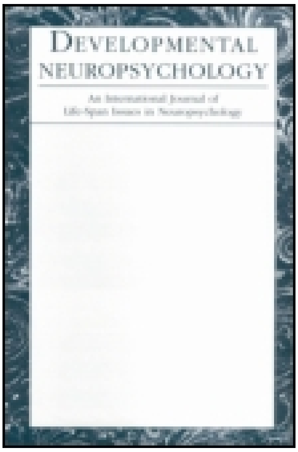

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Nonverbal Cognition in Deaf Children Following Cochlear Implantation: Motor Sequencing Disturbances Mediate Language Delays

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We assessed profoundly deaf children with cochlear implants (CIs) ($N = 24$) and age-matched normal-hearing children ($N = 31$) on several nonverbal cognition measures: motor sequencing, tactile discrimination, response inhibition, visual-motor integration, and visual-spatial processing. The results revealed that the children with CIs showed disturbances solely on motor sequencing and that performance on this task was significantly correlated with scores on the Clinical Evaluation of Language Fundamentals, 4th Edition (CELF–4). These findings suggest that a period of auditory deprivation before cochlear implantation affects motor sequencing skills, which in turn may mediate the language delays displayed by some deaf children with CIs.

Deaf children with cochlear implants (CIs) represent an important clinical population to better understand and study, for both applied health and theoretical reasons. Although many children with CIs are able to develop age-appropriate speech and language abilities, some children obtain little benefit other than the awareness of sound from their implant (American Speech-Language-Hearing Association, 2004). Very little is currently known about general cognitive development

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in this population following implantation, and whether particular cognitive abilities can help explain the enormous variation in language outcomes. Whereas a great deal of research has been devoted to understanding hearing and spoken language development in deaf children with CIs, comparably less research has been focused on factors having to do with non-auditory and nonverbal functions. Because the brain is an integrated functional system (see Luria, 1973), it is likely that a period of auditory deprivation occurring early in development may have secondary cognitive and neural sequelae in addition to the obvious hearing-related effects. For this reason it is important to investigate the non-auditory and nonverbal cognitive abilities in deaf children with CIs.

One cognitive domain that may be fruitful to study is sequencing and serial order behavior (Lashley, 1951). It has been previously suggested that the sense of hearing is intimately bound with time and the perception of serial order (Hirsh, 1967); if so, then a lack of experience with sound during early development may impact cognitive functions having to do with perceiving, manipulating, and organizing temporal sequences of nonverbal stimuli, thoughts, and actions. A deaf or hearing-impaired person has less experience with common auditory features that provide important markers for the passage of time, such as the ticking sounds made by a clock, the ringing of a telephone, or the sound of another person's footsteps coming from around the corner (cf. Ramsdell, 1947; Rileigh & Odom, 1972). Indeed, this function of hearing—automatically detecting *changes* to events occurring in the background—may be so basic and fundamental to hearing individuals that it is simply taken for granted. Sound also provides a signaling function that helps a listener predict what will occur next even when visual information is not available, as in the example of approaching footsteps from around a corner. This characteristic of sound as being a carrier of temporal events implies that lack of experience with sound may have a profound impact on cognitive development beyond the obvious hearing-related sensory effects. Indeed, anecdotal reports suggest that deafness, more so than blindness, results in the subjective impression that time itself seems to stand still, leading to a profound feeling of living in a "dead" world (Ramsdell, 1947).

If deafness results in disturbances to certain selective aspects of nonverbal and non-auditory cognition, it becomes crucial to investigate a broad range of neuropsychological measures in deaf children with CIs, not just skills related to auditory perception, speech perception, and spoken language development. In this article, we explore several measures of nonverbal abilities in deaf children following cochlear implantation, including motor sequencing, visual-spatial memory, visual-motor integration, tactile discrimination, and response inhibition. Importantly, although all measures focus on nonverbal and non-auditory functions, one measure in particular relies heavily on sequencing skills, whereas several others are primarily spatial and/or non-sequential (visual-spatial memory; tactile discrimination; response inhibition). Before describing the study in more detail, we first review previous research exploring nonverbal abilities in deaf and hearing-impaired populations.

