Executive Functioning Skills in Long-Term Users of Cochlear Implants: A Case Control Study

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Objective To investigate differences in executive functioning between deaf children with cochlear implants (CIs) and normal-hearing (NH) peers. The cognitive effects of auditory deprivation in childhood may extend beyond speech—language skills to more domain-general areas including executive functioning. **Methods** Executive functioning skills in a sample of 53 prelingually deaf children, adolescents, and young adults who received CIs prior to age 7 years and who had used their CIs for ≥7 years were compared with ageand nonverbal IQ-matched NH peers and with scale norms. **Results** Despite having above average nonverbal IQ, the CI sample scored lower than the NH sample and test norms on several measures of short-term/ working memory, fluency—speed, and inhibition—concentration. Executive functioning was unrelated to most demographic and hearing history characteristics. **Conclusions** Prelingual deafness and long-term use of CIs was associated with increased risk of weaknesses in executive functioning.

Key words cognitive assessment; deafness and hearing loss; neuropsychology.

Cochlear implantation restores some attributes of hearing to many deaf children, allowing them to develop substantial receptive and expressive spoken language skills (Geers & Sedey, 2011). However, even after implantation at very young ages, children with cochlear implants (CIs) have already experienced a period of profound deafness during critical early periods of brain development, with degraded auditory experience following implantation. Furthermore, despite marked development of speech perception and spoken language skills in most children with CIs, many children consistently lag behind age peers in some areas of speech and language skills (Geers, Nicholas, & Sedey, 2003). Because early auditory experience and verbal skills are a part of a larger functionally integrated system of neurocognitive processes (Geers & Moog, 1987; Luria,

1973), language delays and/or compromised early auditory experiences may affect other neurocognitive abilities (Pisoni et al., 2008; Pisoni, Conway, Kronenberger, Henning, & Anaya, 2010). For example, auditory–verbal experiences and skills have been implicated in the development of executive functioning (processes used for directing and controlling thought and behavior) because they provide tools and experiences (such as language and phonological representations of speech–sound sequences) that are used in developing self-regulation, verbal mediation, working memory, and sequential processing strategies (Barkley, 1997; Conway, Pisoni, & Kronenberger, 2009; Luria, 1973).

In support of this hypothesis, prior research suggests that children with CIs, on average, have more difficulty than normal-hearing (NH) children in several areas of executive functioning. For example, Figueras, Edwards, and Langdon (2008) found that deaf and hearing-impaired children with CIs or hearing aids scored lower than NH children on neuropsychological measures of executive functioning, including inhibition, planning, set-shifting, working memory, and some types of attention. In another study using a standardized parent-report behavior checklist of executive functioning behaviors completed by parents of adolescents with CIs, Beer, Pisoni, Kronenberger, and Geers (2010) found elevated sample mean scores relative to scale norms (indicating more problems) on three composite executive functioning measures as well as several clinical subscales. A substantial amount of research also demonstrates that children with CIs score below age norms on measures of auditory-verbal short-term and working memory capacity, which are also considered to be core foundational components of executive functioning and cognitive control (Pisoni et al., 2008; Pisoni, Kronenberger, Roman, & Geers, 2011).

Executive functioning abilities are critically important in the development of language, behavioral control, and adaptive functioning skills, including academic and social skills (Barkley, 1997; Pisoni et al., 2010). Deficits in verbal working memory capacity, for example, are related to language and learning problems in NH children (Pickering & Gathercole, 2004) as well as in deaf children with CIs (Pisoni et al., 2008). Additionally, deficits in the ability to inhibit behavior and engage in planning prior to action are associated with a host of adaptive functioning delays and psychosocial risks (Barkley, 1997). Thus, a growing body of evidence suggests that a subset of children with CIs may be at risk for deficits in executive functioning skills that are critically important for development and quality of life, including the development of robust receptive and expressive spoken language skills.

Although some limited research has investigated executive functioning in children with CIs, little is currently known about the long-term development of executive functioning in older children, adolescents, and young adults with prelingual deafness who received CIs in early child-hood and have used their CIs for long periods of time. Pisoni et al. (2011) investigated the development of verbal short-term and working memory using a digit span test in a sample of 112 prelingually deaf adolescents after >10 years of CI use. Although the sample improved in verbal short-term/working memory capacity and verbal rehearsal speed over the 8-year study period, these children continued to score well below average relative to scale norms. Moreover, verbal short-term memory scores were found to be related to a broad set of speech and language

outcomes, whereas verbal working memory scores were related only to higher-order language functioning. The results of the Pisoni et al. (2011) long-term outcome study indicated continued delays in verbal short-term and working memory into adolescence and showed that short-term/ working memory skills are closely linked to speech and language outcomes. Although that study provided important information about long-term working memory outcomes, it was narrowly focused on only one area of longterm executive functioning outcomes (verbal short-term/ working memory capacity), and it did not include a matched control sample. No research to date has comprehensively assessed long-term executive functioning outcomes in a sample of prelingually deaf children who received CIs in early childhood, nor have any previous studies of executive functioning in children with CIs used rigorously matched NH control samples.

Investigation of long-term outcomes of CIs is critically important for several reasons: First, the effectiveness of any treatment is based not only on short-term progress but also on long-term developmental endpoints. Second, because cochlear implantation in early childhood was not widely performed prior to the 1990s, the first cohorts of long-term childhood CI users are only now becoming available for study. Third, assessment of long-term outcomes allows the investigation of whether deaf children with CIs "catch up" in adulthood in core neurocognitive areas that are found to be delayed earlier in development.

