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Executive Functioning Skills in Preschool-Age Children With Cochlear Implants

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Abstract

Purpose—The purpose of this study was to determine whether deficits in executive functioning (EF) in children with cochlear implants (CIs) emerge as early as the preschool years.

Method—Two groups of children ages 3 to 6 years participated in this cross-sectional study: 24 preschoolers who had CIs prior to 36 months of age and 21 preschoolers with normal hearing (NH). All were tested on normed measures of working memory, inhibition-concentration, and organization-integration. Parents completed a normed rating scale of problem behaviors related to EF. Comparisons of EF skills of children with CIs were made to peers with NH and to published nationally representative norms.

Results—Preschoolers with CIs showed significantly poorer performance on inhibition-concentration and working memory compared with peers with NH and with national norms. No group differences were found in visual memory or organization-integration. When data were controlled for language, differences in performance measures of EF remained, whereas differences in parent-reported problems with EF were no longer significant. Hearing history was generally unrelated to EF.

Conclusions—This is the first study to demonstrate that EF deficits found in older children with CIs begin to emerge as early as preschool years. The ability to detect these deficits early has important implications for early intervention and habilitation after cochlear implantation.

Cochlear implantation has become the standard of care for children born with a bilateral severe-to-profound sensorineural hearing loss who receive minimal benefit from hearing aids (Sarant & Garrard, 2014). As of 2010, over 28,000 children in the United States had received a cochlear implant (CI), in which an electrode array is surgically implanted into the cochlea to provide direct stimulation to the auditory nerve, bypassing the damaged parts of the inner ear and allowing the brain to perceive and process sound (O'Donoghue,

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Nikolopoulos, & Archbold, 2000). A CI does not restore normal hearing, but with extensive speech and language rehabilitation, most deaf children who receive an implant by age 2 learn to perceive and produce spoken language and enter a mainstream educational setting by first grade, although their speech perception, language, and literacy skills lag, on average, behind those of their peers without hearing loss (Montgomery, Magimairaj, & Finney, 2010; Nittrouer, Caldwell, & Holloman, 2012). Although these findings are encouraging, there is a considerable amount of individual variability in speech, language, and literacy development in children with CIs, and a full range of outcomes are observed (Davidson, Geers, Blamey, Tobey, & Brenner, 2011; Ganek, McConkey Robbins, & Niparko, 2012; Geers & Hayes, 2011; Niparko et al., 2010; Pisoni et al., 2008).

New theoretical and empirical developments suggest that deafness and hearing impairment cannot be viewed in isolation as a simple sensory impairment (Luria, 1973; Myklebust, 1960). The inability to perceive auditory sensory information (including spoken language) and to participate in communicative experiences involving spoken language from birth impacts neural organization and the development of domain-general neurocognitive skills that rely on auditory experiences, speech perception, and spoken language processing (Conway, Pisoni, & Kronenberger, 2009; Luria, 1973; Pisoni et al., 2008). In order to better understand these global outcomes of children with CIs, new research efforts have targeted a broader set of neurocognitive processes beyond traditional product-based, end-point, speech-language measures. Converging evidence suggests that a set of domain-general executive-organizational-integrative (EOI) processes may be impacted by a period of auditory, speech, and language deprivation and delays and that disturbances in these foundational processes may explain an additional source of variance in speech and language outcomes of deaf children with CIs beyond the conventional predictors related to demographic, device, and child variability that have been studied extensively in the past (Geers, Brenner, & Davidson, 2003).

Executive functions are self-regulatory processes that include attention, inhibitory control, nonverbal and verbal working memory, emotion regulation, planning, and problem solving. Executive functions are self-directed actions and are used to purposefully modify one's own behavior in order to make a goal more or less likely to happen (Barkley, 1997a, 2013). Executive functioning (EF) emerges during the first year of life and has a protracted developmental time course that continues throughout early adulthood as it parallels neurological development of the prefrontal system (Steinberg, 2010; M. C. Welsh & Pennington, 1988). Although the component processes of executive function are less differentiated in preschool-age children compared with school-age children, latent variable studies of typically and atypically developing preschool-age children support a two-factor model of executive function, with working memory and inhibitory control identified as separate factors (Miller, Giesbrecht, Müller, McInerney, & Kerns, 2012; Miller, Müller, Giesbrecht, Carpendale, & Kerns, 2013; Schoemaker et al., 2012). Latent variable analysis indicates significant growth in EF of typically hearing children of about two standard deviations between the ages of 4 and 6 years old with large individual differences (Hughes & Ensor, 2011). Furthermore, EF skills in the preschool years predicts SAT scores, attentiveness, concentration, self-control, and ability to cope with stress and frustration during adolescence (Moffitt et al., 2011; Shoda, Mischel, & Peake, 1990). And they are

associated with physical health, financial well-being, and criminal outcomes in adulthood (Moffitt et al., 2011).

Understanding the development of EF skills during the preschool years is particularly important as several longitudinal studies provide evidence that both inhibitory control and working memory skills reliably predict math and literacy in the early elementary grades (Blair & Razza, 2007; Bull, Espy, & Wiebe, 2008; McClelland et al., 2007; Ponitz, McClelland, Matthews, & Morrison, 2009; J. A. Welsh, Nix, Blair, Bierman, & Nelson, 2010). Blair and Razza (2007) reported that inhibitory control during preschool measured with the peg-tapping task (which requires a child to inhibit a prepotent motor response) uniquely predicted children's kindergarten math ability after controlling for language and IQ.

