlear implantation.

Attention to speech and language development

The effects of enhanced attention to speech in young infants may not be incidental, as both theoretical models of infant language acquisition and empirical studies posit important roles for attention to speech in early spoken language development, among many other skills. According to the Word Recognition and Phonetic Structure Acquisition (WRAPSA) model (Jusczyk, 1993), infants innately attend more to some aspects of the speech signal than others. What they attend to is important for encoding acoustic details into memory. Likewise, the developmental framework for Processing Rich Information from Multidimensional Interactive Representations (PRIMIR) also includes attention to speech in the model (Curtin, Byers-Heinlein, & Werker, 2011; Werker & Curtin, 2005). Werker and colleagues proposed that three dynamic filters (the initial biases, the requirements of the specific language task, and the developmental level of the child) work together to direct children’s attention to the language-specific distributional properties, leading to successful word representation. Furthermore, there are many empirical studies demonstrating a relation between attention to speech and speech processing and language development, at least in children with NH. For example, preference for speech over non-speech sounds in infancy predicts later expressive vocabulary in both typically-developing children and children with Autism Spectrum Disorder (ASD) (Kuhl, Coffey-Corina, Padden, & Dawson, 2005; Molfese, 2000; Vouloumanos & Curtin, 2014). Specifically, infants’ attention to speech pitted against sine-waves at 12 months of age predicted expressive vocabulary at 18 months (Vouloumanos & Curtin, 2014). Moreover, 2.5- to 4-year-old children with ASD who preferred listening to non-speech over speech were more likely to exhibit deficits in expressive language ability (Kuhl et al., 2005). Early differences in attention to speech may also predict reading ability in school-age children. For example, neonatal electrophysiological responses to speech and non-speech predicted children who were either dyslexic or were below average readers at 8 years of age (Molfese, 2000).

Whereas enhanced attention to speech seems to benefit spoken language development, reduced attention to speech may affect speech processing. Although there is no direct evidence suggesting a relation between attention and speech processing in infants, previous studies showed the importance of attentional state for learning. For example, Richards and his colleagues (Richards, 1997; Richards & Hunter, 2002) presented infants with different visual stimuli depending on their degree of attention as measured by heart rate, and subsequently tested them on recognition of novel stimulus paired with old stimulus. Infants showed a novelty preference only for the objects presented during attention phases, suggesting the importance of attention for encoding visual information. Therefore, if children with CIs attend less to speech than children with NH do, then the challenge for acquiring spoken language is increased above and beyond what might be predicted simply from the quality of the auditory input provided by the CIs. Even so, the relationship between attention to speech and language development has not been explored in children with CIs. Therefore, the second goal of the current study was to fill this gap and determine whether individual differences in attention to speech post implantation would account for individual differences in speech and spoken language development in children with CIs during 2 to11 years post implantation.

Goals and predictions

The goals of this current study were twofold. The first goal was to examine whether children with CIs show reduced attention to speech as compared with their peers with NH. The second goal was to investigate whether attention to speech is associated with speech perception and spoken language development in children with CIs. To answer these questions, we conducted a 10-year longitudinal study from the time when the CIs were implanted. Specifically, we tested prelingually profoundly deaf children who received CIs and their chronologically age-matched peers with NH on their attention to speech at four time points: less than 1 month (Bin < 1 mo), 3 to 6 months (Bin 3–6 mos), 12 months (Bin 12 mos), and 18 months (Bin 18 mos) post implantation. In addition, we collected a variety of well-established standardized tests tapping different aspects of language abilities, such as speech perception, speech production, and vocabulary, from the children with CIs. These measures included Grammatical Analysis of Elicited Language (GAEL-P, Moog, Kozak, & Geers, 1983), Goldman-Fristoe Test of Articulation (GFTA, Goldman & Fristoe, 1986), The Lexical Neighborhood Test (LNT, Hay-McCutcheon, 1999), Peabody Picture Vocabulary Test (PPVT, 3rd and 4th editions, Dunn, 1997; Dunn & Dunn, 2007)[3], and Pediatric Speech Intelligibility (PSI, Jerger & Jerger, 1984). We collected these many different measures over a period of 10 years for two reasons: First, it is important to identify which aspects of spoken language are related to attention to speech early in the development; second, it is critical to continue to assess children’s speech and language skills with increasing duration of CI use in order to test the validity of attention to speech for predicting the development of spoken language skills.

Our prediction was that early auditory/language experience would affect attention to speech in children with CIs. We further predicted that if speech processing and language development are related, at least in part, to attention to speech in children with CIs, then attention to speech would be associated with measures of speech and language development. Specifically, learners with higher levels of attention to speech would have better scores in speech and spoken language tests.

These questions are important for both theoretical and clinical reasons. From a theoretical perspective, this research provides a unique opportunity to identify the possible mechanism by which early severe-to-profound hearing loss may affect other linguistic and/or cognitive processes, leading to poor language outcomes. From a clinical perspective, the research will contribute to important discoveries about the relationship between attention to speech and spoken language development in children with CIs. These findings will have significant implications to early intervention that focuses on developing attention and listening skills in children with hearing loss.