Few studies have been conducted with samples consisting exclusively of long-term CI users who were implanted in early childhood, and these studies have focused more on speech-language and functional outcomes than on executive functioning (Geers & Sedey, 2011; Ruffin, Kronenberger, Colson, Henning, & Pisoni, 2013; Uziel et al., 2007). Results of these studies have demonstrated sustained improvement in speech perception and language skills even after ≥10-15 years of CI use. A majority of the long-term CI users scored in the average range or higher (>1 standard deviation below the normative mean score) on standardized measures of language, although the mean language score for CI samples fell below that of norm samples (Geers & Sedey, 2011; Ruffin et al., 2013; Uziel et al., 2007). Academically, adolescents who used CIs for ≥10 years have shown performance on written expression and phonological processing tasks that is well below that of NH peers. On the other hand, performance of long-term CI users has been found to be somewhat stronger on measures of reading comprehension and word recognition, with half or more of the CI sample falling in the average range or higher (Geers & Hayes, 2011). Moog, Geers, Gustus, and Brenner (2011) reported generally good psychosocial adjustment in long-term CI users, with almost universal participation in high school activities (including sports) and a rate of part-time work similar to that of NH adolescents. However, little is known of the development of executive functioning skills in long-term users of CIs.

Because of the importance of executive functioning skills for core foundational areas of verbal, cognitive, behavioral, and emotional development following cochlear implantation, it is critically important to understand the long-term developmental course of this functioning in older children, adolescents, and young adults with prelingual deafness who received CIs in early childhood. In this study, we investigated long-term executive functioning outcomes in a sample of prelingually deaf individuals who received CIs prior to age 7 years and who had used their CIs for ≥7 years, compared with a 1:1 age-IQmatched group of NH children. Specifically, the purpose of this research was to determine (1) what types of executive functioning delays (relative to the NH population, using both matched controls and scale norms as benchmarks), if any, continue to exist in children, adolescents, and young adults who have used their CIs for >7 years, and (2) the extent to which long-term development of executive functioning skills is associated with conventional demographic and hearing history variables such as age at implantation and length of CI use.

We sought to obtain a broad assessment of executive functioning skills by evaluating three areas of executive functioning that have been identified as being potentially at-risk in children with CIs (Conway et al., 2009; Pisoni et al., 2010): (1) short-term/working memory capacity, (2) inhibition—concentration, and (3) fluency—speed. Although there is no universal agreement on a single definition of executive functioning, neuropsychological and neuroimaging studies support the relationships between these three areas as important in executive functioning (Barkley, 1997; Gioia, Isquith, Guy, & Kenworthy, 2000; McAuley & White, 2011).

Method *Participants*

CI Sample

Participants in the CI sample were 53 children, adolescents, and young adults (see Table I for demographic and hearing history characteristics). Inclusionary criteria were as follows: (1) severe to profound hearing loss (>70 dB hearing loss in the better hearing ear) prior to age 3 years; (2) cochlear implantation prior to age 7 years; (3) use of a CI for ≥7 years; (4) use of a currently

available state-of-the-art multichannel CI system; and (5) living in a household with English as the primary language. Potential participants were excluded if (1) any additional developmental or cognitive delays other than hearing loss were reported in the medical chart or by parent report; or (2) they received a score >1 SD below the normative mean on the study measure of nonverbal IQ.

NH Control Sample

Participants in the NH control (NH) sample were 53 children, adolescents, and young adults (Table I). NH sample participants were required to pass a basic hearing screening (headphones were used to test each ear individually at frequencies of 500, 1,000, 2,000, and 4,000 Hz at 20 dB), to report no significant developmental or cognitive delays, and to be between ages 7 and 25 years inclusive. NH participants were 1:1 matched for age (± 2 years) and nonverbal IQ (± 1 standard deviation; nonverbal IQ score ≥ 1 SD below the mean was required for study inclusion) with participants in the CI sample. Other characteristics of the NH sample are described in Table I.

Recruitment

CI sample participants were recruited from patient populations receiving clinical services at a large hospital-based CI clinic. Recruitment approaches included contacting all eligible patients who has previously volunteered for research (or who were enrolled in active longitudinal research projects) and informing patients at clinical appointments of the opportunity to participate in the study. An attempt was made to provide notice of the study throughout all clinical and research programs of the CI clinic to recruit a broad sample of participants. The project was also advertised locally to professionals and schools who had contact with CI users. Control sample participants were recruited from the community using flyers and advertisements posted in the same institutions and geographic areas from which the CI sample was recruited, including e-mail and internet sites affiliated with the CI clinic and university.

Procedure

All study procedures were reviewed and approved by the university institutional review board, and participants were fully consented (with assent by children as appropriate) prior to initiation of any study procedures. All study visits took place at a hospital-based clinic, and all children with CIs were evaluated by licensed speech—language pathologists with extensive experience evaluating individuals with CIs; NH control participants were evaluated either by speech—language pathologists or by experienced psychometric

Table I. Demographics and Hearing History

Variable	CI sample	(N = 53)	Control sample ($N = 53$)		
	Mean (SD)	Range	Mean (SD)	Range	
Chronological age (years)	14.4 (4.1)	7.8–25.6	14.7 (4.4)	7.1–24.0	
Age at Implantation (months)	34.8 (19.3)	8.3-75.8	NA	NA	
Duration of CI Use (years)	11.5 (3.2)	7.1-19.8	7.1–19.8 NA		
Age of onset of deafness (months)	2.5 (7.2)	0–36	NA	NA	
Preimplant residual hearing (PTA)	107.8 (10.7)	85-118.4	NA	NA	
Communication mode	4.6 (0.9)	1–5	NA	NA	
Income level	7.1 (2.4)	2–10	2–10 7.3 (2.5)		
Nonverbal IQ (Matrix Reasoning T)	55.9 (6.0)	42–68	56.2 (6.9)	40-70	
Etiology of hearing loss (N)					
Meningitis	4		NA		
Other/unknown	49		NA		
Bilateral/bimodal CI					
Bilateral CI	16		NA		
Bimodal (CI + hearing aid)	3		NA		
Unilateral CI	34		NA		
Sex (female/male)	25/28		31/22		
Race/ethnicity					
Asian	1		3		
Black/African-American	1		6		
Hispanic	2		2		
Multiple races	3		5		
White	46		37		
Processor/strategy					
Nucleus 22/SPEAK	5				
Nucleus 24/SPEAK	2				
Nucleus 24/ACE	31				
Nucleus system 5/ACE	6				
Clarion/MPS	3				
Clarion/HiRes	2				
Sonata/CIS	1				
Combi 40+/CIS	3				