Extending these findings using hierarchical linear modeling, McClelland et al. (2007) reported that children with stronger growth in inhibitory control between preschool and kindergarten, as measured by the head-to-toes task (which also requires children to inhibit a prepotent motor response), also had stronger growth in emerging literacy gains, vocabulary skills, and math skills than children with weaker growth in inhibitory control. Hughes and Ensor (2011) found that preschoolers with low gains in EF between ages 4 and 6 experienced more internalizing and externalizing behavior problems as reported by teachers, whereas high gains predicted self-perceived academic competency at age 6. Finally, a recent study by J. A. Welsh et al. (2010) used path analysis to show that, even after controlling for language skills, growth in executive function (i.e., verbal working memory, inhibitory control, and attention shifting) between pre-kindergarten and kindergarten uniquely predicted reading and math achievement in kindergarten. In sum, these studies all provide converging support for the proposal that domain-general cognitive skills such as EF skills during the preschool age, particularly behavioral regulation and the active control of attention, contribute significantly to the development of domain-specific academic skills in literacy and math. The development of executive skills that children can draw upon to keep themselves from talking a inappropriate times, to participate in circle time amid distractions, to remember to raise their hand, and to remember and complete multistep directions may provide the foundational skills necessary for robust learning in a classroom setting.

Language and Executive Function

Although the nature of the relationship is unclear, there is growing evidence for a link between language skills and EF skills in children with specific language impairment (Montgomery et al., 2010), autism spectrum disorder (Akbar, Loomis, & Paul, 2013), and language disorders (Gathercole, 1990), who all show deficits in particular areas of EF. Little is known, however, about the developmental relationship between language and executive function in children with CIs, many of whom experience language delays and slower rates of growth in language compared with typically hearing peers (Geers, Moog, Biedenstein, Brenner, & Hayes, 2009; Geers & Sedey, 2011; Niparko et al., 2010). Although significant correlations between language and executive function in children with CIs have been reported, suggesting close relations between the two domains, it remains unclear if the

language delay experienced by children with implants contributes to a delay in executive function or if executive delays contribute to delays in language.

Surowiecki et al. (2002) measured visual memory (i.e., recognition memory, delayed recall, paired associative learning), executive function (i.e., attention shifting, spatial working memory, Tower of London), and language in a group of 6- to 14-year-olds with hearing aids or CIs. The authors found that, after controlling for age, visual memory tasks but not executive function tasks were significantly correlated with their measure of global language ability. In a more recent study, Figueras, Edwards, and Langdon (2008) reported significant positive correlations between language and executive function (i.e., planning, set shifting, working memory, impulse regulation) in 8- to 12-year-old children with CIs and hearing aids after partialing out age, degree of hearing loss, and number of years with their current device. The authors concluded that the delay in executive function in children with implants and hearing aids was due to a delay in language acquisition because differences in executive function between children with implants or hearing aids and those with typical hearing were no longer significant after entering language as a covariate.

Although some studies provide evidence for a link between language and particular domains of executive function in school-age children with hearing aids or CIs, the direction and timing of the relationship across development is unknown, as is the impact of auditory-related factors such as age at implantation. It is possible that deaf children with better executive control are better able to attend to and efficiently process information in the environment necessary for language acquisition or that children with better language skills are able to use language for more effortful control over their behavior, emotions, and thinking. It is also possible that the relationship between language and executive function is more specific with regard to particular linguistic and executive skills. For example, in their sample of high-functioning children with autism, Akbar et al. (2013) reported that both general language ability and pragmatic language skills predicted children's performance on direct measures of working memory but not on measures of organization, shifting, or inhibitory control. Furthermore, general language predicted parent reports of working memory skills, whereas pragmatic skills predicted teacher reports of working memory; neither general language nor pragmatic language predicted parent or teacher reports of the other executive skills measured, which included organization, shifting, and inhibitory control.

Longitudinal studies are needed to model the complex relationship between development of the component processes of executive function and the development of various components of language (i.e., syntax, morphology, pragmatics, vocabulary, receptive and expressive language) in children with CIs beginning during the preschool years, a time when the structure and organization of executive function is undergoing significant change and children are becoming more competent language users and beginning to use language as a tool for thinking (Bodrova & Leong, 2007; Nelson, 1996).

Executive and Organizational-Integrative (EOI) Processes

EOI processes encompass the supervisory-attentional, executive, or cognitive-control system that allows for active control of attention, use of working memory, fluent-speeded processing, and integration of multiple sources of information (Norman & Shallice, 1986). These foundational information processes are necessary for efficient allocation of cognitive resources required to interpret novel auditory inputs provided by a CI and to apply this information to higher-order speech perception and spoken language processing operations (Conway et al., 2009; Kral & Sharma, 2012; Pisoni & Cleary, 2003; Pisoni, Conway, Kronenberger, Henning, & Anaya, 2010; Pisoni, Kronenberger, Roman, & Geers, 2011). Early auditory experience and activities with sound patterns promotes the development of EOI processes by providing opportunities to engage in controlled information-processing activities such as sustaining attention over time (e.g., focusing on one talker against background noise), coding and manipulating auditory-verbal-linguistic information in memory during competing cognitive operations (auditory-verbal working memory), integrating sequences into wholes (e.g., chunking auditory patterns into meaningful sounds and linguistic units), and engaging in fluent-speeded processing of sound sequences and temporal patterns (e.g., comprehension of a series of spoken sentences). Cognition and brain development do not occur in isolation from the development of attention, learning, and memory as a result of a predetermined blueprint but rather are the by-products of self-organization of the central nervous system based on experiences provided to the child from the environment combined with core elementary cognitive operations used to encode, store, and retrieve information from memory (Lewis, 2005; Singer, 1986). Hence, early experience with cognitive control processes, sustained attention, sequential organization, and fluent processing of temporal information is essential for the development of robust EOI functions. In turn, EOI functioning is one of the foundational building blocks for processing information in the auditory-verbal modality. As a result, strong bidirectional relations exist between spoken language development and the development of EOI processes (Conway et al., 2009; Figueras et al., 2008; Marchman & Fernald, 2008).

