Executive Functioning and Speech-Language Skills Following Long-Term Use of Cochlear Implants

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Received December 12, 2013; revisions received April 8, 2014; accepted April 29, 2014

Neurocognitive processes such as executive functioning (EF) may influence the development of speech-language skills in deaf children after cochlear implantation in ways that differ from normal-hearing, typically developing children. Conversely, spoken language abilities and experiences may also exert reciprocal effects on the development of EF. The purpose of this study was to identify EF domains that are related to speech-language skills in cochlear implant (CI) users, compared to normal-hearing peers. Sixty-four prelingually deaf, early-implanted, long-term users of CIs and 74 normal-hearing peers equivalent in age and nonverbal intelligence completed measures of speech-language skills and three domains of EF: working memory, fluency-speed, and inhibition-concentration. Verbal working memory and fluency-speed were more strongly associated with speech-language outcomes in the CI users than in the normal-hearing peers. Spatial working memory and inhibition-concentration correlated positively with language skills in normal-hearing peers but not in CI users. The core domains of EF that are associated with spoken language development are different in long-term CI users compared to normal-hearing peers, suggesting important dissociations in neurocognitive development.

Hearing loss is a common condition of childhood, with 3% of children showing some degree of hearing loss and nearly 1% having at least a mild bilateral hearing loss (Mehra, Eavey, & Keamy, 2009). The most significant forms of hearing loss involve severe (greater than 70 decibels of hearing loss) and profound (greater than 90 decibels of hearing loss) deafness (Clark, 1981). Hearing loss, especially in its more severe forms, is associated with specific developmental risks, particularly in the area of speech and language skills (Muse et al., 2013). As a result, early identification and medical interventions to improve access and exposure to sound have been aggressively pursued as public health priorities (Bhatia, Mintz, Hecht, Deavenport, & Kuo, 2013).

During the 1990s, cochlear implantation became available as a medical treatment for children with severe-to-profound sensorineural hearing loss, a majority of whom now routinely receive cochlear implants (Bradham & Jones, 2008). A cochlear implant (CI) is a device consisting of an external component that processes sound into electrical signals that are sent to an internal receiver and electrode array that stimulates the auditory nerve. Cochlear implantation restores some attributes of hearing to many children who have profound hearing loss, although the sensory information provided by a CI is significantly degraded and underspecified relative to normal hearing. Nevertheless, the restoration of auditory experience allows many children with CIs to develop substantial receptive and expressive spoken language skills (Geers & Sedey, 2011). Better spoken language outcomes are related to a set of conventional demographic and hearing history variables such as earlier age at implantation, shorter duration of deafness before implantation, greater residual hearing prior to implantation, and use of an auditory-oral mode of communication (Geers & Sedey, 2011).

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doi:10.1093/deafed/enu011
Advance Access publication June 5, 2014

Despite the remarkable advances in CI technology, many children with CIs lag significantly behind their age-peers in speech and language skills, and considerable unexplained variability is routinely observed in speech and language outcomes in this clinical population (Geers & Sedey, 2011; Niparko et al., 2010). This variability is not fully accounted for by conventional outcome predictors such as characteristics of the CI device, hearing history, demographic, or medical characteristics (Pisoni, Conway, Kronenberger, Henning, & Anaya, 2010). Attempts to better understand the sources of variability in speech and language development following cochlear implantation have led to the discovery that other areas of neurocognitive functioning are also adversely affected in many children with CIs, and much of the heretofore unexplained variability in speech and language outcomes may be due to the contributions of these other areas of neurocognitive functioning. For example, verbal working memory capacity (actively maintaining phonological and lexical memory codes in immediate memory during concurrent mental operations) is atypical and significantly delayed in samples of children with CIs compared to normal-hearing (NH) control samples (Harris et al., 2013). Furthermore, poorer verbal working memory capacity has been found to predict delays in speech perception and language skills in children with CIs, even after accounting for variance contributed by conventional demographic, device, and medical outcome variables (Pisoni, Kronenberger, Roman, & Geers, 2011).

Kronenberger et al. (Kronenberger, Pisoni, Henning, & Colson, 2013) reported delays in three broad domains of executive functioning (EF) in children with CIs compared to an age- and nonverbal intelligence-matched NH control sample: (1) verbal working memory, (2) fluency-speed (processing speed during cognitive operations requiring effortful controlled attention), and (3) inhibition-concentration (the ability to sustain a consistent level of attention and to control responses). EF involves the regulation, allocation, and management of thoughts, behaviors, and emotions in the service of planning, goal-direction, and organization (Gioia, Isquith, Guy, & Kenworthy, 2000). The impact of reduced early auditory experience with sound patterns (from deafness and subsequent degraded and underspecified auditory input from the CI) on downstream neurocognitive development may put CI users at greater risk than normal-hearing peers for delays in the development of critical building blocks of EF abilities, including sequential processing skills (Conway, Pisoni, & Kronenberger, 2009) and self-regulatory language skills (Pisoni et al., 2010).

Previous findings of EF delays in children with CIs (Figueras, Edwards, & Langdon, 2008; Kronenberger et al., 2013) are consistent with the hypothesis that a period of profound deafness during critical early periods of brain development, combined with degraded underspecified auditory experience following implantation, may impact broad, domain-general areas of neurocognitive development (Pisoni et al., 2010). The brain is a highly interconnected information-processing system that develops based on complex interactions between neural activity and sensory stimulation from the environment, including auditory stimulation. As a result, deprivation in early auditory experiences and activities may influence the development of more basic elementary neurobiological and cognitive functions extending well beyond spoken language skills. Importantly, early auditory experiences provide temporal patterns to the developing brain, which may be important for the development of sequential processing abilities such as sustained attention and memory for serially presented items (Conway et al., 2009). Sustained attention and sequential memory processes are critical developmental building blocks for executive functions such as controlled attention, planning, working memory, and fluent-efficient cognitive processing (Pisoni et al., 2010).