Note. Preimplant residual hearing is expressed as mean unaided pure-tone average (PTA) for the frequencies 500, 1,000, and 2,000 Hz in dB HL. Communication mode is coded on a scale from auditory-verbal (6) to mostly sign (1); income is coded on a 1 < 5,500 to $10 \ge 9,000$ scale. Other/unknown etiology for hearing loss includes auditory neuropathy (N=3), large vestibular aqueduct (N=1), Mondini malformation (N=3), familial (at least one other immediate family member also was deaf, of unknown etiology; N=9), and unknown (N=3). Processor/Strategy for bilateral users is coded for most recent implant/upgrade.

technicians. Evaluators completed a training process consisting of mutual observation and scoring, discussion of administration procedures, and supervision by a licensed clinical psychologist. Participants were administered speech and language measures in one test session and neurocognitive tests and questionnaires (including measures for the present study) at a second test session within the next 6 months. Only the test results relevant to executive functioning are presented in this article. Participants were paid \$20 per hour plus travel expenses (e.g., parking costs) to offset expenses related to time and travel.

Demographic variables coded for both samples included age at the time of testing, sex, family income (coded by income ranges on a 1 [<\$5,500] to 10

[≥\$95,000] scale, with values of 3, 5, and 7 corresponding to income values of \$15,000–\$24,999, \$35,000–\$49,999, and \$65,000–\$79,999, respectively), and race/ethnicity. Additional hearing history variables coded for the CI sample included age at onset of deafness, age at time of implantation, duration of deafness (from onset to implantation), preimplant residual hearing (mean unaided pure-tone average [PTA] in the better-hearing ear for the frequencies 500, 1,000, and 2,000 Hz in dB HL), communication mode (coded on a 1 [mostly sign] to 6 [auditory verbal] scale, with values of 1−3 reflecting total communication strategies [sign and speech to varying degrees of emphasis] and 4−6 reflecting oral communication strategies [speech used exclusively with no formal sign language

other than gestures]; Geers & Brenner, 2003), and etiology of hearing loss (reported for descriptive purposes; N was insufficient in any etiology category for further analysis).

Measures

General Approach to Measurement of Executive Functioning Skills

The test battery selected for this study was designed to provide a broad assessment of executive functioning skills by using multiple tests within each of the three executive functioning domains. A measure of nonverbal intellectual ability was also included to provide information about the global intelligence of study participants. Selection of the assessment instruments for this study was guided by the following principles: Minimal auditory or language requirements for understanding directions or completing test items, established psychometrics including reliability and validity in both NH and hearing-impaired populations across the age range of the study, and established age-based norms based on a national sample (expressed in T-scores [normative mean of 50 and standard deviation of 10], scaled scores [normative mean of 10 and standard deviation of 3], or standard scores [normative mean of 100 and standard deviation of 15]). For instances in which participants' ages were older than established norms, the highest norm age table was used to derive norm-based scores. Because norms for all tests were available at least into late adolescence or early adulthood (at which point they were at or near an asymptote), it is unlikely that this procedure resulted in significant distortion of norm estimates; nevertheless, scores based on use of norms out of age range should be interpreted with caution. For the Delis-Kaplan Executive Function System (DKEFS) Number-Letter Switching (NLS) task, norms for 8-yearolds were used to derive scaled scores for two 7-year-old participants.

To minimize the effects of audibility and modality-specific auditory spoken language processing on test results, most of the tests in the study involved the use of only visual materials for test stimuli and manual responses. Spoken directions and instructions were supplemented when necessary with nonverbal demonstrations, examples, and practice. Only three measures (Digit Span, Retrieval Fluency, and Stroop Color–Word Test) required spoken responses, and only one measure (Digit Span) required auditory perception of test items. Although efforts were made to have all participants complete all tests, some participants were unable to complete some of the tests owing to fatigue or (in the case of the CI participants) difficulty with the spoken language components of the tests noted

above. For any measure not completed by all participants, N is reported below.

Measure of Nonverbal Intellectual Ability

The Matrix Reasoning subtest of the Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999) requires subjects to complete a pattern of geometric designs based on an underlying concept. A substantial amount of research supports the reliability and validity of Matrix Reasoning tests in general and the WASI Matrix Reasoning test in particular as excellent measures of fluid nonverbal intelligence (Wechsler, 1999). WASI Matrix Reasoning T-scores were available for the full age range of the study and were used to measure global nonverbal intellectual ability.

Measures of Short-Term and Working Memory Skills

Three memory span subtests were used to measure sequential verbal and visual short-term/working memory: The Digit Span (N = 52 CI, 53 NH) subtest of the Wechsler Intelligence Scale for Children, Third Edition (WISC-III; Wechsler, 1991; the WISC-III version of this subtest was used to maintain continuity with prior studies at the clinic) and Spatial Span subtest of the Wechsler Intelligence Scale for Children, Fourth Edition-Integrated (WISC-IV-I; Wechsler et al., 2004) require subjects to reproduce a series of spoken digits or sequential locations, respectively, in forward order (Span Forward) or backward order (Span Backward). The Visual Digit Span subtest of the WISC-IV-I requires repetition of a rote forward order of digits that are visually presented to subjects in printed format. Scaled scores for these three span tests were used to measure sequential verbal short-term and working memory capacity in the auditory and visual modalities. WISC norms are available for Visual Digit Span and Spatial Span (Wechsler et al., 2004) as well as Digit Span Forward and Digit Span Backward (Kaplan, Fein, Kramer, Delis, & Morris, 1999) through age 16 years, 11 months.