Because of the importance of auditory and verbal experiences and activities in the development of EOI processes, the reduced and degraded auditory input associated with deafness and use of a CI may alter and/or disturb the typical developmental process of EOI skills. Recent findings investigating EOI skills in school-age children and adolescents with CIs provide support for this hypothesis. Children with CIs score lower than normal-hearing peers on measures of working memory capacity (Pisoni & Cleary, 2003), verbal rehearsal speed, short-term memory retrieval speed (Burkholder & Pisoni, 2003), visual sequence learning (Cleary & Pisoni, 2001), and visual attention (Horn, Davis, Pisoni, & Miyamoto, 2005; Quittner, Smith, Osberger, Mitchell, & Katz, 1994). Compared with normal-hearing children, more children with CIs show below-average growth in verbal short-term memory and verbal working memory capacity, and slow working memory growth rates have been found to predict later speech and language outcomes (Harris et al., 2012; Kronenberger, Pisoni, Harris, et al., 2013; Pisoni et al., 2011).

A recent study of the EF skills of 53 children, adolescents, and young adults who used their CIs for 7 years or longer reported delays on a range of neurocognitive tests of EOI skills

relative to the normal-hearing population (Kronenberger, Pisoni, Henning, & Colson, 2013). Despite above-average nonverbal IQ in the CI sample, participants scored lower than a control sample on auditory and visual measures of short-term and working memory, verbally mediated fluency-speed skills such as retrieval fluency, and measures of inhibition-concentration such as the trail-making test and continuous performance tests. Studies using parent-report behavior checklist measures of EF have also found poorer scores in children with CIs or hearing aids relative to controls. Parents of 54 adolescents ages 16 to 18 with CIs rated their children as having more problems related to executive function, including inhibitory control, planning/ organizing, and shifting of attention compared with the control sample (Beer, Pisoni, Kronenberger, & Geers, 2010). Similarly, parents of 45 school-age children ages 5 to 18 with CIs rated their children as having more problems with working memory, inhibitory control, and behavioral regulation (Beer, Kronenberger, & Pisoni, 2011; Beer et al., 2010). Other researchers studying children with CIs and hearing aids have reported similar delays in EF, particularly on language-mediated tasks of inhibitory control and working memory capacity (Figueras et al., 2008; Surowiecki et al., 2002). In their study comparing the executive skills of 8- to 10-year-old children with deafness to hearing peers of the same age, Figueras et al. (2008) reported significant differences between the groups on the number of errors made on the tower task (which requires the development and execution of a plan for rearrangement of objects according to a set of rules), number of errors and time to complete the day/night task (child must provide a verbal response [e.g., day] to a visual stimulus associated with the opposite concept [e.g., a moon]), and number of correct sorts and attempted sorts on a card sorting test.

In summary, core foundational components of EOI functions display rapid development in the preschool years in typically hearing preschoolers, and deficits in EOI development during preschool years are related to long-term academic, social, and vocational outcomes. Taken together, research suggests that several core EOI functions may be at risk in school-age children, adolescents, and young adults with CIs. Furthermore, because EOI functions influence speech and language development (Harris et al., 2012; Kronenberger, Pisoni, Harris, et al., 2013; Pisoni et al., 2011), delays and/or disturbances in EOI development may put children with CIs at greater risk for speech and language delays, which are already of clinical concern for this population. As a result, EOI development in children with CIs during preschool ages may be a critically important source of variance affecting multiple areas of long-term outcome in addition to the development of conventional/ traditional speech-language skills. However, the majority of research on EOI skills has focused on school-age children, adolescents, and young adults, and little is known about the process and timing of development of EOI functions in prelingually deaf children who receive CIs at younger ages. There has been no research on the development of EOI skills in children with CIs during the preschool years, despite the fact that EF undergoes rapid initial development in normal-hearing children during that time frame (Garon, Bryson, & Smith, 2008). Do differences in EOI skills found for older children with CIs emerge (and can they be detected) during preschool ages? If differences are found, which areas of EOI are particularly at risk in this age range? This knowledge is crucial not only for understanding the process of EF development but also for identifying ages for early assessment and intervention.

In order to address this critical gap in our knowledge of EF, we compared the EOI skills of preschool children with CIs to two different developmental benchmarks: (a) the skills of a control group of children with typical hearing and (b) nationally representative norms. The relations between EOI scores and demographic/hearing-history history variables and language skills were also investigated in order to better understand the underlying factors contributing to early development of EOI skills in preschool children with CIs.

Method

Participants

Two groups of children participated in this study: a group of 24 deaf children with CIs (10 girls, 14 boys) and a group of 21 children with normal hearing (NH) (eight girls, 13 boys). In order to be eligible for the study, children in the CI group had to receive their implant prior to age 3 and to be between ages 3 and 6. In addition, the following inclusionary criteria were also required for children with CIs: (a) onset of hearing loss prior to age 36 months, (b) profound hearing loss bilaterally (>90 dB HL in the better-hearing ear), (c) a home environment in which English was the primary language spoken (not bilingual), (d) current or prior enrollment in an aural rehabilitative program and/or educational setting that encouraged the development of speaking and listening skills, (e) absence of any additional handicapping conditions other than hearing loss (e.g., autism spectrum disorders and multiple handicaps), and (f) use of multichannel CIs. All CI participants for this research project were recruited from populations currently being seen for clinical services at a large university hospital-based CI clinic or responded to advertisements posted in the community.

General inclusion criteria for NH participants included (a) normal hearing and language development as assessed by parent report at the time of enrollment (including never having been enrolled in any program for hearing impairment and never having used a sensory aid or assistive device for listening); (b) pure-tone average (PTA) within normal range as assessed by a hearing screening in the soundfield for 500, 1000, and 2000 Hz; (c) absence of any history of neurological or developmental conditions that require chronic management by a physician or special accommodations in the school or at home; (d) English as a first language in the school setting and at home; and (e) age 3–6 years. Children with NH were recruited through advertisements posted in the same hospital and community settings that were used for recruitment of children with CIs.