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[3] PPVT-3 edition was administered during early period of data collection, whereas PPVT-4 was administered during later period when it became available. Throughout this paper we will refer to PPVT-3 and PPVT-4 as PPVT.

Method

Participants

A total of 102 children participated in this study. All children came from English-speaking families in a Midwestern town in the United States. The CI group consisted of 22 children (10 girls, 12 boys) with severe to profound hearing loss, who were recruited from a university medical center's cochlear implant program. None of them had any comorbidity. The CIs were activated between 7.6 and 27.6 months of age (*M* = 16.97 months, *SD* = 5.47). Children with CIs were tested 1 to 7 times between 1 day and 18 months post implantation for a total of 93 testing sessions. An additional 33 testing sessions were conducted but not included in the data due to crying/fussiness (7), failure to reach the habituation criterion (9), or experimenter/equipment error (6). The testing sessions were grouped into four bins: Bin < 1 mo (less than 1 month of CI use; 41 sessions); Bin 3–6 mos (3 to 6 months of CI use; 30 sessions); Bin 12 mos (12 months of CI use; 11 sessions); and Bin 18 mos (18 months of CI use; 11 sessions). Additional demographic information for the children with CIs and the number of testing sessions included during each bin is displayed in Table 1. Eighty typically-developing children with NH (40 girls, 40 boys) were recruited as chronological age-matched control participants. These children were all born full-term, and had no history of hearing loss, speech delay, or cognitive disorder.

Each CI session was matched to a NH session based on the chronological age, thus 93 NH sessions in total. However, it was not logistically feasible to match all testing sessions for any given child in the CI group to only one child with NH. Therefore, although all 93 CI testing sessions were matched with a NH testing session, most of the children from the CI group were matched to more than one child with NH while most of the children with NH were matched to only one child with a CI. Similar to the CI group, the NH sessions were also grouped into four bins following their matched CI sessions, resulting in 41 sessions for Bin < 1 mo, 30 sessions for Bin 3–6 mos, 11 sessions for Bin 12 mos, and 11 sessions for Bin 18 mos.

Stimulus Materials

Auditory stimuli—Four speech sounds were recorded by the same female speaker: a 4-second discontinuous CVC pattern, with 8 repetitions of the 368-millisecond [hɑp] and 150 milliseconds of silence between each repetition; a 4-second continuous vowel [ɑ] with minimal pitch change (from 217 to 172 Hz); a 4-second [í] with a rising pitch contour (from 167 to 435 Hz); and a 4-second [ì]with a falling pitch contour (from 417 to 164 Hz). These four sounds were chosen because they are used in clinical trials and are among the first sound contrasts that children with hearing loss can discriminate. Each stimulus was digitized onto a 4-second .wav file.

Visual stimuli—The visual stimuli consisted of an attention getter (a laughing baby) and a visual display (a white and red static checkerboard pattern).

Apparatus and procedures

Infants were tested using the central fixation procedure (Best et al., 1988), which was successfully adapted by Houston et al. (2003) to assess speech perception skills in infants with CIs. Each child was seated on the caregiver's lap in front of a TV monitor in the middle of a quiet and comfortable double-walled IAC sound booth. Speech stimuli were presented to the children via loudspeakers on the TV monitor at a comfortable level of 70±5 dB SPL. The presentation of the stimuli was controlled by an experimenter in an adjacent control room using a MacIntosh computer operating the Habit program software (Oakes, Sperka, & Cantrell, 2015). The experimenter observed the children via a monitor that was linked to a camera in the testing booth. Caregivers listened to a combination of loud music and speech babble over sound-attenuating enclosed headphones (Peltor Aviation Headset 7050) so that they were not able to hear the stimuli presented to the infants. Likewise, the experimenter was blinded from the stimuli and experiment conditions while in the control booth.

Children were randomly assigned to one of the four conditions: [hɑp], [ɑ], [í], and [ì]. They were presented with two types of trials: sound trials and silent trials. Sound trials consisted of the visual display (checkerboard pattern) and one of the 4-second sound files, which was the same sound throughout the testing session. Silent trials consisted of the visual display only. Before each trial, children were presented with the attention getter to orient them to the center of the TV monitor. When the child was fixated on the attention getter, the experimenter initiated the trial. Each trial continued until the child looked away from the visual display for 1 second. The duration of the child's looking time towards the checkboard was measured for each trial. The test trials were grouped into blocks of four in which two sound trials and two silent trials were presented in random order. There were 20 blocks in total. The experiment ended when the child met the habituation criterion: mean looking time during a block of trials that was at least 50% shorter than the mean looking time during the first block of trials. The dependent measure was the average looking times to speech trials and the average looking times to silent trials across trials and blocks for each participant. If children prefer speech sounds, they would look longer to the visual display during the sound trials than during the silent trials.