Two holistic visual memory subtests were used to measure simultaneous (no sequential processing) visual memory skills: The Picture Memory and Design Memory subtests of the Wide Range Assessment of Memory and Learning, Second Edition (WRAML-2; Sheslow & Adams, 2003) present subjects with pictures of scenes or abstract designs (respectively). Subjects must recognize changes from the pictures of scenes (Picture Memory) or draw the designs from memory (Design Memory). Scaled scores (available for the full age range of this study) were obtained from both subtests.

Measures of Fluency-Speed

Verbally mediated fluency-speed skills were assessed with the Retrieval Fluency (N = 52 CI, 53 NH) subtest of the Woodcock-Johnson Tests of Cognitive Abilities, Third Edition (WJ-III; Woodcock, McGrew, & Mather, 2001), which requires subjects to rapidly generate words from specific semantic categories. Visual-perceptual fluencyspeed skills were measured with the Pair Cancellation and Visual Matching subtests of the WJ-III, which involve rapid identification of pictures or matching numbers (respectively) within visual arrays. Visual association fluency-speed skills were assessed with the Coding subtest from the WISC-IV-I, a measure of the ability to rapidly reproduce a sequence of symbols based on a corresponding sequence of numerals (each numeral corresponds to a unique symbol). Finally, visual-motor fluency-speed skills were evaluated using the Coding Copy subtest, which requires rapid reproduction of the symbols from the Coding subtest, without the corresponding numerals. Scores on the WJ-III measures are expressed as standard scores (available for the full age range of this study), whereas those for the WISC-IV-I are expressed as scaled scores.

Measures of Inhibition/Concentration

The Color-Word score of the Stroop Color-Word Test (expressed as a T-score based on norms for children up to age 14; 14-year-old norms were used for older children and adults because Stroop adult norms use an entirely different scoring algorithm; Golden, Freshwater, & Golden, 2003) was used to measure the ability to inhibit an overlearned/ automatic process (word reading) in favor of a more effortful, less automatic process (naming ink color), for a series of color words (either red, blue, or green) that are printed in ink colors that differ from the actual word. The NLS Condition of the Trail-Making Test (N = 52 CI, 53 NH) of the DKEFS (Delis, Kaplan, & Kramer, 2001) requires subjects to connect a series of numbers and letters randomly spaced on a page by alternating between numbers and letters (e.g., 1-A-2-B, etc.). DKEFS NLS scaled scores measure mental flexibility/shifting and attention-concentration skills. The Test of Variables of Attention (TOVA; Leark, Dupuy, Greenberg, Corman, & Kindschi, 1996) (N = 50 CI, 51 NH) is a continuous performance test that requires subjects to respond (with a button press) to a target stimulus (a square at the top of a screen) while not responding to a nontarget stimulus (a square at the bottom of a screen). Standard scores (available for the full age range of the study) for Response Time Variability (variation in response speed across all target stimuli of the test), Omissions (failing to respond to a target), and Commissions (responding inaccurately to a nontarget)

reflect consistent attention, vigilance, and impulsivity, respectively.

Statistical Approach and Analysis

Comparison With Developmental Benchmarks

Scores on the executive functioning measures for the CI sample were compared with two types of developmental benchmarks: (1) scores obtained from the matched NH sample and (2) scale norms. First, scores from measures in each of the three executive functioning domains were compared between the CI and NH samples using three separate Multivariate Analyses of Variance (MANOVAs), to evaluate CI vs. NH differences at a domain level. Specific test/subtest scores were then compared (CI vs. NH) using independent samples t-tests with 2-tailed p-values. Second, CI sample scores on the executive functioning measures were compared with mean norm scores using 1-sample *t*-tests. This analysis provided an evaluation of the deviation of the CI sample performance from national averages for age, although the CI group differed from nationally representative norm samples on some background variables (most notably, nonverbal IQ). Furthermore, to provide an estimate of the proportion of clinically significant scores on the executive functioning measures, the percentage of the CI and control samples scoring 1 or 2 standard deviations (SD) or more below the norm sample mean for each measure was also calculated. The -1 SD cutoff is emphasized in these analyses because it is a generally accepted (albeit arbitrary) criterion for differentiating average from below average performance. The -2 SD cutoff is much more stringent (and reflects much more severe deficits in functioning) and would be expected to include few participants given the sample sizes in this study (i.e., because \sim 2% of the normal curve falls below 2 SD below the mean, only 1 out of 50 participants would be expected to fall at this value given a normal distribution); as a result, analyses using the -2 SD cutoff should be interpreted with caution. The analyses reporting numbers of participants scoring below average relative to norms provide information about frequency of clinically significant scores in the CI sample, allowing for estimation of risk at the level of the individual as opposed to a group average.

Relations Between Executive Functioning Scores and Demographic and Hearing History Factors

To investigate relations between executive functioning scores and demographic and hearing history factors (current age, age at implantation, duration of use, onset age of deafness, best pre-implant PTA, communication mode, income, bilateral vs. unilateral implant, and gender),

correlations (Pearson for continuous variables; point-biserial for dichotomous variables) were calculated between executive functioning scores and demographic/hearing history factors within the CI subsample. Because of the large number of correlations (153) between demographic/hearing history variables and executive functioning scores, only correlations significant at the p < .001 level are discussed in the text to reduce the influence of alpha error on study results.