Procedure

All children in this study were recruited as part of a longitudinal study investigating neurocognitive and speech and language development in preschool children with CIs. The data reported for this article were obtained from the first annual testing visit. Speech-language pathologists with significant experience testing deaf children with CIs administered tests to all children with CIs; children with NH were tested either by the speech-language pathologists or by an experienced research technician who was trained for reliability by the speech-language pathologists. All children took a core battery of tests of EOI functioning (additional tests were administered to a subset of children who were able to continue after the core battery, but results of those tests are not reported here) as well as a set

of conventional speech-language tests. Testing required two visits. The core EOI test battery consisted of three nonverbal neurocognitive tests, each of which were selected to evaluate a specific core area of EOI skills (short-term/working memory, inhibition-concentration-vigilance, and organization-integration). All children also completed a test of global nonverbal ability.

Testing was administered in the mode of communication used in the child's school environment: 22 children with CIs used oral communication, and two children with CIs used total communication (speech and Signed Exact English). Parents completed behavior checklists, including a checklist of EOI functioning. All study procedures were reviewed and approved by the Institutional Review Board, and informed consent was obtained from all parents of participating children.

Demographic variables coded for both samples included chronological age at the time of testing, sex, family income (coded by income ranges on a 1 [under $5,500] to 10 [$95,000 and over] scale, with values of 3, 5, and 7 corresponding to income values of $10,000–$14,999, $25,000–$34,999, and $50,000–$64,999, respectively), and race–ethnicity. Additional hearing-history variables coded for the CI sample included age at onset of deafness, age at time of hearing aid fit and implantation, duration of deafness (from onset to implantation), preimplant residual hearing (mean unaided PTA in the better-hearing ear for the frequencies 500, 1000, and 2000 Hz in dB HL), and 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]) based on Geers and Brenner (2003), and etiology of hearing loss. Etiology of hearing loss in the CI sample was unknown for 18 children; syndromic for two children (Waardenburg syndrome); and auditory neuropathy, genetic, meningitis, and mondini malformation, for one child each. A summary of the demographics is provided in Tables 1 and 2.

Measures

All EOI measures were selected to be developmentally appropriate for children ages 3 through 6 with a history of hearing or language impairment and are components of well-known, normed (for the entire age range of the study) test batteries. Norm-based scores for the tests are scaled scores ($M = 10$, $SD = 3$), standard scores ($M = 100$, $SD = 15$), or T scores ($M = 50$, $SD = 10$) based on comparison of the participant's score with a nationally representative sample of same-age peers. These nationally representative norms were used as our second developmental benchmark for evaluating the performance of preschoolers with CIs relative to typically developing, normally hearing peers. Not all children received each EOI measure due to attention, fatigue, or time constraints; therefore, group sample sizes for each measure are included in the description. Even when accompanied by verbal directions, all test directions were also visually demonstrated for the children; only a nonverbal response was required for all tests.

Short-term/working memory—The Memory for Designs subtest of the NEPSY–II (Korkman, Kirk, & Kemp, 2007) is a nonverbal visual memory test that requires subjects to remember the location of visual designs on a grid. One child with an implant and one child with NH had difficulty understanding the task and did not receive a score (CI, $n = 23$; NH, $n = 20$). Children look at a picture of a 4×4 grid with designs in specific locations and attempt to reproduce the grid from memory on a blank card. Scores on Memory for Designs are expressed as scaled scores. The norms for the NEPSY–II are based on a nationally representative sample of 1,200 typically developing children ages 3 to 16.

Inhibition-concentration-vigilance—The Attention Sustained subtest of the Leiter International Performance Scale—Revised (Roid & Miller, 1997) is a nonverbal timed cancellation test that requires children to identify and cross out a target picture (e.g., flower) within a larger visual stimulus array of other background pictures (e.g., flowers, butterflies, mushrooms). Two children with CIs did not understand the task and did not receive a score (CI, $n = 22$; NH, $n = 21$). A total score is calculated by subtracting the number of errors from the number of correct identifications of the target picture. Raw scores are converted to scaled scores. The norms for the Leiter–R are based on a nationally representative sample of 1,719 typically developing children and young adults ages 2 to 20.

Organization-integration—The Beery Developmental Test of Visual-Motor Integration (Beery VMI; Beery & Beery, 2004) is a measure of visual-motor reproduction of designs of increasing complexity. All children were able to complete this subtest (CI, $n = 24$; NH, $n = 21$). The Beery VMI was developed to assess the extent to which children can integrate their visual and motor abilities. Children are asked to either reproduce/imitate a visual design drawn by the examiner or to copy a design from a picture, using paper and pencil. Raw scores are converted to standard scores. The norms for the Beery VMI are based on a nationally representative sample of 2,512 typically developing children ages 1 to 18.

Parent-reported EOI skills—The Behavior Rating Inventory of Executive Function (BRIEF for age 6; Gioia, Isquith, Guy, & Kenworthy, 2000) and BRIEF–Preschool Version (BRIEF-P for ages 3–5; Gioia, Espy, & Isquith, 2003) are parent-report behavior checklists that measure several domains of executive function behaviors in everyday life. Parents are asked to rate how often behaviors related to EF have been problematic for their child in the past 6 months by selecting Never, Sometimes, or Often. The Inhibitory Control, Working Memory, and Planning/Organizing sub-scales of the BRIEF and BRIEF-P were included in the present study because they represent the parent-report counterparts to the performance measures of EOI (additional BRIEF subscales that are not on the BRIEF-P were not included in this study). The BRIEF provides a more ecologically valid measure of how difficulties in EF play out in everyday life, compared with the typical performance measures of EF obtained from clinical neuropsychological assessments. Two parents of children with CIs and one parent of a child with NH did not complete the BRIEF (CI, $n = 22$; NH, $n = 20$). Respondents for both groups of children were most frequently the mother, with two father and one grandmother respondents for the CI group and three father respondents for the NH group. BRIEF/BRIEF-P subscale T scores were used to measure parent-reported EOI skills. The norms for the BRIEF-P are based on a nationally representative sample of 460 typically

developing children ages 2–5, and norms for the BRIEF are based on a large sample of 604 boys and 815 girls ages 5–18 with no history of special education or psychotropic medication usage.