Outcome measures

Standardized tests of vocabulary (PPVT), spoken word recognition (LNT, GAEL-P, PSI), and articulation (GFTA) were administered to the children with CIs over 2 to 11 years post implantation. The standardized measures and descriptions are shown in Table 2. We grouped the data gathered during the 2–11 years post implantation into 20 intervals, with 6 months as one interval: Interval 1: 1–1.5 years post implantation; Interval 2: 1.5–2 years post implantation; Interval 3: 2–2.5 years post implantation, and so forth. Note that these measures were not obtained from some of the children due to several reasons: not being old enough for specific tests; or moving away and no longer participating in the research study. In addition, due to the longitudinal nature of the study, not all the children with CIs participated in all the intervals. Note also that the standardized tests that were administered to the children with CIs also varied as a function of their chronological age, such that some tests (e.g., GAEL-P, PSI) were administered only during early periods because these tests are not valid for older children. Total numbers of intervals for each test that have been administered on each child in the CI group are displayed in Table 1.

Results

Attention to speech

To assess whether repeating speech sounds engaged children' attention more than silence, mean looking times during the sound and the silent trials across blocks were computed for each child. Descriptive statistics for attention to speech at the four bins are shown in Table 3. Inspection of Table 1 reveals that our data was unbalanced and some of the participants were repeatedly measured; therefore, a mixed model analysis is most appropriate (Baayen, 2011). These analyses were implemented using the lmer function, part of the lme4 package (Bates, Mächler, Bolker, & Walker, 2015) in the *R* environment (R Development Core R Core Team, 2014). Although mixed models are a relatively new statistical tool in the developmental field, it is popular for hierarchically-organized data in a wide variety of disciplines, especially in settings where repeated measurements are made on the same statistical unit. In contrast to a more traditional approach with data aggregation and repeated measures ANOVA analysis, lmer allows controlling for the variance associated with random factors without data aggregation. Therefore, we fitted 4 mixed-effects models with Type (sound trial, silent trial) and Hearing status (CI, NH) as fixed factors; Condition ([hɑp], [ɑ], [í], [ì]) and Session as random factors; and Looking time as the dependent variable for each bin (Bin < 1 mo, Bin 3–6 mos, Bin 12 mos, and Bin 18 mos). Because some children were repeatedly measured, we also included Participant as a random intercept to control for the influences associated with this factor. The full model, fitted with the complete structure, was translated to *lmer(Lookingtime ~ Hearing status*Type +(1/Condition) + (1|Participant) + (1|Session) + (1|Hearing status:Session) + (1|Type:Session), data=mydata)*. For the sake of brevity, we present only the *F* tests from the lmer results here. The reported *F* and *p*-values were estimated using the anova() function on lmer objects in package lmerTest (Kuznetsova, Brockhoff, & Christensen, 2015) in *R*. The post hoc contrast comparisons were conducted using the lsmeans() function in package lsmeans (Lenth, 2016) and were adjusted by Tukey correction.

At Bin < 1 mo (41 CI and 41 NH sessions, 20 unique CI participants; 37 unique NH participants), there was a significant interaction of Hearing status and Type, $F(1, 40) = 7.63$, $p = .009$. In addition, both the main effects of Hearing status and Type were significant, $F(1, 44.11) = 6.53$, $p = .014$, and $F(1, 40) = 7.47$, $p = .010$, respectively. An inspection of the interaction revealed that children with NH looked significantly longer during the sound trials ($M = 9.44$, $SD = 4.15$) than during the silent trials ($M = 7.12$, $SD = 2.95$), $t(77.1) = 3.89$, $p < .001$; whereas children with CIs looked equally long during the sound trials ($M = 6.25$, $SD = 4.91$) and during the silent trials ($M = 6.04$, $SD = 3.43$), $t(77.1) = .36$, $p = .720$. This suggests that the children with CIs showed reduced attention to speech as compared to their peers with NH with less than one month of CI experience.

At Bin 3–6 mos (30 CI and 30 NH sessions, 20 unique CI participants, 28 unique NH participants), the main effect of Type was significant, $F(1, 29) = 5.36$, $p = .028$, because both groups of children, in aggregate, looked longer during the sound trials ($M = 7.86$, $SD = 4.69$)

than during the silent trials ($M = 6.43$, $SD = 3.61$), $t(29) = 2.31$, $p = .028$; however, the main effect of Hearing status and the interaction of Hearing status and Type were not significant, $F < 1.00$, $p > .326$. This suggests that with 3 to 6 months of CI experience, the children with CIs demonstrated similar attention to speech as their same-aged peers with NH.

At Bin 12 mos (11 CI and 11 NH sessions, 11 unique CI participants, 11 unique NH participants), there was a marginally significant main effect of Type, $F(1, 20) = 3.45$, $p = .078$. Both groups tended to look longer during the sound trials ($M = 7.48$, $SD = 3.87$) than during the silent trials ($M = 6.45$, $SD = 2.70$), $t(20) = 1.86$, $p = .078$. However, neither the main effect of Hearing status nor the interaction of Hearing status and Type was significant, $F < .97$, $p > .336$. This suggests that with 12 months of CI use, the children with CIs showed similar degree of attention to speech as compared to their same-aged peers with NH.

At Bin 18 mos (11 CI and 11 NH sessions, 11 unique CI participants, 11 unique NH participants), no main effects or interactions were significant, $F < 1.41$, $p > .251$, suggesting that both groups did not show any preference for sound versus silent trials. Looking times during the four bins are shown in Figure 1.