A large body of research has demonstrated that EF skills such as working memory and cognitive control are critical for the development of speech and language skills in NH children (Gathercole & Baddeley, 1993). Language and EF are dependent on each other for development, particularly through childhood (Barkley, 2012; Singer & Bashir, 1999; Ylvisaker & DeBonis, 2000). Research with NH children who have speech and language delays demonstrates weaknesses in multiple EF areas, even when potential confounds such as age, nonverbal IQ, and verbal IQ are accounted for (Henry, Messer, & Nash, 2012). Furthermore, working memory, fluency-speed, and inhibition have been specifically identified as at-risk in NH children with language delays (Henry et al., 2012). Both working memory and fluency-speed are related to the development of reading skills in NH children (Gathercole, Alloway, Willis, & Adams, 2006; Gathercole & Pickering, 2000; Nittrouer, Caldwell, Lowenstein, Tarr, & Holloman, 2012), and long-term academic achievement in language subjects is predicted by early working memory skills (Gathercole, Tiffany, Briscoe, & Thorn, 2005).

Recent findings have also documented close links between several types of EF skills and speech and language outcomes in children with CIs, accounting for prior unexplained variance in speech-language development in this clinical population (Figueras et al., 2008; Pisoni et al., 2010). Of the three broad EF areas identified by Kronenberger et al. (2013) as at-risk in long-term CI users, working memory has received the most investigation showing associations with language in children with CIs. Verbal-phonological working memory capacity is significantly associated with speech perception skills (Cleary, Pisoni, & Kirk, 2000; Nittrouer, Caldwell-Tarr, & Lowenstein, 2013), grammar (Willstedt-Svensson, Löfqvist, Almqvist, & Sahlén, 2004), vocabulary (Cleary et al., 2000; Geers, Pisoni, & Brenner, 2013; Nittrouer et al., 2013; Wass et al., 2008), reading (Geers et al., 2013), novel word learning (Willstedt-Svensson et al., 2004), and conversational communication (Ibertsson, Hansson, Asker-Arnason, Sahlén, & Mäki-Torkko, 2009; Lyxell et al., 2008) in children with CIs. Furthermore, electrophysiological studies using the mismatch negativity (MMN) paradigm (Ortmann et al., 2013) have found relations between digit span scores and MMN frontal activity associated with good speech perception skills in children with CIs. In contrast to the research showing strong relations between verbal working memory and language skills in children with CIs, measures of visuospatial working memory are typically not related to language skills in CI samples (Lyxell et al., 2008; Wass et al., 2008). However, investigations of visuospatial working memory and language skills are scarce, and more research is needed in this area.

Almost no research has investigated associations between other areas of EF, such as fluency-speed and inhibition-concentration, and spoken language skills in children with CIs. One study using a delay measure of inhibition found significant relations with vocabulary, language, and speech intelligibility, but measures of attention and concentration were unrelated to spoken language outcomes in that study (Horn, Davisa, Pisoni, & Miyamoto, 2004). There has been almost no research on domain-general EF fluency-speed and spoken language outcomes in children with CIs, although some studies have investigated verbal processing speed such as rapid automatized naming speed and response times to verbal stimuli. The results of these studies have been equivocal, with some research indicating strong relations between verbal processing speed and language outcomes (Pisoni et al., 2011) and others finding either no association or inconsistent associations (Nittrouer et al., 2013; Nittrouer et al., 2012). In summary, little is currently known about associations between domain-general EF inhibition-concentration and fluency-speed in children with CIs, despite evidence that these factors are important in language development in NH children (Henry et al., 2012).

Because there are substantial differences in the development of speech-language and EF skills between children with CIs and NH children (Kronenberger et al., 2013), it cannot be assumed a priori that EF and speech-language skills are related in the same way in these two populations. Furthermore, published research on the development of EF and speech-language skills in children with CIs is sparse and has been limited to verbal working memory or global EF measures (Figueras et al., 2008; Pisoni et al., 2011). EF may play an important role in the development of speech and language functioning in children with CIs, offering the potential to better understand and intervene to maximize speech and language development over and above advances in CI technology. Conversely, because speech-language skills are foundational building blocks for the development of EF, delays in the development of spoken language skills in children with CIs may contribute to delays in EF.

The present study is the first investigation of associations between a broad set of EF domains, including working memory, fluency-speed, and inhibition-concentration, and speech-language skills in a sample of prelingually deaf, early-implanted children, adolescents, and young adults following long-term use of CIs, compared to a NH sample. We sought to address two primary questions in this study:

1. What is the association between core EF domains and speech-language skills in long-term CI users? Based on earlier research findings (Figueras et al., 2008; Pisoni et al., 2011), we predicted that verbal working memory and inhibition-concentration would be related to measures of speech perception and spoken language processing.
2. Do associations between EF domains and language skills differ in CI users compared to NH peers? Verbal working memory and fluency-speed are likely to be particularly important in language learning for CI users, who may need greater processing capacity or faster processing speed in order to accommodate the increased cognitive load and greater risk of perceptual errors resulting from degraded auditory information provided by the CI and underspecified phonological and lexical representations of spoken language (Luce, Feustel, & Pisoni, 1983; Pisoni et al., 2010). Furthermore, CI samples show deficits in verbal working memory and fluency-speed relative to NH samples (Kronenberger et al., 2013), and prior research has found strong relations between measures of verbal working memory capacity and speech-language skills in CI users (Pisoni et al., 2011; Cleary et al., 2000; Geers et al., 2013; Nittrouer et al., 2013; Wass et al., 2008). Based on these findings, we hypothesized that verbal working memory and fluency-speed would be more strongly related to language skills in long-term CI users than in NH peers.