Language—The Preschool Language Scale, Fourth Edition (PLS–4; Zimmerman, Steiner, & Pond, 2002) is a standardized assessment of general language ability for children ages birth to 6 years, 11 months. Responses for individual items on the PLS–4 are obtained through caregiver report, spontaneous observation during interactions with the examiner, and/or responses elicited during the evaluation. The PLS–4 was administered in the child's current communication mode, and children using total communication were able to respond using sign or spoken response or a combination of both. One child with CI did not receive the PLS–4 because of fatigue (CI, $n = 23$; NH, $n = 21$). The Total Language Score, which is a standard score with a mean of 100 and standard deviation of 15, was used in all analyses. The norms for the PLS–4 are based on a nationally representative sample of 1,564 children ages 2 days to 6 years, 11 months.

Measure of global nonverbal ability—The Picture Similarities subtest of the Differential Ability Scales (Elliott, 2007) is a nonverbal reasoning task used to assess global nonverbal intelligence (IQ). Children are shown a row of four pictures and are asked to place a picture card under one of the four pictures on the display that shares an element or concept. Raw scores are converted to T scores.

Data Analysis

Scores on the three performance and three parent-report measures of EOI skills were compared with two different developmental benchmarks. First, EOI scores from the CI group were compared with the NH group using independent samples t tests. Next, in order to compare the CI group's performance with well-established benchmarks for typical development, EOI scores from the CI sample were compared with scale norms for the test batteries (which are based on nationally representative samples with typically developing, normally hearing children) using one-sample t tests. Finally, Pearson product–moment correlations were calculated to investigate the relations between EOI scores and demographic/hearing-history variables and language ability.

Results

Comparison With Control Group

The CI and NH groups did not differ in age, $t(43) = 0.535$, $p = .595$; sex ($p = .525$, using Fisher's exact tests); parent's income level, $t(40) = 0.979$, $p = .333$; or nonverbal IQ, $t(41) = 1.432$, $p = .160$. Although no differences were observed between the two groups in income level, which can be considered a measure of socioeconomic status, four children with CIs came from families where the average income was less than $25,000 annually, whereas none of the families of children with NH reported incomes in this range. The CI group had significantly poorer general language skills than the NH group, $t(42) = 6.77$, $p < .001$.

On the performance measures of EOI, children with CIs scored lower than those with NH on the inhibition-concentration measure, $t(41) = 4.98$, $p < .001$; no significant group differences were found on measures of visual memory, $t(41) = 1.41$, $p = .166$, or organization-integration, $t(43) = 1.34$, $p = .189$. (See Table 3 for means and standard deviations.) On the BRIEF, compared with children with NH, children with CIs were rated by their caregivers as having significantly more problem behaviors on the Inhibit, $t(40) = 2.78$, $p = .008$, and Working Memory subscales, $t(40) = 2.14$, $p = .039$. However, no significant group differences were observed on the Plan/Organize subscale, $t(40) = 1.13$, $p = .263$.

In order to better understand the impact of individual children on group means and to offer a clinical interpretation of EOI performance in the preschoolers, the percentage of preschoolers in each group with EOI scores in the clinical range is also presented in Table 3. The percentage of preschoolers with CIs with scores in the clinical range is greater than preschoolers with NH across all performance and parent-report measures of EOI, with the exception of the Plan/Organize subscale of the BRIEF. One quarter of all preschoolers with CIs fall within the clinical range on Attention Sustained compared with zero NH preschoolers. Almost half the preschoolers with CIs fell within the clinical range on parent-reported problems with Inhibitory Control and Working Memory compared with 15% and 30% of preschoolers with NH.

Although the CI and NH groups did not differ significantly on nonverbal IQ, in order to account for potential differences in fluid intelligence, additional analyses comparing the groups were run using analyses of covariance, with nonverbal IQ as a covariate. Results were similar to the findings obtained with the *t* tests with the exception of parent-reported Working Memory, which showed a nonsignificant trend between the groups after controlling for nonverbal IQ, $F(1, 37) = 3.127$, $p = .085$.

Correlations between language and all EOI measures revealed significant associations for the CI group but not the NH group and only for parent-reported EOI in contrast to the performance tests of EOI (although the correlation between the Attention Sustained test and language nearly reached the cutoff for statistical significance, $p = .053$) (Table 4). Children with poorer language were reported by their parents to have more problems related to Working Memory ($r = -.533$, $p = .013$) and to Planning/Organizing ($r = -.524$, $p = .015$). However, controlling for language level, group differences in Attention Sustained remained significant, $F(1, 39) = 4.782$, $p = .035$, whereas group differences in parent-reported Working Memory and Inhibitory Control were no longer significant, $F(1, 38) = 0.313$, $p = .579$, and $F(1, 38) = 0.290$, $p = .593$, respectively.

Comparison With Scale Norms

Relative to published national norms for each test administered, children with CIs had significantly poorer performance on the Attention Sustained (inhibition-concentration measure) subtest of the Leiter–R, $t(21) = 2.19$, $p = .040$, but did not show significant differences on measures of visual memory; Memory for Designs on the NEPSY–II, $t(22) = 0.08$, $p = .934$; or organization-integration, Beery VMI, $t(23) = 0.44$, $p = .667$. Caretakers of children with CIs reported significantly more problems with Inhibitory Control and Working Memory compared with the normative sample of the BRIEF/BRIEF-P, $t(21) = 3.29$, $p = .$

003, and $t(21) = 3.92$, $p < .001$, respectively; no significant differences were found between children with CIs and the norms on the Plan/Organize subscale, $t(21) = 1.04$, $p = .309$.

Relationship Between Demographic and Hearing-History Variables and EOI Scores

Correlations among all EOI measures and CI participant characteristics revealed only one statistically significant relationship. Longer duration of CI use was related to fewer problems with planning and organization ($r = -.580$, $p = .006$) based on the parent-reported Plan/Organize scores.