Taken together, these results suggest that children with NH preferred the sound trials over the silent trials at Bin < 1 mo, Bin 3–6 mos, and trended toward the same direction at Bin 12 mos; however, they did not show any preference at Bin 18 mos. Children with CIs did not show any preference for sound trials at Bin < 1 mo. However, they showed a similar degree of attention to speech as compared to their peers with NH at Bin 3–6 mos, Bin 12 mos, and Bin 18 mos.

Attention to speech during infancy and language outcomes in children with CIs

The next question we turned to is whether individual differences in attention to speech are associated with speech perception and language outcomes at later points, specifically in children with CIs. To answer this question, we calculated an attention to speech (ATS) score by subtracting looking time during the silent trials from the looking time during the sound trials (Sound-Silent) for Bin 3–6 mos, with positive values indicating a preference for sound. If a child was tested more than once during this bin, the ATS scores were averaged.

Specifically, we fitted regression models with the ATS score from the Bin 3–6 mos as a predictor, CI participant and Interval as random variables, and each of the outcome scores as the dependent variable. These analyses were implemented using the lm() function, part of the lme4 package (Bates et al., 2015) in *R*. Each full model, fitted with the complete structure, was translated to $lm(Outcome \sim ATS_{Bin\ 3–6\ mos} + (1|CI\ participant) + (1|Interval), data=outcome)$[4]. The reported results were estimated using the summary() function in package lmerTest (Kuznetsova et al., 2015). Table 4 summarizes five multiple regression models evaluating the effects of attention to speech from Bin 3–6 mos on measures of speech and language development. Results showed that for LNT, the regression model was

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[4] The reason that we modeled Interval as a random factor was because the available outcome measures was not sufficient to reveal a developmental change (see supplementary materials for the data attrition information). Due to the nature of our data distribution, modeling interval as a random factor would be more appropriate. As suggested by one of the reviewers, we also ran additional model with Interval as a fixed factor; the results did not differ.

significant, $p = .007$ and the adjusted R^2 was .09, suggesting that attention to speech during Bin 3–6 mos predicts LNT scores in children with CIs. Other regression models were not significant, *p*s > .240. These findings suggest that deaf children's attention to speech during 3 to 6 months post implantation may serve as a valuable predictor for later spoken word recognition.

To determine which demographic factor(s) contribute to explaining CI infants' attention to speech during Bin 3–6 mos, we fitted a multiple regression model with age at implantation and residual hearing as continuous predictors, communication mode (oral vs. total communication) as a categorical factor, and ATS score, as the dependent variable: $lm(ATS_{Bin\ 3\text{–}6\ mos} \sim Age\ at\ implantation + Residual\ hearing + Communication\ mode, data=demographic)$. The regression model was not significant, $F(3, 18) = 2.12$, $p = .140$, and the adjusted R^2 was .158. These findings suggest that none of the demographic factors evaluated were associated with attention to speech in children with CIs 3 to 6 months post implantation.

Discussion

In this section, we discuss the findings in terms of the questions raised at the outset of this paper: 1) whether children with CIs show reduced attention to speech as compared to their same-aged peers with NH, and 2) whether attention to speech post implantation predicts standardized speech and spoken language test scores that might reflect spoken language skills in children with CIs. We also consider limitations of this study and propose some future directions.

Effects of early severe-to-profound hearing loss on attention to speech

First, we found that children with CIs showed reduced attention to speech as compared to their chronologically age-matched peers with NH at Bin < 1 mo. However, these differences should be interpreted with caution, because audiologists tend to be conservative with programming CI processors during the first few weeks post implantation as they are still assessing the threshold and comfortable levels for the CI recipients. Therefore, reduced attention to speech within the first month post implantation in infants with CIs may be due to poor access to auditory input. Due to these considerations, we will focus our discussion on the findings from Bin 3–6 mos, Bin 12 mos, and Bin 18 mos. Second, children with CIs, similar to their peers with NH, showed enhanced attention to speech during Bin 3–6 mos. The rapid change of the ability to attend to speech over the 3 to 6 months post implantation suggests that experience with sounds via CIs improves young CI recipients' attention to speech. Finally, neither group showed a significant preference for speech during Bin 12 mos or Bin 18 mos. This may be because our stimuli consisted of repetitions of monosyllables, which older children in both the CI and the NH groups found not very interesting. It could also be that due to developmental change, older children begin to pay less attention to isolated speech as they explore a world of dynamic multimodal stimulation to all the senses.

Taken together, the CI and the NH groups in our study, despite the differences in their hearing experience, showed similar levels of attention to speech during Bins 3–6 mos, Bin 12 mos, and Bin 18mos. In addition, there is a gradual decline in attention to speech in both the CI and the NH groups with the increasing of chronological age. These findings seem to be in contrast with Houston et al. (2003)'s findings that attention to speech is greatly reduced in the CI group who had 6 months hearing experience as compared to the control group with matched *hearing* age. In what follows, we explain how these seemingly opposite findings may in fact be complementary in providing a complete picture in helping us to understand attention to speech in children with CIs.