Method

Participants

Participants were 64 CI users and 74 NH peers aged 7–27 years who did not differ significantly in age ($t(136) = 1.17$, $p = 0.243$), family income ($t(122) = 0.15$, $p = 0.883$), or nonverbal intelligence ($t(136) = 0.94$, $p = 0.348$); the NH group had slightly more females than the CI group (Fisher's exact $p < 0.04$) (Table 1). CI participants were recruited from a large university hospital-based CI clinic as well as local advertisements; NH participants were recruited using advertisements posted in the same locations.

Inclusionary criteria for the CI sample were (1) severe-to-profound hearing loss prior to age 3 years; (2) pediatric cochlear implantation prior to age 7 years;

Table 1 Sample characteristics

	CI sample (*N* = 64)		NH Sample (*N* = 74)	
	Mean (SD)	Range	Mean (SD)	Range
Demographics and hearing history				
Chronological age (years)	15.0 (4.9)	7.8–27.4	16.0 (4.9)	7.1–25.3
Age at implantation (months)	35.6 (19.6)	8.3–75.8	NA	NA
Duration of CI use (years)	12.1 (3.9)	7.1–22.4	NA	NA
Age of onset of deafness (months)	2.9 (7.6)	0–36	NA	NA
Preimplant residual hearing (PTA)[a]	107.5 (11.1)	85–118.4	NA	NA
Communication mode[b]	4.6 (1.0)	1–5	NA	NA
Income level[c]	7.1 (2.5)	2–10	7.0 (2.6)	1–10
Nonverbal intelligence[d]	55.2 (7.5)	32–68	56.3 (6.9)	38–70
Sex (female/male)	27/37		45/29	
Speech and language domains[e]				
Language	−0.53 (1.00)	−2.66 to 0.89	0.49 (0.52)	−0.75 to 1.56
Speech perception	0.17 (0.70)	−1.73 to 0.90	NA	NA

Note. CI = cochlear implant; NH = normal hearing; SD = standard deviation.
[a]PTA = preimplant unaided pure-tone average for frequencies 500, 1000, 2000 Hz in decibels hearing loss.
[b]Communication mode is coded mostly sign (1) to auditory-verbal (6) (Geers & Brenner, 2003)
[c]Income is coded on a 1 (under $5,500) to 10 ($95,000 and over) scale (Kronenberger et al., 2013).
[d]Nonverbal intelligence is T-score of the Wechsler Abbreviated Scale of Intelligence (Wechsler, 1999), Matrix Reasoning subscale, based on national norms.
[e]Domain scores for speech and language are mean z-scores for constituent measures.

(3) use of a CI for 7 years or more; (4) use of a modern multichannel CI system; and (5) living in a household with English as the primary language. Inclusionary criteria for NH participants were (1) normal hearing (required to pass a basic hearing screening) and (2) age 7–25 years. Potential CI or NH participants were excluded if any additional developmental or cognitive delays were documented in the medical chart or by parent-report.

The majority of participants in the CI sample (N=54/64, 84%) were deaf at birth; therefore, duration of deafness and age at cochlear implantation were very highly correlated ($r=0.92$, $p<0.001$). To eliminate redundancy in analyses, duration of deafness is not analyzed further, since it is essentially identical to age at cochlear implantation. Seven participants developed profound deafness after birth as a result of meningitis; other etiologies for hearing loss in the CI sample were auditory neuropathy ($N = 3$), large vestibular aqueduct ($N = 1$), Mondini malformation ($N = 3$), familial (another immediate family member was deaf, of unknown etiology; $N = 9$), ototoxicity ($N = 1$), and unknown ($N = 40$). Consistent with FDA requirements for pediatric CI candidacy, average preimplantation residual hearing (pure tone average, PTA; average decibels hearing loss for frequencies 500, 1,000, and 2,000 Hz) was in the profound range for hearing loss (Table 1). Three participants were implanted below age 12 months; 20 participants were implanted between age 12 and 23 months; 17 participants were implanted between age 24 and 35 months; 12 participants were implanted between ages 36 and 59 months; and the remaining 12 participants were implanted between ages 60 and 76 months. Participants had used their CIs for, on average, 12.1 years, and about 1/3 of participants had bilateral CIs ($N = 21$) compared to about 2/3 with unilateral CIs ($N = 40$; the remaining 3 participants used a CI in one ear and hearing aid in the other ear). All participants reported using their CIs regularly, and almost all participants ($N = 59/64$; 92%) communicated using an auditory-oral or speech-emphasis mode of communication at school and/or in their primary living and working environments, as coded by a standard communication mode rating scale (Geers & Brenner, 2003) (Table 1).

Procedures

Study procedures were approved by the university institutional review board, and written consent (and assent if appropriate) was obtained prior to initiation of any study procedures. Data for this study were obtained as a part of a larger study of speech, language, and EF skills in children, adolescents, and adults with CIs (Kronenberger et al., 2013). All participants completed speech and language measures in one test session and neurocognitive tests at a second test session within the next 6 months (95% of the sample completed both sessions in a 30 day period; mean = 9.4 days, SD = 20.4 days). Language tests were administered in the current mode of communication used at school or (for those not in school) in the participant's preferred mode of communication (oral vs. total communication). Speech perception tests were presented at 65 decibels in a sound field in a sound-treated audio booth at 0^{o} azimuth approximately 3 feet from the participant using a high-quality loudspeaker. Participants were paid $20 per hour plus travel expenses.