Results

Comparison of CI and NH Samples

As expected from the matching procedure, CI and NH samples did not differ on age (t(104) = 0.36, p = .724) or nonverbal IQ scores (t(104) = 0.27, p = .786) (Table I). Although not matched 1:1 on other parameters, the CI and NH samples also did not differ on family income (t(94) = 0.51, p = .615) or gender (p = .33 by Fisher's Exact Test) (Table I).

Relative to the NH control sample, the CI sample lower on memory measures (MANOVA F(7.96) = 8.47, p < .001). Specifically, significant differences (CI < NH) were found in forward digit span, whether presented in the auditory (Digit Span Forward) or visual (Visual Digit Span) modality, as well as in backward digit span. No differences between the samples were found for other visual or spatial memory measures (Table II). On nearly all measures of fluency-speed, the CI sample scored lower than the NH sample (MANOVA F(5,99) = 4.66, p < .001), with significant differences for Coding, Visual Matching, and Retrieval Fluency, and nonsignificant trends (p < .10) for Coding Copy and Pair Cancellation. Similarly, the CI sample scored lower than the NH sample on nearly all measures of inhibitionconcentration (MANOVA F(5,94) = 5.34, p < .001), with only Stroop Color-Word scores failing to reach statistical significance (Table II). For the statistically significant comparisons between the CI and NH samples, most effect sizes were in the medium (Cohen's d = 0.50; Cohen, 1992) to large (d = 0.80 or higher) range.

Comparison of CI Sample With Scale Norms

Despite having above average nonverbal IQ scores (t(52) = 7.20, p < .001, relative to the norm sample mean T-score of 50), the CI sample scored lower than the norm mean score on most measures of executive functioning (Table II). For the most part, scores that differed significantly from norms for the CI group were the same as those that differed from the NH control sample. On memory measures, the CI sample scored below scale

norms on WISC-III Digit Span Forward (t(51) = 8.68,p < .001) and Digit Span Backward (t(51) = 2.87,p = .006), WISC-IV Visual Digit Span (t(52) = 3.62,p = .001), and WRAML-2 Picture Memory (t(52) = 3.58, p = .001). On the other hand, the CI sample scored higher than scale norms on WISC-IV Spatial Span Backward scores (t(51) = 2.50, p = .016). On measures of fluency-speed, the CI sample scored below normative means on WISC-IV Coding (t(52) = 2.74, p = .008), WJ-III Retrieval Fluency (t(51) = 4.19, p < .001), and WJ-III Visual Matching (t(52) = 3.26, p < .002). On measures of inhibition-concentration, the TOVA response time variability, commission, and omission scores of the CI group fell below scale norms (t(49) = 4.47, 4.95, and 5.95, respectively, all p-values < 0.001), and D-KEFS NLS scores for the CI group showed a nonsignificant trend in the direction of being lower than norms (t(51) = 1.89,p = .065).

More than 30% of the CI sample demonstrated clinically significant delays (1 standard deviation or more below the normative mean) on measures of digit span forward, digit span backward, visual digit span, picture memory, coding, visual matching, and TOVA scores (Table II). By comparison, ~16% of the norm sample would be expected to score in that range. The proportion of the NH sample demonstrating clinically significant delays was consistently lower than that of the CI sample and, in almost all cases, was within a range similar to that expected in the norm sample (e.g., near 16%). On only one test (WRAML-2 Picture Memory) did >30% of the NH sample score ≥1 standard deviation below the norm mean.

At a more extreme level, at least 10% of the CI sample scored 2 SD or more below the normative mean (a range encompassing approximately the lower 2% of the norm sample) on verbal short-term/working memory and inhibition–concentration measures, with the exception of Stroop Color–Word and Digit Span Backward (Table II). For fluency–speed, the percentages of the CI sample scoring 2 or more SD below the normative mean were not as large, although most exceeded the 2% expected in a normative sample. The percentage of the control sample scoring 2 or more SD below the normative mean was \leq 2% for most executive functioning measures, consistent with the distribution of the normative samples.

Relationships Between Executive Functioning Scores and Demographic/Hearing History Variables

The large majority (137/153) of correlations between demographic/hearing history variables and executive functioning scores had p > .05 (a table of all correlations is

Table II. Cochlear Implant (CI) and Normal Hearing (NH) Sample Comparisons in Executive Functioning Domains

Executive Functioning Domain	CI		NH	·			
	Mean (SD)	%	Mean (SD)	%	ES	t	95% CI
Short-term/working memory							
Digit span forward	6.7 (2.7)	56/29	10.3 (3.0)	17/8	1.3	6.4***	2.5 to 4.7
Digit span backward	8.9 (2.9)	40/4	10.1 (3.2)	25/0	0.4	2.1**	0.0 to 2.4
Visual digit span	8.4 (3.1)	34/13	11.9 (2.2)	4/2	1.3	6.4***	2.4 to 4.5
SS forward	9.8 (2.6)	21/0	10.7 (2.9)	13/0	0.3	1.6	-0.2 to 1.9
SS backward	10.9 (2.4)	8/0	11.4 (2.9)	8/2	0.2	1.0	-0.5 to 1.6
Design memory	9.7 (2.5)	17/6	9.8 (2.5)	13/0	0.1	0.3	-0.8 to 1.1
Picture memory	8.6 (2.8)	38/6	8.4 (2.6)	40/8	0.1	0.4	-1.3 to 0.9
Fluency-speed							
Coding	9.0 (2.7)	34/2	10.2 (3.0)	19/0	0.4	2.2**	1.0 to 2.3
Coding copy	9.8 (3.0)	26/4	10.9 (3.1)	15/4	0.3	1.7*	-0.1 to 2.2
Visual matching	91.9 (18.1)	34/8	104.6 (13.9)	8/0	0.8	4.0****	6.4 to 18.9
Retrieval fluency	92.1 (13.6)	29/6	101.8 (10.0)	6/0	0.8	4.2****	5.1 to 14.3
Pair cancellation	98.6 (11.1)	9/2	102.4 (9.2)	6/0	0.4	1.9*	-0.1 to 7.8
Inhibition-concentration							
NL switching	9.2 (3.2)	25/10	11.1 (1.8)	4/0	0.7	3.8****	0.9 to 3.0
Stroop color-word	48.7 (11.1)	26/6	52.1 (11.0)	11/2	0.3	1.6	-0.8 to 7.7
TOVA RT variability	85.7 (22.6)	44/28	97.0 (16.4)	22/4	0.6	2.9***	3.5 to 19.0
TOVA commissions	83.5 (23.5)	54/30	98.8 (14.2)	22/6	0.8	4.0****	7.6 to 22.9
TOVA omissions	76.9 (27.5)	46/42	94.1 (18.8)	22/10	0.7	3.7****	8.0 to 26.5