First, it is possible that attention to speech is determined by experience-independent processes, which develops regardless of the experience with the input (see Tomblin, Barker, and Hubbs (2007) for discussion). However, this does not necessarily suggest that there is no difference between the NH and the CI groups with regard to their attention to speech skills. Note that in contrast to children with NH, children with CIs do not have access to speech sounds before implantation; this early period may be a critical period for infants to develop strong listening skills. Indeed, NH infants' attention to speech is higher at 6 months compared to at 9 months (Houston et al., 2003). In addition, our findings show a gradual decline in attention to speech with age. There is also evidence that the neural circuitry is specialized for processing speech during the first 4 months (Shultz, Vouloumanos, Bennett, & Pelphrey, 2014). On this account, a period of severe-to-profound hearing loss early in life affects the developmental pattern of attention to speech in children with CIs, such that they miss the sensitive periods for developing strong attention to speech skills and never reach the same level of attention to speech that young infants with NH have. This may have major consequences for infants with CIs to acquire speech perception skills that are critical for learning spoken language.

It is also possible that attention to speech is driven by experience-dependent processes, such that infants' attention to speech is shaped by experience with the input. If this were the case, then the similar degree of attention to speech we observed between children with CIs and their chronologically age-matched peers with NH may be due to an interaction between two factors: (1) relatively immature attention-to-speech mechanisms due to less hearing experience, which should result in more attention to speech than age-matched peers; and (2) weakened attention-to-speech mechanisms due to atypical hearing experience, which may result in less attention to speech than age-matched peers. In other words, children with CIs in our study, who were younger than children with NH in terms of their hearing age, should have shown higher level of attention to speech as compared to the control group (note that we discussed earlier that children in our study showed a gradual decline in attention to speech with age, either due to the nature of our stimuli or developmental change). However, the degraded nature of the input and atypical developmental course of auditory and/or language experience that children with CIs received may have reduced their attention to speech as compared to their hearing age-matched controls. The interaction of these two factors may have led to the seemingly similar degree of attention to speech between children with CIs and children with NH in our study.

What might cause reduced attention to speech in children with CIs compared to their hearing age-matched peers with NH? We consider two possible explanations, which were briefly mentioned above. First, it is possible that a lack of *auditory exposure* early in life, including in utero, as well as the degraded nature of input via CIs, may affect the development of basic neurocognitive processes, leading to reduced ability to respond to speech signal from their auditory environment (Conway et al., 2009). Indeed, neurophysiological studies provide evidence that sensory deprivation at birth has dramatic effects on the organization of sensory cortices and brain circuitry in response to challenges that interfere with its normal development, such as visual deprivation, auditory deprivation, etc. (Merabet & Pascual-Leone, 2010; Mills et al., 2004; Rauschecker & Korte, 1993; Voss & Zatorre, 2012). As just one example, deaf individuals show greater recruitment of occipital-parietal cortical areas related to visual attention processing compared with their NH controls (Bavelier et al., 2001; Bavelier et al., 2000; Neville & Lawson, 1987). In addition, the degraded speech signal provided by CIs may also contribute to the differences because the acoustic signal transmitted to the auditory nerve by CIs is underspecified relative to the speech signal received by normally functioning cochlea, which may be inherently less interesting (Zeng, 2004).

An alternative hypothesis is that early *language deprivation* leads to differences in attention to speech between the CI and the NH group. Some evidence suggests that the cognitive processes required for modality-independent processing are not affected by hearing loss in these children, who achieve typical language and social milestones in infancy (Hall et al., 2017; Marshall et al., 2015; Peterson & Siegal, 2000; Petitto & Marentette, 1991). For example, Hall et al. (2017) examined executive function in a group of Deaf children from Deaf families, who have a history of auditory but not language deprivation. They found that scores among the Deaf signers were age-appropriate and similar to scores among their typically-developing peers. It should be noted that the children with CIs in our study lacked exposure to natural human language (spoken or signed) prior to implantation. Therefore, it is not possible to tease apart these two hypotheses. Future studies are encouraged to examine the source of attentional deficits in children with CIs early in development.

In addition, we also found that none of the demographic factors examined – age at implantation, amount of residual hearing, and communication mode – was associated with attention to speech in children with CIs at 3 to 6 months post implantation. These findings may seem surprising, because these variables are often found to be correlated with performance on language tasks in deaf infants who received CIs later (Fryauf-Bertschy et al., 1997; Houston, Stewart, Moberly, Hollich, & Miyamoto, 2012; Kirk et al., 2002; Miyamoto, Svirsky, & Robbins, 1997; Svirsky, Teoh, & Neuburger, 2004). This may be due to that all of the infants in our study received cochlear implants relatively early (prior to 2 years of age) and their residual hearing was rather homogenous. Moreover, these results should be interpreted with caution given the small number of children with CIs.