Three EF domains were assessed based on prior neuropsychological research with NH children (Gioia et al., 2000; McAuley & White, 2011) and recent findings that these domains are atypical and at-risk in CI users (Conway et al., 2009; Kronenberger et al., 2013): (1) working memory; (2) fluency-speed; and (3) inhibition-concentration. A measure of nonverbal intelligence was also obtained in order to account for potential confounds related to intellectual ability and fluid reasoning skills. In order to reduce potential confounds associated with audibility of stimuli, all EF measures selected for this study had minimal auditory processing requirements and well-established psychometrics in both NH and hearing-impaired populations. Only the measures of verbal working memory involved verbally mediated stimuli and required spoken responses, and only the Digit Span test required auditory perception of spoken test stimuli. All of the EF measures of visual working memory capacity, fluency-speed, and inhibition-concentration used only visual stimuli that were minimally verbally mediated (e.g., in most cases, stimuli were symbols) and that required no spoken verbal response, in order to control for the effects of audibility, verbal, language, and speech production for response output.

All speech perception, language, EF, and nonverbal intelligence tests were administered according to standardized procedures, and (with the exception of the speech perception tests) provided norm-based scores based on nationally representative samples of NH, typically developing children. For language, EF, and nonverbal intelligence tests, norm-based scores were used for all analyses reported in this study. In cases where the participants' ages were out of the norm range (which was the case for the WISC-IV Coding and Coding Copy subtests for participants aged 17 years and older), norms for the oldest available age were used. Norm tables for the WISC-IV (Wechsler et al., 2004) demonstrate that norm scores level off near the upper age ranges for these subtests, supporting the use of the oldest age norms with participants who are above the norm range for these tests. Furthermore, correlations of all EF domain scores with chronological age were nonsignificant in this study (see Results section for correlations between age and EF domain scores), showing that this strategy for obtaining norm-based scores for older individuals on the WISC-IV did not influence results. For the speech perception tests, raw scores were used because national norms are not available for any of these clinical tests. Spoken directions and instructions were supplemented when necessary with nonverbal demonstrations, examples, and practice in order to ensure that all participants fully understood the tasks prior to completing the assessments. In some cases, participants were unable to complete tests as a result of failure to understand or sufficiently adhere to instructions; for measures not completed by all participants, N is reported in the Measures section.

Measures

Measures of EF and nonverbal intellectual ability. *Working memory* capacity was assessed using five immediate memory span tests: The *Digit Span Forward* ($N = 63$ CI, 74 NH) and *Digit Span Backward* ($N = 63$ CI, 73 NH) subtests from the Wechsler Intelligence Scale for Children, Third Edition (WISC-III) (Wechsler, 1991), and the *Visual Digit Span* subtest from the Wechsler Intelligence Scale for Children, Fourth Edition, Integrated (WISC-IV-I)(Wechsler et al., 2004), measure immediate memory capacity for sequences of digits that are either spoken using live voice (Digit Span Forward or Backward) or presented visually in printed format (Visual Digit Span) to the participant. Sequences must be repeated in either forward (Digit Span Forward and Visual Digit Span) or backward (Digit Span Backward) order. The *Spatial Span Forward and Backward* ($N = 63$ CI, 74 NH) subtests from the WISC-IV-I (Wechsler et al., 2004) assess memory span for sequences of spatial locations. The examiner points sequentially to individual blocks on a board, and the participant then reproduces (by pointing) the same sequence of blocks in either forward or backward order. *Fluency-speed* skills were assessed with three subtests measuring different areas of processing speed during tasks that required focused, sustained attention: identification and cancellation of visual stimuli (*Pair Cancellation* subtest of the Woodcock–Johnson Tests of Cognitive Ability, Third Edition; WJ-III (Woodcock, McGrew, & Mather, 2001)), association and reproduction of visual stimuli (*Coding* subtest from the WISC-IV-I (Wechsler et al., 2004)), and visual-motor copying (*Coding Copy* subtest from the WISC-IV-I (Wechsler et al., 2004)). *Inhibition-concentration* abilities were measured using three scores from the *Test of Variables of Attention* (a continuous performance test that requires participants to press a button when a square appears at the top of a computer screen or to not respond when the square is presented at the bottom of the computer screen (Leark, Dupuy, Greenberg, Corman, & Kindschi, 1996); $N = 61$ CI, 72 NH): Response time variability (variability of reaction times to stimuli across all responses), Omission errors (failing to respond to a target stimulus), and Commission errors (responding to a nontarget stimulus). *Nonverbal intelligence* was assessed using the *Matrix Reasoning* subtest from the Wechsler Abbreviated Scale of Intelligence (WASI) (Wechsler, 1999), which required participants to complete a pattern of visual geometric designs based on an underlying concept. All measures of EF used in this study are well established, are widely used in clinical and research settings, and have strong psychometrics, including excellent internal consistency, test-retest reliability, and construct validity.

Measures of speech and language skills. Speech perception skills (obtained for the CI sample only, since NH children routinely score at ceiling levels on these measures when administered under quiet conditions) were evaluated using a measure of isolated word recognition (total score on the Lexical Neighborhood Test (LNT) (Kirk, Pisoni, & Osberger, 1995); $N = 63$) and three sentence recognition scores from two tests (Auditory-Visual Lexical Neighborhood Sentence Test [AVLNST], Kirk et al., 2007; Auditory-Only scores, $N = 62$; and Hearing in Noise Test for Children [HINT-C], Nilsson, Soli, & Gelnett, 1996, sentences presented in quiet, $N = 62$ and in speech-shaped noise at +5 decibels signal-to-noise ratio, $N = 57$). For these speech perception measures, participants repeat individual words (LNT) or sentences (AVLNST and HINT-C) that are presented in audio-only format without visual lipreading cues. The LNT, AVLNST, and HINT-C have been extensively used in studies of deaf children with CIs, and their validity is well established. Receptive *vocabulary* was assessed with the *Peabody Picture Vocabulary Test, Fourth Edition* (PPVT-4) (Dunn & Dunn, 2007), which requires the participant to identify a picture that corresponds to a vocabulary word that is spoken or signed by the examiner. Core *language* processing skills were evaluated with the *Clinical Evaluation of Language Fundamentals, Fourth Edition* (CELF-4) (Semel, Wiig, & Secord, 2003), Core Language Score ($N = 59$ CI, 74 NH), which is a composite score based on several subtests measuring word knowledge, conceptual understanding of words and sentences, sentence recall/repetition skills, and generation of sentences in spoken language. The PPVT-4 and CELF-4 have excellent psychometrics and are widely used in the field of language development.