Note. CI = cochlear implant sample; NH = normal-hearing sample. "%" = sample percentage scoring one standard deviation or more/two standard deviations or more below the normative mean. SS = Spatial Span; NL = Number-Letter; RT = response time.

T-test is for CI vs. NH (df=99–104; see Method section for N per measure), with effect size (ES) for this test expressed as Cohen's d. 95% CI is 95% confidence interval for difference in NH-CI scores.

available from the authors), and none of the conventional demographic/hearing history factors were found to be significantly correlated with nonverbal IQ scores (all p > .05). The only correlations reaching the corrected p < .001 level of significance were the following: For fluency/speed, use of an auditory–oral communication mode was related to higher Visual Matching (r = .50, p < .001) scores. For inhibition–concentration, Stroop Color–Word scores were positively related to older age (r = .48, p < .001) and longer duration of CI use (r = .44, p = .001).

Discussion

This study is the first investigation of several domains of executive functioning in long-term CI users comparing their performance with developmental norms and with scores from a closely matched NH control sample. Study results demonstrate moderate to marked weaknesses in the CI sample across broad areas of executive functioning relative to matched NH participants and scale norms, despite above average nonverbal intelligence. Specifically, long-term CI users scored below matched NH peers and developmental norms on measures of verbal memory

capacity, visuospatial and verbal fluency/speed, and inhibition—concentration, with effect sizes in the medium to large range (Cohen, 1992). Approximately 1/3 to 1/2 of the CI sample scored in a below average range on most of these measures, a rate that is at least twice as high as the norm and matched control samples. The smallest deviations of the CI group from the control group and developmental norms were found on measures of nonverbal visual—spatial memory skills. Demographic and hearing history variables were, in almost all cases, not significantly related to executive functioning scores in the CI group.

The present findings suggest that a significant subgroup of long-term CI users may not "catch up" to their NH age peers in several core areas of executive functioning, despite having the benefit of CI use for at least 7 years. Approximately 1/3 to 1/2 of the sample of CI users scored 1 or more SD below the mean on measures of verbal short-term/working memory capacity, fluency–speed (with the exception of Pair Cancellation), and inhibition–concentration. Because of the critical importance of executive functions for broad behavioral, academic, and social adjustment (Barkley, 1997), even moderate delays or deficits in these areas are functionally important and are

^{*}p < .10, **p < .05, ***p < .01, ****p < .001.

therefore appropriate intervention targets. Furthermore, >10% of the sample of CI users scored in a more extreme range of 2 or more SD below the normative mean on almost all measures of verbal short-term/working memory and inhibition–concentration, suggesting that risk of more severe deficits in these areas is not uncommon.

Prior research has demonstrated poorer performance on continuous performance test (CPT) measures of attention and concentration in samples of children with hearing aids or CIs (Horn, Davis, Pisoni, & Miyamoto, 2005; Quittner, Smith, Osberger, Mitchell, & Katz, 1994; Smith, Quittner, Osberger, & Miyamoto, 1998). Results from those earlier studies have also suggested that longer use of a CI might be related to improved CPT performance, as a result of a positive influence of access to auditory experience on the development of attention skills (Quittner et al., 1994; Smith et al., 1998). Unlike those prior studies, however, the current study investigated much longer durations of CI use (≥7 years, compared with ≤6 years in most prior research) and used a large rigorously 1:1 matched control sample of NH children. Furthermore, prior studies used verbally mediated symbols (typically numerals) as the stimuli for CPT tests, whereas the current study used a CPT test (the TOVA) with visualspatial location of a shape as the target. It may be that improvement on CPT performance in the prior studies was mediated by improved phonological coding, verbal rehearsal skills, and verbal fluency with increased CI use, whereas the present study used a task that was less dependent on verbal coding and verbal mediation. The finding of well below average performance on the CPT task for the CI participants in the current study (TOVA) indicates that deficits in attention, concentration, and inhibition persist for years after cochlear implantation in a large proportion of children with CIs.

Chronological age and duration of CI use had no consistent relationship with executive functioning, indicating that beyond 7 years of CI use, duration of experience with the CI was not related to executive functioning outcomes. However, these correlations should be regarded with caution, because participants with more years of CI use also received their CIs in an earlier era and therefore may not have received the full benefits and improvements of the most modern CI devices and processing strategies at the time that they were implanted. To address this possible cohort effect, a longitudinal design would be needed.