Discussion

Preschool-age children with CIs demonstrated poorer performance on measures of inhibition and concentration and were reported to have significantly more problems related to inhibitory control and working memory by their parents when compared with two sets of developmental benchmarks: (a) age-matched peers with NH drawn from the same recruitment sites and (b) published nationally representative age-based norms for subscales of tests administered to participants. Inhibitory control includes the child's ability to resist impulses, to stop a behavior or thought in order to respond, to sustain attention/focus, and to control interference that comes from competing stimuli in the environment (Barkley, 1997b). Problems with inhibitory control, such as not noticing when one's behavior is bothering other people, continually acting silly or out of control, or being easily distractible, have functional consequences at home and in preschool or child care that can adversely impact early social and learning experiences (Gioia et al., 2003; Watson & Bell, 2013). Working memory reflects a child's ability to maintain information in mind and manipulate information in immediate conscious memory for the purpose of completing a future task (Best & Miller, 2010). Problems with working memory in the everyday life of preschool-age children include having trouble carrying out instructions with more than one step, having difficulty finishing tasks, and forgetting what to do in the middle of an activity, all of which have functional consequences for early learning (Gioia et al., 2003).

We found that a greater percentage of children with CIs fell within the clinically significant range across all measures of EOI compared with children with NH, with the exception of parent-reported problems related to Planning/ Organizing where the percentages were about equal. Almost half the children with CIs scored in the clinical range on parent-reported problems with Inhibitory Control and Working Memory. So although significant group differences were not found across every EOI measure assessed in this study, the finding that such a high percentage of children with CIs score in the clinically significant range, much higher than that of children with NH, provides strong evidence that EOI skills are already at risk in preschool-age children with CIs. The present study is the first investigation to demonstrate that the EOI deficits found in older children and young adults with CIs (Beer et al., 2011; Beer et al., 2010; Kronenberger, Pisoni, Henning, Colson, & Nguyen, 2012; Pisoni, Kronenberger, Henning, & Colson, 2011) begin to emerge as early as preschool ages. This is a highly significant theoretical and clinical finding because it indicates that the emergence of several specific EOI deficits occurs at substantially younger ages than have been investigated in past research. As a result, monitoring and tracking of these critical areas

of potential risk should begin at much younger (e.g., preschool) ages than previously assumed in the past.

Preschoolers with CIs did not demonstrate any deficits or delays in performance measures of visual memory or organization-integration compared with peers with NH and normative benchmarks. There are several potential explanations for these findings. First, it is very likely that a period of profound deafness followed by degraded auditory experiences from a CI impacts some types of EOI skills more than others, placing some EOI skills at a higher risk for atypical development than others regardless of age. This explanation alone, however, is unlikely to fully explain all of the present results, given findings of deficits in broader areas of memory, organization, and integration found in older children with CIs (Pisoni et al., 2011). Alternatively, differences in the developmental timing of various types of EOI skills may place some skills at greater risk during preschool ages, whereas other skills may not be affected until later ages. For example, the EOI skills required to control concentration and inhibition may develop earlier than complex visual memory and organization skills, placing the former skills at greater risk than the latter skills during the preschool ages (Garon et al., 2008; Miyake et al., 2000). In addition, it is possible that other EOI risks are actually present during preschool ages but are not easily measurable at these younger ages with the testing instruments available.

Another important factor in understanding the lack of differences found between the scores of children with CIs and developmental benchmarks in visual memory and organization-integration is the method of measurement of these abilities. In the present study, visual memory was measured with a simultaneous, holistic, visuospatial memory test involving visually presented designs. Differences in memory processes found between children with CIs and developmental benchmarks at older ages have all used sequential, span-based tests (Cleary, Pisoni, & Geers, 2001; Pisoni & Cleary, 2004) . Interestingly, results from a subgroup of the current CI sample, which was administered an extended battery of tests that included a sequential span-based memory test from the Leiter–R (Forward Memory), did reveal differences when compared with the controls, $t(28) = 2.793$, $p = .009$, with 15% of children with CIs and 5% of children with NH falling into the clinical range (i.e., 1 *SD* below the mean). However, this extended battery was administered only to children who were able to continue after the core battery of EOI tests were completed; this subgroup of children is a smaller sample consisting of 13 children with CIs and 17 with NH, who may have differed from children who were unable to complete additional tests. Therefore, these results should be viewed with caution. Nevertheless, these findings raise the possibility that differences related to sequential memory span might be present at preschool ages (Conway, Pisoni, Anaya, Karpicke, & Henning, 2011; Conway et al., 2009; Pisoni & Cleary, 2004).

EOI skills were generally unrelated to demographic and hearing-history variables, with the exception that longer duration of implant use was related to fewer parent-reported problems with planning and organizing. The types of behaviors indicative of planning and organizing skills on our parent-report measure of EF skills involve planning and/or completion of tasks in a sequential and ordered manner and are likely to be influenced by language skills as both are sequentially ordered. Therefore, planning and organizing skills may be particularly sensitive to the development of linguistic and sequential processing after cochlear

implantation, resulting in a positive relationship between length of implant use and planning and organizing skills. Other hearing-history variables such as age at implantation, pre-implant PTA, and age at testing were unrelated to EOI scores, although the range of these variables was very limited within the study sample. Differences between the CI and NH groups in nonverbal IQ did not account for the differences observed in EOI scores between the groups. This pattern suggests that differences in demographic or intellectual ability alone cannot explain the EOI weaknesses found in the present CI group.

General language ability as measured by the PLS–4 was unrelated to any of the EOI measures in preschoolers with NH. However, language was significantly correlated with parent-reported EOI but not behaviorally based performance measures of EOI in children with CIs; the parents of children with lower language skills reported more difficulty in everyday situations that involved working memory and planning and organizing skills. When language was entered as a covariate, group differences in the performance measure of inhibition and concentration (Attention Sustained) remained significant; however, parent-reported working memory and inhibitory control were no longer significant. It is possible that group differences on the Attention Sustained subtest remain even after controlling for differences in language because difficulties with inhibition and concentration that require the active control of attention are highly robust in preschoolers with CIs and that good language skills are not enough to buffer them against the impact of early auditory deprivation and degraded auditory input. Longitudinal studies are needed, however, to explain how continued development of both language and inhibition-concentration skills throughout childhood may impact their influence on one another.