NONVERBAL ABILITIES IN THE DEAF

It is now well-established that deaf children with CIs are impaired on verbal short-term memory (Dawson, Busby, McKay, & Clark, 2002; Marschark, 2006; Pisoni et al., 2008; Pisoni & Cleary, 2003, 2004; Pisoni & Geers, 2001). Furthermore, deaf children with CIs also show slower verbal rehearsal speed and short-term memory scanning rates for verbal material (Burkholder & Pisoni, 2003, 2006). This work suggests that even after restoring hearing via a CI, a period of auditory deprivation impacts the ability to encode, manipulate, rehearse, and remember verbal material. Reduced exposure to sound and spoken language appears to limit one's ability to represent and manipulate phonological representations of speech in working memory, an idea referred to as the *linguistic coding* or *inner speech* hypothesis (see Marschark, 1993).

However, additional research has also suggested that deafness may affect nonverbal serial memory as well. A small handful of studies have attempted to compare nonverbal short-term memory in deaf versus hearing populations using non-linguistic visual or gestural stimuli that are not easy to code verbally (e.g., abstract designs, shapes, or hand gestures). For instance, Todman and Seedhouse (1994) assessed hearing children and age- and nonverbal reasoning-matched profoundly deaf children on a short-term memory task requiring the production of particular visual-action codes (e.g., open-mouth, bang-table). The deaf children were significantly worse when the constraints of the task required the action codes to be recalled in correct serial order; however, when the task allowed children to recall the action codes in a free recall format, the deaf children actually surpassed their hearing peers. Interestingly, other research also suggests that deaf individuals may perform better than hearing individuals on tasks requiring visual-spatial cognitive processing (e.g., see Bavelier, Dye, & Hauser, 2006; Marschark, 2006). This pattern suggests that deaf children may have a specific disturbance with encoding and/or recalling the *serial order* of items in memory, even when verbal codes are not actively used.

Several other studies have shown that deafness may be associated not just with disturbances to visual memory and learning for serial stimuli, but also with more general visual-motor abilities relating to the temporal organization of behavior. For example, Savelsbergh, Netelenbos, and Whiting (1991) found that deaf children were impaired on ball-catching ability, suggesting that a lack of auditory stimulation during early development can lead to disturbances to the development of complex movement coordination. Summarizing results from several studies, Wiegersma and Van der Velde (1983) concluded that deaf children were impaired on general dynamic motor coordination and the execution of movements. Similarly, Schlumberger, Narbona, and Manrique (2004) found that deaf children (both with and without CIs) showed delays in the development of complex motor sequence production.

Taken together, these findings suggest that a period of auditory deprivation may result in secondary disturbances to aspects of nonverbal cognition, especially skills related to the representation and organization of sequences of visual and visual-motor patterns (Conway, Pisoni, & Kronenberger, 2009). Furthermore, even after having their hearing restored via a CI, it is very likely that many deaf children with CIs still suffer delays in the development of neural circuits that underlie these information processing systems. Thus, differences resulting from neural and cognitive reorganization due to deafness may be at least partly responsible for the enormous variability observed in speech and language outcome measures following implantation.

Consistent with this idea, some initial research has indeed shown that selective aspects of nonverbal cognitive abilities—especially those involving the temporal organization of behavior—may be linked to the development of spoken language skills in deaf children with CIs. Horn, Pisoni, Sanders, and Miyamoto (2005) and Horn, Pisoni, and Miyamoto (2006) found that pre-implant measures of fine motor development can predict post-implant speech and language outcomes in profoundly deaf children who have received a CI. Similarly. Knutson et al. (1991)

found that the ability to detect and respond to visual input sequences accounted for a substantial amount of language variance in post-lingually deaf adults with cochlear implants. These results suggest that perceptual-motor abilities and spoken language acquisition are closely coupled processes in both deaf children and adults with CIs (cf. Broesterhuizen, 1997; Lenneberg, 1967; Locke, Bekken, McMinn-Larson, & Wein, 1995; Siegel et al., 1982). Furthermore, motor sequencing skills (as assessed by a finger tapping task) were found to be correlated with reading ability in unimpaired readers (Carello, LeVasseur, & Schmidt, 2002), suggesting that non-verbal sequencing skills are linked to aspects of language development more generally.