Three of the executive functioning subtests that were significantly different between the CI and NH groups involved rapid phonological coding and processing of verbal stimuli: Digit Span, Visual Digit Span, and Retrieval Fluency. However, only the Digit Span subtest requires

auditory perception of spoken language. The other two subtests (Visual Digit Span and Retrieval Fluency) involved verbal processing but were not dependent on hearing or audibility of the test stimuli. Comparison of the Digit Span Forward and Visual Digit Span subtests is therefore particularly relevant, as both involve rapid encoding and retrieval of sequences of digits, differing primarily in the modality of presentation of the test items: Auditory for Digit Span Forward and visual for Visual Digit Span. The fact that Visual Digit Span scores for the CI group also fell below scores for NH controls and developmental norms indicates that delays in Digit Span Forward in the CI population are not solely the result of auditory perception and modality-specific auditory encoding deficits. Hence, auditory perceptual factors alone cannot account for the delays observed in scores on these tests. Importantly, all of these subtests are at least in part dependent on rapid phonological coding, verbal rehearsal, verbal retrieval speed, and efficient use of lexical knowledge, which are known to be at high risk in children with CIs (Pisoni et al., 2011). Hence, the use of executive functioning skills that rely in part on language-based cognitive processing may be particularly at risk for children with CIs, regardless of whether auditory sensory processing operations are involved.

In this study as in prior research, the CI sample scored lower on measures of digits recalled forward than on a measure of digits recalled backward. Similar to findings of the current study, Pisoni et al. (2011) found that smaller percentages of a CI sample scored in normatively deviant ranges on a measure of digit span backward than on a measure of digit span forward, although the CI sample scored well below norms on both tasks. In a longitudinal study, Harris et al. (2013) also found that both digit span forward and digit span backward scores consistently lagged behind those of developmental norms, with larger deviations for forward than for backward digit span. These results suggest that the concurrent cognitive task required by digit span backward (digit reversal) may have less of an effect (relative to norms) on children with CIs than the basic short-term verbal memory task inherent in both subtests of digit span. Further research on working memory dynamics is recommended to better understand the ability of children with CIs to process short-term verbal memory information concurrent with other mental operations, in order to better understand the relative roles of short-term rehearsal and central executive management of cognitive resources in working memory in children with CIs (Baddeley, 2007).

In addition to findings with more verbally mediated memory tests, the CI sample scored lower than the matched NH sample on several subtests that consisted of visual stimuli including Coding, Visual Matching, TOVA, and NLS. Significant differences on the Coding and Visual Matching tests suggest that fluency–speed differences between the groups are probably not completely mediated by auditory–verbal skills. Similarly, TOVA scores measure sustained attention, concentration, and inhibition without requiring auditory coding or verbal processing. Hence, findings of executive functioning weaknesses extended beyond exclusively auditory–verbal tests, consistent with a more domain-general deficit in executive functioning in a proportion of CI users.

Several possible factors may explain a potential domain-general weakness in executive functioning in children with CIs. Conway et al. (2009), for example, summarize research demonstrating that children with CIs may have domain-general sequential processing deficits resulting from reduced access to auditory experiences, which provide sequential input to the developing brain. These sequential processing deficits may produce downstream effects on domain general (i.e., not dependent on auditory-verbal processing alone) areas of executive functioning that require sustained, serial processing, such as sustained attention and working memory for sequential stimuli (Conway et al., 2009). It is also notable that processing speed is involved to a degree in almost all of the tests (even in areas other than fluency-speed) that differentiated CI and NH samples, suggesting that efficiency of core information processing operations may be a factor differentiating the groups. Furthermore, Pisoni et al. (2011) showed that underspecified cognitive representations (particularly verbal representations) in children with CIs may have a general effect on efficiency and speed of processing, ultimately affecting working memory. The present findings support a hypothesis of domain-general executive functioning risks in children with CIs, extending beyond first-order sensory factors related directly to audibility and auditory functioning. Future research is recommended to identify the mechanisms (such as sequential processing and/or fluency-speed) underlying this domain-general executive functioning weakness.

Despite significant group differences between CI and NH samples on several measures of executive functioning, the CI sample scored within normal limits relative to developmental norms on several measures that are heavily dependent on visuospatial skills: Spatial Span, Design Memory, Coding Copy, and Pair Cancellation. For some of these variables, the differences between the CI and NH samples were nearly significant (Coding Copy, Pair Cancellation) or in the predicted direction consistent with prior research (Spatial Span Forward) (Conway)

et al., 2009; Pisoni et al., 2008). For these measures, the NH sample scores were more consistent with their (above average) nonverbal IQ, whereas the CI sample scores were lower than expected given their above average nonverbal IQ. Differences on these measures may therefore reflect insufficient power (need for larger N) and the fact that the CI sample had a higher nonverbal IQ than the samples used to derive developmental norms (which, by definition, should be approximately average in IQ). For other scores (especially Design Memory), however, the matched groups were nearly equivalent and scored at or near developmentally normal limits. This latter finding suggests that differences between CI and NH samples may be smallest for executive functions involving more exclusively holistic simultaneous visual–spatial skills.

One primary purpose of this study was to assess whether differences exist in executive functioning between children with CIs and developmental benchmarks, which is a necessary first step for a better understanding of the impact of auditory deprivation on executive functioning. Given the finding that such delays in executive functioning were found, an important next step in the research is identifying the core underlying factors that may account for these delays. In particular, language development may play an important protective or exacerbating role in executive functioning risk for children with CIs. Hence, investigation of the possible mediating role of different components of speech and language skills (e.g., vocabulary, comprehension, language-mediated concept formation) in executive functioning development may provide some insight into the causal information processing mechanisms. Because existing research suggests that the causal relationship between language and working memory development is likely to be reciprocal (Harris et al., 2013; Kronenberger et al., 2012), longitudinal methods would be ideally suited for investigation of this topic. Additionally, although ~30-50% of the CI sample showed clinically significant deficits in the form of below average executive functioning scores, at least half of the sample scored within average ranges (within 1 SD of the normative mean or higher). Therefore, many children with CIs have adequate executive functioning skills, and identification of the protective factors present in this group may assist in understanding and remediating deficits in at-risk children with CIs.