The finding that language was unrelated to performance measures of EOI and that significant group differences in EOI as reported by parents were no longer significant after controlling for language suggests that language plays different roles across the two types of EOI assessments used in this study. Parent-reported EOI provides us with an ecologically valid measure of the functional consequences of a deficit in executive function as evidenced in everyday real-world behaviors. Parents' ratings of the behaviors on the BRIEF/BRIEF-P are much more likely to be mediated by their child's language ability than performance measures of EOI obtained in the clinic. Many of the BRIEF/BRIEF-P items ask parents to rate behaviors that are often exhibited using language (e.g., acting out of control) or are requested using language (e.g., cannot remember all things when given more than one thing to do). Nevertheless, even performance tasks such as the Attention Sustained subtest used to measure inhibition-concentration-vigilance are likely to be mediated by language to some degree, as children may use private speech to guide their performance (Fernyhough & Fradley, 2005; Vygotsky, 1986).

Detection of EOI deficits as early as preschool age has important clinical and educational implications. The primary focus of habilitation after implantation for families who choose oral communication is typically skill-based speech and language development (e.g., articulation, vocabulary, and syntax). Assessment and treatment of deficits in EOI processes such as working memory, inhibition, concentration, organization, flexibility, and speed are less often considered in conventional treatment plans developed by clinical CI teams, even though preschool programs designed to strengthen EF skills, and interventions and training

programs to improve working memory, are proving to yield benefits to children at risk for adverse outcomes (Diamond & Lee, 2011; Raver et al., 2011; Röthlisberger, Neuenschwander, Cimeli, Michel, & Roebers, 2011). The Tools of the Mind curriculum, for example, developed by two educators and developmental scientists (Bodrova & Leong, 2007) based on the research of Russian sociocultural psychologist Lev Vygotsky (1978), reports impressive gains in the development of EF skills in at-risk preschoolers through the use of specific behavioral techniques designed to scaffold early executive skills—these techniques range from holding a drawing of an ear to remind a child of his or her active role as the listener in child–child book reading, to pre-planning a bout of dramatic play with a friend using spoken language and drawings, to encouraging the use of private speech to self-regulate behaviors (Barnett et al., 2008; Diamond, Barnett, Thomas, & Munro, 2007).

The PATHS curriculum (Promoting Alternative THinking Strategies; Kusche & Greenberg, 1994)—designed to improve self-control; increase emotional awareness; and integrate emotion, language, cognition, and behavior in at-risk children—was implemented in a classroom of 29 children with severe to profound deafness who used hearing aids and total communication (Greenberg & Kusche, 1998). After 1 year, children in PATHS showed improvements in social problem solving, impulsivity, emotional adjustment, frustration tolerance, and reading comprehension. Finally, several computer-based training programs to improve inhibitory control and working memory have also been shown to improve EF skills in preschool-age children (Thorell, Lindqvist, Bergman Nutley, Bohlin, & Klingberg, 2009) and school-age children with CIs (Kronenberger, Pisoni, Henning, Colson, & Hazzard, 2011).

Evidence of the foundational roles of parent–child interactions (e.g., maternal scaffolding behaviors during problem solving) and family environments (e.g., family chaos and inconsistent parenting) for supporting neurocognitive development is also rapidly growing (Bernier, Carlson, & Whipple, 2010; Bibok, Carpendale, & Muller, 2009; Carlson, 2009; Hughes & Ensor, 2009), based on the principle that the family serves as an integral source for targeted intervention. A recent family study from our center investigated the impact of the family environment on executive function in children with CIs and found that families placing a high emphasis on personal achievement and greater organization in the home reported fewer problems related to executive function in their children (Holt, Beer, Kronenberger, Pisoni, & Lalonde, 2012). A second study showed that families of preschool-age children with CIs who reported higher levels of support and lower levels of conflict in the home also reported fewer problems with emotional control in their children (Rubinstein, 2002). There is increasing evidence that early executive control is highly predictive of a wide range of short-term and long-term academic, social, and health outcomes throughout the life span (Kusche & Greenberg, 1994); hence, identification and intervention efforts to address EOI risks and vulnerabilities during preschool may have wide-ranging impacts on quality of life longitudinally for children with CIs.

Our study is the first investigation to assess EOI processes at preschool ages. It provides the first empirical evidence that delays and/or disturbances in speech and language alone should not form the sole basis for decision making about the needs for special services or interventions for preschoolers with CIs. Increasing academic demands that begin at early

school ages are known to rely heavily on robust EOI skills developed in preschool, and these more complex and sophisticated academic endeavors (e.g., reading comprehension, writing, and multistep math) are delayed in children with CIs (Bull et al., 2008; Geers & Hayes, 2011; Montgomery et al., 2010). Routine assessment and careful monitoring of EOI skills in children with CIs beginning in preschool would allow for more individualized and targeted interventions of specific executive skills in preschool, at home, and in speech-language therapy that support speech and language development.

One strength of the present study is the use of both performance-based neurocognitive measures and ecologically valid parent-report ratings of executive function. The convergence of significant findings across both types of measures adds confidence that the differences in EOI abilities found using performance measures also have functional consequences as evidenced by parent report of significant problems with inhibitory control and working memory in everyday life. In addition, significant findings across both types of measures provide replication that adds confidence to the validity of the findings. Another strength of the present study is the use of two developmental benchmarks—scores of a group of children with NH and normative data from nationally representative samples for the test batteries administered—as comparison data for assessing EOI functioning of children with CIs. The convergence of significant and nonsignificant differences and similarities across both types of benchmarks also attests to the robust nature of the present EOI results—the children with CIs who performed more poorly than NH children on measures of inhibition and concentration also performed more poorly compared with the normative benchmarks.