Attention to speech and language development

Second, and more importantly, attention to speech predicts later word recognition in the children with CIs. Specifically, we found that children with CIs who looked longer during the speech compared to the silent trials at 3 to 6 months post implantation scored higher on LNT measures gathered during 2 to 11 years post implantation. These findings lend support to the WRAPSA and the PRIMIR theoretical models that attention to the language-specific properties lead to successful word recognition and representation (Curtin et al., 2011;

Jusczyk, 1993; Werker & Curtin, 2005). In addition, these findings also provide the first empirical evidence connecting attention to speech to later spoken language development in children with CIs, suggesting that attention to speech early in life may provide a foundation for subsequent speech and language development.

The findings that attention to speech only predicts LNT scores, but not PSI or GAEL-P scores, may be due to three reasons. First, LNT, which is an open-set word recognition task, can be fundamentally different from the other two closed-set word recognition tasks, PSI and GAEL-P. This is because the information processing demands, particularly with respect to their level of competition between potential responses, are quite different (Clopper, Pisoni, & Tierney, 2006). Indeed, previous studies showed robust effects of lexical competition and talker variability in open-set tasks but not in closed-set tasks, suggesting that open-set tests of spoken word recognition may be better assessments of speech recognition skills (Kirk, Pisoni, & Miyamoto, 1997; Mullennix, Pisoni, & Martin, 1989). Second, the number of data points gathered for PSI ($N = 33$) and GAEL-P ($N = 39$) were much smaller than for the LNT ($N = 68$). Thus, it is possible the differences are due to differences in their statistical power. Third, the PSI and GAEL-P measures were gathered during early period of post implantation, between Intervals 3–7 and 3–6, respectively, whereas the LNT was gathered over a longer time span, between Intervals 3 and 22. Previous studies showed that greater improvements in speech perception are generally observed with increased duration of CI use (Miyamoto, Kirk, Svirsky, & Seghal, 2000). Therefore, it is possible that attention to speech may be better at predicting word recognition over a longer period post implantation.

The finding that variability in the ability to attend to speech in children with CIs contributes to explaining variability in open-set word recognition raises a fundamental question as to why higher level of attention to speech is associated with better word recognition. Although it is clear that there is potential advantage afforded a child who has higher levels of attention to speech, less is known about the nature of the relationship between attention to speech and language development. It is possible that greater attention to speech would allow infants with CIs more access to speech, leading to better encoding, storage, and retrieval of acoustic-phonetic and phonological information into memory. This process may in turn accelerate the segmentation of words from continuous speech and eventually bootstrap language learning at higher levels.

However, a predictive relationship between attention to speech and word recognition does not necessarily entail a direct causal relationship. It is possible that attention to speech and later word recognition share variance because they are both affected by other factors, such as general cognitive and/or linguistic abilities. Therefore, it is the individual differences in other domains, rather than in attention to speech per se, that relates to individual differences in word recognition later in the life. Second, the observed association between attention to speech and later word recognition may also be explained by variation in CI infants' auditory abilities. To disambiguate the multiple factors that may be at play in language development in children with CIs, future studies taking a multivariate approach to investigate the relationship between infants' auditory processing, linguistic skills, general cognitive abilities, and their language development are encouraged.

Our findings give rise to a challenge: How to enhance attention to speech early on for infants with CIs. There is growing evidence that spoken language learning in both typically-developing infants with NH and infants with hearing loss may be enhanced by their social interest in speech, especially the kind of speech that is directed to them; namely, infant-directed speech (IDS). Recent studies has also demonstrated an association between attention to IDS and language skills in children with hearing loss (Robertson, von Hapsburg, & Hay, 2013; Wang, Bergeson, & Houston, 2017). In ongoing research, we are exploring the relationship between the quality and quantity of IDS in the listening environment, attention to IDS, and later spoken language outcomes in infants with CIs. In addition, music training may also serve to enhance attention to speech (Barton & Robbins, 2015; Strait, Slater, O’Connell, & Kraus, 2015). For instance, length of music training during childhood is associated with reduced response variability to the attended speech in school-aged child and adult musicians (Strait et al., 2015). Finally, as we noted above, if attention to speech is affected by language deprivation independently from any influence of auditory deprivation, then exposing deaf children with natural sign language before implantation may serve as a protective role in their ability to attend to speech post implantation thus lead to better spoken language. Recent studies provided empirical evidence that exposure to sign language from fluent sign language users can facilitate spoken language development post implantation (Davidson, Lillo-Martin, & Chen Pichler, 2014; Kozak, Chen Pichler, Quadros, Cruz, & Pizzio, 2013).

A limitation of our study is that we only tested children’s attention to speech versus silence. Therefore, it is possible that the findings are about auditory attention in general, rather than attention to speech per se. Nevertheless, existing evidence demonstrates that children with CIs distinguish between different types of auditory stimuli. For instance, infants with CIs prefer infant-directed speech over adult-directed speech (Wang et al., 2017); furthermore, different patterns of neural activation to speech vs. nonspeech have also been found in children with CIs as compared to normal hearing group. Specifically, Sevy et al. (2010) shows a right-sided brain activation to speech in children with CIs; this is in contrast to the adults with NH and children who show left-hemisphere activation to speech. Despite these findings, future work will need to address this question by comparing attention to speech versus nonspeech sounds in children with CIs.