When interpreting the results of this study, it is important to emphasize that both samples had significantly above average nonverbal intellectual ability. Although this adds confidence that executive functioning delays in the CI sample are not a result of global intellectual delays, it also raises the need for additional study of executive

functioning in CI samples with average and below average intellectual abilities. An additional factor to consider in interpreting the present results is the potential impact of subtle and undetected/unknown neurological vulnerability in the CI sample. Although efforts were made to exclude children with known neurological and neurodevelopmental conditions in addition to deafness, it is possible that undetected/unknown neurological conditions in some CI participants may have affected their results. Therefore, differences between the CI and NH groups could be a result of currently unknown neurological conditions in addition to differences in auditory experience.

Several other study characteristics should also be taken into account when interpreting the results of this study. First, the study sample was cross-sectional; therefore, cohort effects could have influenced correlations with any of the age variables. Second, the study was powered to detect medium to large effect sizes, and therefore it is possible that small, but substantive, effect sizes were not detected. This caveat is particularly relevant for several of the executive functioning measures that approached significance levels in comparison with the NH control sample. Third, because multiple measures were evaluated for each area of executive functioning, a large number of statistical tests were conducted, raising the possibility of alpha error. Although we attempted to reduce this possibility through the use of MANOVA analyses for group comparisons and by using a more stringent p-value for the correlational analyses, replication of study findings with another study sample would add more confidence to the results. Fourth, some participants were tested at ages exceeding the norm tables available for tests. Specifically, WISC-III and IV norm tables end before age 17 years and SCWT norms end before age 15 years (the child version of the Stroop Color-Word Test (SCWT) was used; Golden et al., 2003). However, inspection of the norm tables for these tests demonstrates that the growth of average scores with increasing age on these tests has slowed significantly by the time that the highest norm age is reached. For example, the mean norm scores for all WISC-III/IV memory span subtest scores are <1 raw score point different (and in most cases are identical) from ages 15-16 years, and the Coding raw score ranges corresponding to a normative mean scaled score of 10 overlap at those ages. Similarly, the mean norm score for SCWT Color Word scores for 13-14-yearolds is only 2 words more (out of a total of 40 words) than that for 11-12-year-olds.

Finally, although we attempted to cluster our measures into the three theoretically based at-risk executive functioning domains for children with CIs, it is important to emphasize that no measure falls exclusively within a

single domain. All of our inhibition-concentration measures, for example, also included a component of fluency and speed, and the median correlation between the five inhibition-concentration and the five fluency-speed measures was r = .26 (a table of these correlations is available on request from the authors). Although this value is lower than the median correlation between the processing speed measures themselves (r = .41), it nevertheless does suggest some overlap between the inhibition-concentration and fluency-speed measures. Additionally, all executive functioning measures demand controlled attention to some degree, which also produces conceptual and empirical overlap. For example, the correlations between Digit Span Forward and TOVA scores in the combined sample were r = .35 for commissions and r = .27 for omissions (p < .01). However, these values are lower than the intercorrelations of Digit Span Forward with other measures of verbal working memory such as Digit Span Backward and Visual Digit Span (r = .40 and .55, respectively, p < .001). Nevertheless, it is possible that shared characteristics of the measures such as controlled attention and fluency-speed affected differences between the samples, even for measures that were not explicitly labeled as indexes of attention or fluency-speed. Such a finding would be consistent with other studies of children with hearing impairment and CIs demonstrating poorer visual attention skills relative to controls and to scale norms (Horn et al., 2005; Kelly et al., 1993; Quittner et al.,

In summary, this study is the first 1:1 case-controlled matched investigation of a broad set of executive functions in early-implanted deaf children with CIs using neurocognitive measures that place minimal demands on audibility and auditory processing. Results showed multiple areas of weakness in executive functioning for children with CIs relative to matched NH controls and to scale norms, supporting hypotheses that domain-general executive functions in the areas of working memory, fluencyspeed, and inhibition-concentration are at high risk in this clinical population. Although verbally mediated executive functions appear to be at particular risk (even in the absence of auditory stimuli), scores on nonverbal measures of fluency-speed and inhibition-concentration were also found to be below average, consistent with domain-general influences of auditory deprivation in long-term CI users. Therefore, this finding has significant implications beyond the specific area of cochlear implantation, because it demonstrates that sensory deprivation and related influences may have cognitive effects extending well beyond the information-processing domains related directly to the impaired area of sensory processing. Investigation of the effects of such deprivation early in childhood should not be limited only to sensory domain-specific cognitive factors.

Clinically, these findings have significant implications for the way in which functional outcomes for children with CIs are evaluated and monitored. As many as 1/3 to 1/2 of children with CIs may be at risk for below average functioning in some areas of executive functioning, and as many as 1/10 or more may have severe deficits in those areas. Currently, speech and language skills receive almost sole emphasis in follow-up functional testing of children with CIs. Our results suggest that executive functioning skills should also be closely monitored and regularly assessed in children with CIs, with appropriate interventions as needed. Furthermore, interventions and experiences that can enhance development of executive functioning and cognitive control may reduce the impact of auditory deprivation and verbal delays. Based on the current findings, fluency-speed, inhibition-concentration, and verbally mediated working memory appear to be at particular risk and therefore present the most significant intervention targets. Although additional research is needed to better understand the development of executive functioning skills in early childhood following cochlear implantation, novel early interventions addressing these long-term at-risk areas of executive functioning may have the potential for improving global adjustment in some individuals with CIs.

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