Some limitations of the present study are the small sample size and the young age of children in the study, which limited the complexity, number, and variety of measures of EOI functioning available. In addition, both the CI and the NH groups had above-average nonverbal IQ scores. Future research should broaden the assessment of EOI functioning in preschool children with CIs who have lower nonverbal IQ scores and who are matched with children with NH of similar nonverbal IQ levels. In addition, because these deficits occur so early in life and evidence for relations between family environments and the development of executive function is increasing, future research should also focus on the identification of specific family factors and behaviors that support neurocognitive development to ultimately inform individualized and targeted early intervention possibilities and to explore these relationships in larger, more culturally diverse samples. Finally, longitudinal data will be critically important for investigating the emergence and developmental trajectory of EOI skills across the preschool ages in the same group of children. A longitudinal study is currently under way in our center, and reports of the developmental results will be forthcoming (Castellanos et al., 2013).

Conclusion

The development of EOI processes such as working memory and inhibition-concentration may be impacted by a period of auditory, speech, and language deprivation, followed by exposure to degraded auditory input received from a CI, placing children with early hearing loss at high risk for deficits in several specific areas of EOI processing. This study found

that preschool-age children with severe to profound prelingual deafness who use CIs were significantly delayed in attention and inhibitory control and parent-reported working memory when compared with a control group of children with NH and compared with national norms using both performance and parent-report measures. Children were not delayed in EOI processes related to holistic visual memory or visual organization-integration, but evidence was found for delays in sequential working memory. In addition, a greater percentage of preschoolers with CIs fell within the clinically significant range compared with preschoolers with NH across all EOI domains assessed in this study, with the exception parent-reported problems related to Planning/Organizing, with close to half the CI group falling in the clinically significant range on parent-reported problems with Inhibitory Control and Working Memory. This is the first study to document that the differences in EOI processing found in school-age children with CIs emerge as early as the preschool years.

Acknowledgments

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Table 1

Participant characteristics.

Variable	Cochlear implant (*n* = 24)		Normal hearing (*n* = 21)	
	n	% of sample	*n*	% of sample
Bilateral/bimodal CI				
Bilateral CI	17	70.83		
Bimodal (CI + hearing aid)	1	4.16		
Unilateral CI	6	25.0		
Gender				
Female	10	42	8	38
Male	14	58	13	62
Race				
Black or African American	2	8	3	14
Multiracial	2	8	1	5
White	20	83	17	81
Ethnicity				
Hispanic	1	4	0	0
Non-Hispanic	23	96	21	100

Note. CI = cochlear implant.

Table 2

Participant characteristics by hearing status.

Variable	Preschoolers with CI (*n* = 24)		Preschoolers with NH (*n* = 21)	
	M (SD)	Range	*M (SD)*	Range
Age at implantation (months)	20.01 (7.87)	10.41–36.60		
Age at hearing aid fit (months)	8.65 (8.19)	1–30		
Duration of implant use (years)	2.73 (1.14)	0.53–5.18		
Preimplant residual hearing (PTA)[a]	99.79 (13.33)	73.33–118.43		
Communication mode[b]	4.75 (0.85)	2–5		
Chronological age (years)	4.36 (1.14)	3.13–6.94	4.19 (1.05)	3.21–7.0
Income level[c]	6.81 (3.20)	1–10	7.57 (1.57)	5–10
Nonverbal IQ[d]	53.45 (10.63)	33–81	58.52 (12.54)	41–78
Language[e]	77.96 (24.33)	50–128	117.62 (11.86)	91–138

Note. NH = normal hearing.

[a]Unaided pure-tone average (PTA) in the better ear for the frequencies 500, 1000, and 2000 Hz in dB HL.

[b]Communication mode is coded on a scale from *mostly sign* (coded 1) to *auditory-verbal* (coded 6) with a code of 4 = cued *speech*.

[c]Income level is coded on a scale from *under $5,000* (coded 1) to *$95,000 and over* (coded 10) with a code of 6 = *$35,000-$49,999* and a code of 7 = *$50,000-$64,999*.

[d]T score from the Differential Ability Scales, II Picture Similarities subtest.

[e]Standard score from Preschool Language Scale, Fourth Edition Total Language Score.

Table 3

Differences in executive-organizational-integrative (EOI) processes between children with cochlear implants and children with normal hearing.

Measure	Preschoolers with CI		Preschoolers with NH	
	M (SD)	% in clinical range	*M (SD)*	% in clinical range
Performance measure				
Attention sustained[a]***	8.95 (2.24)	27.3	12.43 (2.34)	0
Memory for designs[b]	9.96 (2.50)	17.4	11.05 (2.59)	5.0
Visual-motor integration[c]	101.54 (17.31)	12.5	107.38 (10.75)	4.8
Parent-report measure				
Inhibitory control[a]**	59.32 (13.28)	45.5	49.60 (8.65)	15.0
Working memory[b]*	60.55 (12.61)	45.5	52.85 (10.48)	30.0
Plan/organize[c]	52.68 (12.06)	22.7	48.75 (10.21)	25.0

Note. Clinical range is one standard deviation below the mean for performance measures and one standard deviation above the mean for parent-report measures.

[a]EOI domain = inhibition-concentration-vigilance.

[b]EOI domain = memory.

[c]EOI domain = organization-integration.

*
$p < .05$.

**
$p < .01$.

***
$p < .0001$.

Table 4

Pearson correlations between language and EOI processes for children with cochlear implants and normal hearing.

| EOI measure | Language[a] | | | |
| | Preschoolers with CI | | Preschoolers with NH | |
	r	P	r	P
Attention sustained	.428	.053	.068	.771
Memory for designs	.113	.617	−.338	.146
Visual-motor integration	.118	.591	.211	.360
Inhibitory control	−.409	.066	−.052	.827
Working memory	−.533	.013[*]	−.239	.309
Plan/organize	−.524	.015[*]	−.102	.669

[a]PLS–4 Total Language Score.

[*]$p < .05$.