Conclusions

Differences in speech and language outcomes in children with CIs are not fully explained by conventional demographic and medical factors. Some of the unexplained variance may be due to differences in cognitive processes that provide the foundations for the development of speech and language skills. Our findings suggest that a period of severe-to-profound hearing loss early in life affects attention to speech in children with CIs, which, in turn, may have a negative effect on their later speech perception. These results inform early language acquisition theories, such as WRAPSA and PRIMIR, and bring insights into our understanding of the role of early severe-to-profound hearing loss on cognitive processes. In addition, there are potential clinical implications for these outcomes; specifically, early intervention programs may consider including attention to speech evaluation and habilitation.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

Acknowledgments

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Research highlights

- Early severe-to-profound hearing loss affects attention to speech in children with cochlear implants.
- Attention to speech 3 to 6 months post implantation predicts later language in children with cochlear implants.

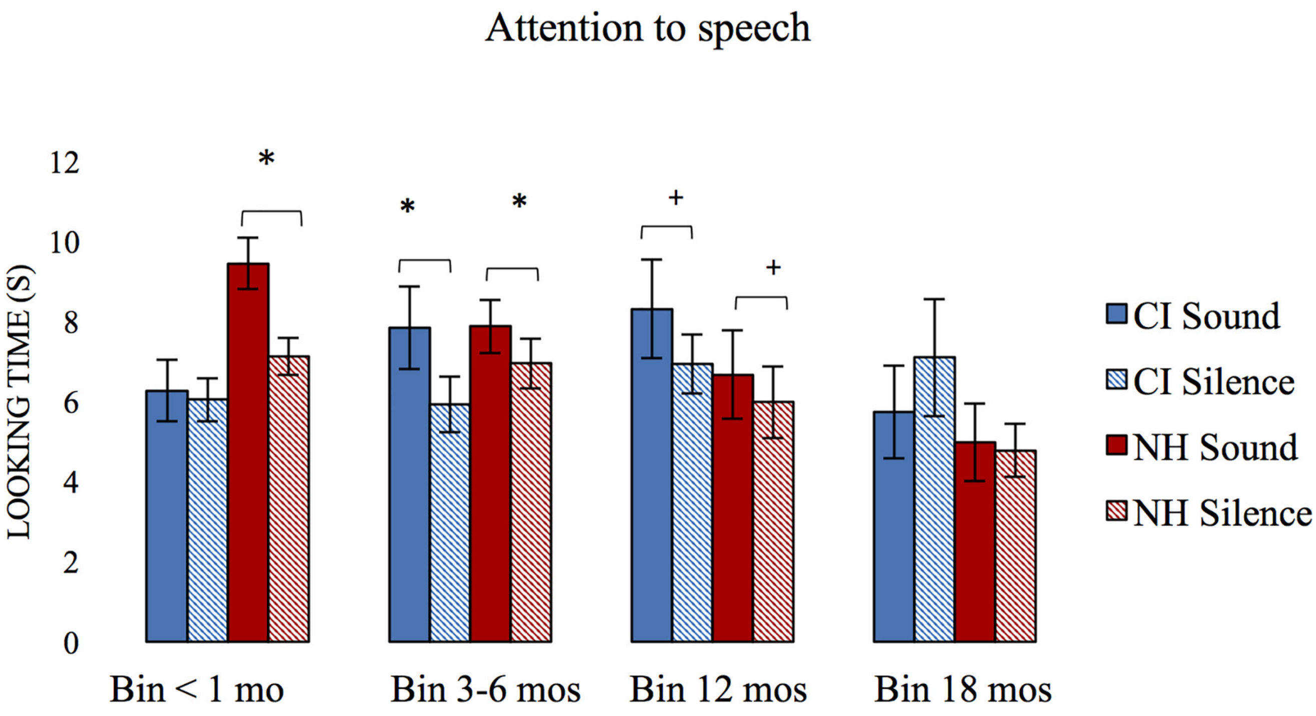

Figure 1.

The average looking times (in seconds) during sound and silent trials during the four bins for children with CIs and their peers with NH. Error bars indicate standard error. *: $p < .05$; $^{+}$: .05 < $p < .01$.

Table 1

Demographic information and data points available for children with cochlear implants (CIs)