Statistical Approach and Analysis

In order to empirically validate the EF and speech-language domains described above, test scores were subjected to two separate principal components analyses (PCAs): One PCA was carried out on the 11 EF scores, and a second PCA was carried out on the 6-speech perception and language test scores. PCA is a statistical method used to identify groups of variables that share variance, indicating that they are statistically related. PCA was also used in this study to reduce the number of variables in subsequent analyses by creating composite domain scores. For each PCA, the number of components selected for rotation (Promax rotation) was based on scree plot inspection and the eigenvalue > 1 convention. A composite score for each component was then derived by calculating the mean of all test scores with loadings > 0.60 on that component. Z-transformed scores, based on the mean and standard deviation of the entire sample, were used to calculate these composite scores. Similar methods for obtaining domain scores (e.g., PCA and/or summed z-scores) have been used successfully in other studies of executive and speech-language functioning of children with CIs (Figueras et al., 2008; Geers, Brenner, & Davidson, 2003).

Following the derivation of the composite domain scores, correlations between the domain scores for EF, speech perception, and language functioning were calculated separately for the CI and NH samples. Finally, a regression analysis was carried out to predict language domain skills based on hearing status (CI vs. NH) and EF domain scores, controlling for baseline nonverbal intelligence. Interaction terms between hearing status and each of the EF domain scores were tested to evaluate whether the relations between EF and language were moderated by hearing status (i.e., whether EF scores are differentially related to language for the CI sample compared to NH peers). Because speech perception scores were obtained only for the CI sample (NH children routinely score at ceiling levels on these measures when the test signals are presented in the quiet), regression equations were not calculated for the speech perception scores, and correlations were not computed for speech perception in the NH sample.

Results

Executive Functioning and Speech-Language Domains

The PCA of the 11 EF measures yielded four components with eigenvalues > 1: Verbal Working Memory, Fluency-Speed, Inhibition-Concentration, and Spatial Working Memory (Table 2; other factor analytic methods were also applied to these scores, and they produced similar results. Scale loadings on EF domains were consistent

Table 2 Principal components analysis (PCA) of executive functioning measures

	Component/domain			
	1	2	3	4
Measure	Verbal WM	Fluency-speed	Concentration	Spatial WM
PCA loadings				
Digit Span Forward	**0.82**	0.03	−0.14	0.18
Digit Span Backward	**0.63**	0.18	0.05	−0.02
Visual Digit Span	**0.85**	0.07	−0.06	−0.07
Spatial Span Forward	0.12	0.00	−0.07	**0.83**
Spatial Span Backward	−0.06	0.03	0.15	**0.78**
Coding	0.09	**0.81**	0.13	0.00
Coding Copy	0.19	**0.83**	0.03	−0.15
Pair Cancellation	−0.10	**0.76**	−0.08	0.16
TOVA response time variability	−0.18	0.19	**0.91**	0.01
TOVA Commissions	0.53	−0.28	0.46	−0.05
TOVA Omissions	0.04	−0.07	**0.87**	0.07
Executive functioning domain scores				
CI sample—mean (SD)	−0.48 (0.71)	−0.23 (0.85)	−0.31 (0.97)	−0.15 (0.86)
NH sample—mean (SD)	0.41 (0.63)	0.20 (0.77)	0.26 (0.77)	0.12 (0.81)
t (df)	7.75 (134)***	3.13 (136)**	3.81 (131)***	1.90 (135)

Note. CI = cochlear implant; TOVA = Test of Variables of Attention; WM = working memory; SD = standard deviation. PCA loadings are from pattern matrix following promax rotation of components with eigenvalue > 1. Eigenvalues for the first five components were 3.84, 1.68, 1.15, 1.07, and 0.74. PCA loadings > 0.60 are indicated in bold. Executive functioning domain scores are the mean z-transformed scores of measures with loadings > 0.60. T-tests compare CI and NH sample on executive functioning domain scores. ****p* < .001; ***p* < .01.

with the a priori conceptual groupings of the EF measures, with two exceptions: The working memory measures fell into two separate modality-specific components (verbal vs. spatial) and the Test of Variables of Attention (TOVA) Commissions score showed moderate loadings on two components (Inhibition-Concentration and Verbal Working Memory) but did not have a single distinct, high loading on only one component. As a result, separate domain scores were created for verbal working memory and spatial working memory, and the TOVA Commissions score was dropped from further analysis.

For the 6-speech perception, vocabulary, and language measures, the PCA produced two components with eigenvalues > 1 (eigenvalues of the first three components were 3.7, 1.4, and 0.4). Consistent with our expectations, the first component (speech perception) had high loadings (>0.82) for the four speech perception scores (from the Lexical Neighborhood Test, Auditory-Visual Lexical Neighborhood Sentence Test, and Hearing In Noise Test for Children in Quiet and in Noise). The second component (language) had high loadings (>0.94) for the receptive vocabulary (PPVT-4) and core language processing (CELF-4) scores.

Composite scores for components reflecting the EF domains (verbal working memory, spatial working memory, fluency-speed, and inhibition-concentration) and the speech-language domains (speech perception and language) were then created by taking the mean of the z-transformed scores (based on the total sample means and standard deviations) of the tests with high loadings (>0.60) on components for those domains. The CI group scored lower than the NH group on three of the four EF composite domain scores (verbal working memory, fluency-speed, and inhibition-concentration; Table 2), as well as the language domain score ($t(131) = 7.60$, $p < 0.001$; Table 1). The two groups did not differ significantly on spatial working memory ($p = 0.06$; Table 2).

Demographic and hearing history variables were found to be unrelated to EF domain scores in both the CI and NH groups. In the CI sample, EF domain scores were unrelated to chronological age, age at implantation, years of CI use, onset age of deafness, preimplantation residual hearing, communication mode, and family income (all $r < 0.26$, all $p > 0.05$). Similarly, chronological age and family income were unrelated to all EF domain scores in the NH sample (all $r < 0.18$, all $p > 0.05$). For the speech perception domain in the CI sample, however, participants who were younger ($r = -0.28$, $p = 0.04$), who had better residual hearing prior to implantation (r = -0.34, p = 0.01), who had higher auditory-oral communication mode scores (r = 0.47, p< 0.001), and who came from higher income families (r = 0.32, p = 0.02) had better speech perception skills. For both the CI and NH participants, higher family income was associated with better language skills (r's = 0.42 and 0.54, respectively; p< 0.01). No other demographic or hearing history variables were significantly related to the speech perception or language domain scores.

Relations Between Executive Functioning, Speech, and Language

The CI and NH samples showed different patterns of correlations between the EF domain scores and the speech and language domain scores (Table 3). For CI users, verbal working memory capacity and fluency-speed were significantly related to both the speech perception and language composite scores. In contrast, fluency-speed was not significantly related to language skills in the NH sample, and the verbal working memory-language correlation was lower in the NH sample than in the CI sample (Table 3). Additionally, spatial working memory and inhibition-concentration were unrelated to speech or language skills in the CI sample but were significantly related to language skills in the NH sample. Partial correlations statistically controlling for nonverbal intelligence produced essentially the same results as bivariate zero-order correlations (Table 3), establishing that the association between EF and speech-language skills was not a result of shared variance with fluid, nonverbal IQ.

In the regression analysis, nonverbal intelligence, hearing status, EF domain scores, and hearing status by EF interactions accounted for over 60% of the variance in the composite language scores (F(10,116) = 19.708, p< 0.001; Table 4). Better language skills were significantly predicted by higher nonverbal intelligence, normal hearing status, and larger verbal working memory capacity. Significant interactions were also found between working memory capacity (both verbal and spatial) and hearing status. Figure 1 displays this finding using separate median splits for verbal and spatial working memory capacity within both samples (CI and NH) to obtain high versus low working memory groups. The mean language domain score was plotted for each group. CI users with high verbal working memory capacity showed much better language scores than CI users with low verbal working memory capacity. The association between spatial working memory and the language composite score was much weaker for CI users. In contrast, for the NH sample, both verbal and spatial working memory showed nearly identical relations with the language score.

Discussion

As hypothesized, specific domains of EF were related to speech perception and language skills in long-term CI users, and these domains differed in several important ways from EF domains that were related to language skills in NH peers. For the CI sample, verbal working memory capacity and fluency-speed were significantly related to language and speech perception skills, whereas spatial working memory and

Table 3 Association of executive functioning and speech-language skills

Executive functioning domains	Language domain		Speech perception domain
	CI (*N* = 59)	NH (*N* = 74)	CI (*N* = 57)
Verbal working memory	0.64***	0.47***	0.30*
(*N* = 63 CI, 73 NH)	0.57***	0.30*	0.27*
Spatial working memory	0.14	0.40***	0.15
(*N* = 63 CI, 74 NH)	0.05	0.33**	0.12
Fluency-speed	0.32*	0.13	0.33*
(*N* = 64 CI, 74 NH)	0.26*	0.11	0.30*
Inhibition-concentration	0.07	0.39***	-0.04
(*N* = 61 CI, 72 NH)	0.01	0.25*	-0.06

Note. CI = cochlear implant; NH = normal hearing; SD = standard deviation. Top values are zero-order Pearson correlations; bottom values are partial correlations with nonverbal intelligence (WASI Matrix Reasoning score) controlled. *N*s for domain scores reflect number of participants completing all measures comprising the composite domain score.
**p* < .05; ***p* < .01; ****p* < .001.

Table 4 Regression predicting language domain score

Predictor	Beta	*t*	*p*
Nonverbal intelligence	**0.20**	**3.20**	**.002*** **
Hearing status: CI vs. NH	**0.25**	**3.40**	**.001*** **
Verbal working memory	**0.83**	**6.66**	**.001*** **
Spatial working memory	**-0.25**	**2.40**	**.018***
Fluency-speed	0.08	0.77	.442
Inhibition-concentration	0.08	0.86	.394
Hearing status × verbal WM	**-0.48**	**4.42**	**.001*** **
Hearing status × spatial WM	**0.27**	**2.84**	**.005** **
Hearing status × fluency-speed	-0.04	0.44	.662
Hearing status × inhibition-concentration	0.01	0.12	.908

Note. CI = cochlear implant; NH = normal hearing; WM = working memory. Nonverbal intelligence = Wechsler Abbreviated Scale of Intelligence Matrix Reasoning T-score. Values are coefficients and t-test results for regression equation predicting language domain score. For entire equation, $F(10,116) = 19.708$, $p < .001$; $R^2 = 0.63$. Regression coefficients for variables in bold font are statistically significant, $p < .05$.
*$p < .05$; **$p < .01$; ***$p < .001$.