					No. of sessions for bin					Outcome measures-No. of intervals				
ID	**Sex**	**Age at CI**	**Mean PTA**	**Com Mode**	**<1 mo**	**3–6 mos**	**12 mos**	**18 mos**	**Total**	**GAEL-P**	**GFTA**	**LNT**	**PPVT**	**PSI**
CI01	M	7.6	86	AC	1	2	0	1	4	2	2	2	4	2
CI02	F	17.29	72	AC	2	2	1	0	5	0	0	0	0	0
CI03	F	12.2	89	AC	3	2	1	1	7	3	6	7	8	1
CI04	F	11.08	90	TC	0	1	0	1	2	1	0	1	1	0
CI05	F	23.67	65	TC	2	2	0	0	4	2	0	0	0	0
CI06	F	17.42	90	TC	3	1	0	1	5	0	5	8	9	3
CI08	F	19.07	79	TC	0	1	1	1	3	3	4	5	6	4
CI12	M	16.96	86	TC	2	1	1	1	5	2	2	1	5	3
CI14	M	12.72	87	TC	2	2	1	1	6	2	6	6	8	2
CI16	M	13.87	73	AC	2	2	0	1	5	1	0	0	1	1
CI17	F	21.66	82	TA	1	0	0	0	1	0	0	0	0	0
CI18	M	11.84	90	AC	3	1	1	1	6	2	7	5	8	2
CI19	F	10.29	78	TC	2	2	1	1	6	3	0	4	5	2
CI22	M	22.12	59	AC	2	2	1	0	5	2	5	5	7	1
CI23	M	24.16	72	AC	2	1	1	0	4	1	2	1	3	2
CI25	M	16.11	90	AC	3	1	0	1	5	4	5	3	8	2
CI26	F	27.61	76	TC	2	1	0	0	3	3	7	5	8	3
CI28	M	16.8	81	TC	2	2	0	0	4	2	5	2	6	2
CI29	M	16.5	90	AC	2	2	0	0	4	2	3	2	3	1
CI31	M	24.28	72	AC	3	1	1	0	5	1	3	4	7	1
CI32	M	17.14	90	AC	1	0	1	0	2	1	0	0	0	0
CI33	F	8.29	90	AC	1	1	0	0	2	2	6	7	7	1
Mean		16.97	81.61	Total	41	30	11	11	93	39	68	68	104	33
SD		(5.47)	(9.23)		Unique participant No.					19	15	17	18	17

Note. PTA = Mean Unaided Pure-tone average before implantation in dB; Com Mode: The type of communication program the infant was following in speech language therapy; OC (oral communication): exclusively spoken; TC (total communication): a combination of spoken language and Signed-Exact English. GAEL-P = Grammatical Analysis of Elicited Language – Pre-Sentence Level; GFTA:

Goldman-Fristoe Test of Articulation; LNT = Lexical Neighborhood Test; PPVT = Peabody Picture Vocabulary Test; PSI = Pediatric Speech Intelligibility Test. Unique participant No.: the number of participants who were administered the corresponding test.

Table 2

Standardized tests administered

Construct	Test	Abbreviation	Scales Used	Intervals administered
Speech perception	Grammatical Analysis of Elicited Language (Moog et al., 1983)	GAEL-P	Pre-sentence level	3–6
Speech articulation	Goldman-Fristoe Test of Articulation (Goldman & Fristoe, 1986)	GFTA	Errors	3–16
Speech perception	The Lexical Neighborhood Test (Hay-McCutcheon, 1999)	LNT	Recorded multi-talker: Easy phoneme & Hard phoneme	4–22
Receptive vocabulary	Peabody Picture Vocabulary Test (Dunn, 1997; Dunn & Dunn, 2007)	PPVT		3–22
Speech perception	Pediatric Speech Intelligibility (Jerger & Jerger, 1984)	PSI	Sentences- auditory only	3–7

Note: Each interval represents a period of 6 months. Interval 3: 1–1.5 years post implantation; Interval 4: 1.5–2 years post implantation, and so forth.

Table 3

Age (Mean, SD) and looking time (Mean, SD) during the sound and the silent trials for the CI and the NH groups

	CI group				NH group		
Bin	**Age**	**CI age**	**Sound**	**Silent**	**Age**	**Sound**	**Silent**
< 1 mo	17.77 (5.29)	.55 (.41)	6.26 (4.91)	6.05 (3.43)	17.80 (5.56)	9.44 (4.15)	7.12 (2.95)
3–6 mos	20.62 (5.52)	4.50 (1.69)	7.84 (5.62)	5.92 (3.82)	20.60 (5.70)	7.87 (3.62)	6.94 (3.36)
12 mos	28.87 (5.23)	11.74 (.55)	8.3 (4.08)	6.93 (2.46)	28.94 (5.30)	6.66 (3.65)	5.98 (2.96)
18 mos	31.42 (3.78)	17.85 (.48)	5.73 (3.82)	7.09 (4.85)	31.54 (3.62)	4.98 (3.24)	4.77 (2.20)

Note: Age reported in months; looking time in seconds.

Table 4

Multiple regression models predicting standardized test scores (GAEL, GFTA, LNT, PPVT, and PSI administered at 2–11years post implantation) from attention to speech measures (ATS score) gathered during infancy; Beta (standard error).

	Outcome measures				
Predictor	**GAEL**	**GFTA**	**LNT**	**PPVT**	**PSI**
Bin 3–6 mos	−.14(.34)	.57(.72)	1.64(.59) **	−0.54(.46)	−0.03(.19)
Adj R^2	.020	.005	.090	.003	.031
Model p	.676	.434	.007 **	.240	.884

Note.

$^{**}p < .01$,

$^{*}p < .05$,

$^{+}.05 < p < .01$.

GAEL-P = Grammatical Analysis of Elicited Language – Pre-Sentence Level; GFTA: Goldman-Fristoe Test of Articulation; LNT = Lexical Neighborhood Test; PPVT = Peabody Picture Vocabulary Test; PSI = Pediatric Speech Intelligibility Test.