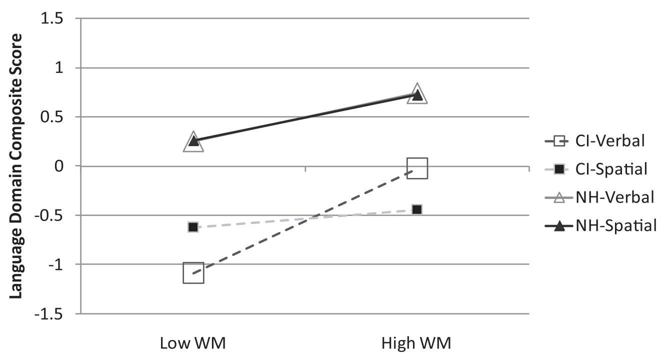

Figure 1 Mean language domain composite scores for high versus low verbal or spatial working memory (WM) subgroups in the cochlear implant (CI) and normal-hearing (NH) Samples. Language domain composite scores are the mean z-transformed scores for the PPVT-4 and CELF-4 Core Language scores. Lines represent the mean language domain score for the CI (square end markers) and NH (triangle end markers) samples for high or low (based on median split) verbal WM (unfilled end markers) and spatial WM (filled end markers).

inhibition-concentration were not. However, a different profile of associations between EF and language skills emerged for the NH sample, suggesting that the role of EF in language development (and, conversely, the role of language in EF) differs in important ways for early-implanted, long-term CI users when compared to NH peers.

The finding of significant relations between verbal working memory and speech perception in children and adolescents with CIs replicates earlier research findings (Pisoni et al., 2011; Cleary et al., 2000; Nittrouer et al., 2013) demonstrating that phonological working memory is especially important for rapidly encoding and processing degraded and underspecified speech signals transmitted to the auditory nerve by a CI. Similarly, the present findings are also consistent with prior research demonstrating strong relations between verbal working memory capacity and language skills in a CI sample (Cleary et al., 2000; Geers et al., 2013; Nittrouer et al., 2013; Wass et al., 2008). The finding of strong relations between verbal working memory and speech-language skills in CI users probably reflects a reciprocal effect: Individuals with faster and more efficient verbal working memory skills are able to encode, store, process, and retrieve more robust, highly-detailed phonological and lexical representations of speech signals, leading to improved language development. Conversely, stronger language skills make verbal encoding and working memory faster and more efficient (Hulme, Maughan, & Brown, 1991; Singer & Bashir, 1999; Ylvisaker & DeBonis, 2000).

This study is among the first to demonstrate that the association between language and verbal working memory is stronger for CI users than for NH peers.

Our results are consistent with other findings from studies showing stronger relations between verbal working memory and vocabulary in CI users compared to NH peers (Nittrouer et al., 2013). Furthermore, prior studies suggest that the association between verbal working memory and reading skills is stronger in CI than in NH samples (Lyxell et al., 2008; Nittrouer et al., 2012). Several recent studies using the MMN paradigm have also found a different pattern of associations between working memory and MMN activation in NH samples compared to CI samples (Ortmann et al., 2013; Watson, Titterington, Henry, & Toner, 2007). Thus, an emerging body of converging research suggests stronger associations between verbal working memory and language-related outcomes in children with CIs compared to their NH peers.

This study also provides new results showing that the association between language and spatial working memory is stronger for NH peers than CI users. Earlier research reported no differences between CI users and NH peers in spatial working memory abilities (Kronenberger et al., 2013; Lyxell et al., 2008; Wass et al., 2008). However, this is the first study to investigate differences in the association of language and spatial working memory in prelingually deaf, long-term CI users compared to NH peers.

Taken together, the constellation of findings obtained in this study suggests that language and working memory skills may interact in fundamentally different ways during development in children and adolescents with CIs compared to NH peers: Verbal working memory is an especially crucial and inseparable foundational component of language development following cochlear implantation, and the development of verbal working memory is strongly dependent on language ability throughout childhood. Because CI users who communicate in an auditory-oral or speech-emphasis modality (with little or no sign) (Geers & Brenner, 2003) depend very heavily on compromised underspecified phonological and lexical representations during verbal processing and language learning (Pisoni et al., 2011), the capacity demands on their active verbal working memory during routine spoken language processing are likely to be much greater than the verbal working memory demands for typically developing children with NH (Kronenberger, Pisoni, Henning, Colson, & Hazzard, 2011). These differences may underlie the differential associations found here between verbal working memory and language skills in early-implanted children and adolescents with CIs compared to NH peers.

Another novel finding in the present study was the association between nonverbal, domain-general fluency-speed and spoken language skills in the sample of long-term CI users. This result is theoretically significant because all of the fluency-speed measures in the present study were obtained using exclusively visual-nonverbal tasks, with no auditory or language processing demands. This pattern of results suggests that domain general (e.g., not limited to audibility or spoken language) information-processing speed is an important component of speech perception in CI users, likely because rapid, highly efficient cognitive operations are necessary to reliably encode and process highly degraded underspecified speech signals in real-time. Similarly, fluency-speed may also be important for the development of language skills by providing access to more language experiences and knowledge as well as more rapid and efficient processing of verbal information. Prior research has demonstrated that verbal processing speed skills, such as verbal rehearsal speed and short-term memory scanning speed, are strongly related to speech and language development in children with CIs (Burkholder & Pisoni, 2003; Pisoni & Cleary, 2003; Pisoni et al., 2011), although other research has found less consistent associations between some types of verbal processing speed and language-related skills using different experimental paradigms (Nittrouer et al., 2013; Nittrouer et al., 2012). This study extends the earlier findings on verbal processing speed by showing that domain-general, nonverbal fluency-speed abilities are also related to speech and language processing operations in children with CIs.

Importantly, fluency-speed was not significantly correlated with language skills in the NH sample, although the lack of a significant interaction between hearing status and fluency-speed in regressions predicting language outcomes (Table 4) suggests that this finding should be interpreted with caution. The dissociation between CI and NH samples in the pattern of correlations between fluency-speed and speech-language skills could suggest that fluency-speed may be more important for spoken language development in children with CIs than in NH peers. Because spoken language processing skills are much faster and more highly automatized in NH samples, the contribution of fundamental, domain-general fluency-speed to language development may be less significant for NH children. On the other hand, spoken language skills place a considerably higher information-processing load on CI users, as a result of degraded auditory input, limitations on audibility and hearing in noise, and underspecified phonological and lexical representations. As a result, individual differences in fluency-speed and the time-course of perceptual processing of speech and spoken language may be uniquely important for language development in CI users.

Contrary to expectations, only the NH sample demonstrated a significant association between language skills and measures of inhibition-concentration skills. Despite a near-zero correlation between inhibition-concentration and language scores in the CI sample, the nonsignificant interaction between hearing status and inhibition-concentration in regression equations predicting language outcomes suggests that additional research is needed to better understand this finding. It is possible that this result could suggest that the ability to sustain attention and concentration is less important for language development in children with CIs than it is for NH peers or that CI users are less dependent on language processing for controlling inhibition and concentration. For example, it is possible that the additional working memory demands of spoken language processing for children with CIs cause an early saturation of their verbal processing capacity. This early saturation of processing capacity would limit the value of further sustaining controlled attention and vigilance after the capacity limit is reached. For NH children, on the other hand, greater capacity and fluency-speed of processing and coding large amounts of phonological and lexical information would produce a greater value of sustaining concentration over longer periods of time in order to have full access to linguistic experience and knowledge. Alternatively, NH children might actively recruit spoken language skills more consistently to engage in inhibitory behaviors, whereas CI users might rely more on abilities that are not language-mediated for the development of inhibitory and controlled behavior.

It is important to note that the method of measuring inhibition-concentration in this study using a visual continuous performance test may have influenced results. In contrast to our current findings, other research using delay-based measures of inhibition has found relations with language in samples of children with CIs. On the other hand, traditional verbally mediated CPT tests have not found such associations, which is consistent with our results (Horn et al., 2004). Additional research with a broader set of measures of inhibition-concentration is recommended to better understand the association between this domain of EF and spoken language outcomes in CI users.

In the regression equations, relations between speech-language skills and fluency-speed for children with CIs and between language skills and inhibition-concentration for NH children were no longer statistically significant. This does not appear to be due to shared variance with nonverbal intelligence, because those relations remained statistically significant in partial correlations when nonverbal intelligence was statistically controlled. Because regression coefficients reflect only unique variance after accounting for other predictors in the regression equation, it is possible that relations between fluency-speed (for children with CIs), inhibition-concentration (for NH children), and language skills share variance with working memory, which was related to language skills in both the CI and NH samples. Future research with larger sample sizes will be necessary to better understand the elementary core processing mechanisms, mediators, and moderators of the significant, differential relations observed between fluency-speed, inhibition-concentration, and language skills in CI versus NH samples.

Limitations of this study include its cross-sectional, correlational design, which only allows for identification of associations but cannot determine direction of causality. In fact, it is most likely that the association between EF and language skills is reciprocal and bidirectional in nature. Future studies using longitudinal and experimental designs should be undertaken to address these questions of causal direction (Kronenberger & Pisoni, 2014). Additionally, larger and more diverse samples of CI users should be assessed in future studies; this project studied long-term CI users who were prelingually deaf and implanted during early childhood. Larger samples would also allow for the investigation of EF and speech-language relations at different ages and developmental stages. Although chronological age was unrelated to any of the measures of EF in this study, the study sample had a broad age range encompassing a wide range of development. Future research should also investigate the generalization of these results to other populations with hearing loss, including children with hearing aids and users of American Sign Language. Given the high prevalence of hearing loss in the pediatric population (Mehra et al., 2009) and the importance of early identification and intervention (Bhatia et al., 2013), a significant public health need exists to better understand the neurocognitive foundations of speech and language delays in the large subgroup of children with hearing loss, particularly if these core underlying neurocognitive processes differ significantly from those observed in normal-hearing children, as we found in the present study for children with hearing loss who use CIs.

Clinically, the results obtained in this study have several important implications not only for the CI population but potentially for other populations with hearing loss. While verbal working memory has been previously shown to be a core foundational component of language development of children with CIs, this is the first study to demonstrate that domain-general fluency-speed skills are additional critical information-processing components of speech-language development in this clinical population. Another important finding of this study is the differential pattern of correlations among the EF measures and language in the two groups. We found that not only is language development delayed in CI users but that the basic reciprocal influences of language skills and EF may have differential developmental effects in CI users compared to NH peers. Our results suggest that understanding variability in language development following cochlear implantation and creating novel interventions to improve language skills in children with CIs cannot be inferred exclusively from what is currently known about the development of language in children with NH, but must be tailored to specific and unique factors in speech, language, and EF development following hearing loss and cochlear implantation. Conventional audiological and speech-language outcome measures routinely used to assess children with CIs who may be at greater risk for language delay and impairment should be broadened substantially to include EF measures, particularly assessments of verbal working memory capacity and fluency/speed. Verbal working memory skills and fluency-speed may also be efficacious targets for novel, high-yield interventions designed specifically for children with CIs, especially for children who fail to display optimal spoken language benefits from their CI after several years of use in linguistically rich stimulating environments (Kronenberger et al., 2011).

Funding

This research was supported by grant R01 DC009581 from the National Institute on Deafness and Other Communication Disorders to DBP.

Conflicts of Interest

No conflicts of interest were reported.

Acknowledgments

The authors thank Terri Kerr, Allison Ditmars, Allison Woody, and Sami Gharbi for their help with this study.

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