The objectives of the present study are to explore the effects of auditory deprivation on nonverbal cognition and to investigate whether individual variability in nonverbal cognitive abilities may be related to language outcomes after cochlear implantation. Given the considerations reviewed earlier, we predicted that deaf children with CIs would show a selective disturbance specifically on visual-motor sequencing abilities (as assessed by a fingertip tapping task) but not on visual-spatial and other non-sequencing cognitive functions. Furthermore, we predicted that this measure of visual-motor sequencing would also be significantly correlated with spoken language outcomes in the deaf children with CIs: the better children's performance on visual-motor sequencing, the better their spoken language outcomes will be.

METHOD

Two groups of children participated, a group of deaf children with CIs, and an age-matched group of normal-hearing (NH) children. All children were given several clinical neuropsychological tests that assessed aspects of nonverbal cognition. In addition, for both groups of children, we collected measures of verbal short-term memory and vocabulary acquisition to serve as covariates. We also used a global clinical language measure to assess language outcome in the CI children.

The deaf children with CIs were tested by a trained Speech Language Pathologist (4th author) at the DeVault Otologic Research Laboratory, Department of Otolaryngology, Indiana University School of Medicine, Indianapolis. The NH children were tested by the second author in a sound-attenuated booth in the Speech Research Laboratory at Indiana University, Bloomington. For both groups of children, the study consisted of a single session lasting 60–90 minutes, with breaks provided as needed. Before beginning the experiment, all NH children received and passed a brief pure-tone audiometric screening assessment in both ears.

Participants

Deaf children with CIs. Twenty-four prelingually, profoundly deaf children (9 females) who had received a CI by age 4 years were recruited through the DeVault Otologic Research Laboratory at the Indiana University School of Medicine, Indianapolis. All the children had profound bilateral hearing loss (90-dB or greater) and had used their implant for a minimum of 3 years. All subjects were native speakers of English and had hearing parents. All children had a single implant except for two children with bilateral implants and one child who had a hearing aid in the non-implanted ear. For the three children with bilateral hearing, testing was

TABLE 1
Participant Characteristics

	CI Children			*NH Children*				
Measure	*M*	*SD*	*Range*	*M*	*SD*	*Range*	*t(53)*	*p*
Sample size	24	—	—	31	—	—	—	—
Age	90.0	19.4	61–118	88.8	11.4	65–107	0.3	0.76
Age implant	21.0	8.2	10–39	—	—	—	—	—
CI duration	69.1	19.0	36–98	—	—	—	—	—
FD	4.9	1.6	2–8	7.2	1.9	5–12	−4.8	0.0001
BD	2.5	1.5	0–5	3.8	1.1	2–6	−3.8	0.0001
PPVT	85.5	12.1	59–107	114.7	11.9	90–139	−9.0	0.0001

Age is given in months; Age implant: age at cochlear implantation (in months); CI duration: duration of cochlear implant use (in months); FD: forward digit span score; BD: backward digit span score; PPVT: Peabody Picture Vocabulary Test scaled score.

conducted with only one CI activated (the original implant). Although several of the children had been exposed to Signed Exact English (SEE), none of the children relied exclusively on sign or gesture, and all children were tested using oral-only procedures. Aside from hearing loss, there were no other known cognitive, motor, or sensory impairments. Etiology of deafness included unknown ($N = 17$), genetic ($N = 3$), ototoxicity ($N = 1$), and mondini dysplasia ($N = 2$). Table 1 summarizes the demographic characteristics of these 24 children. For their time and effort, the children's parents/caregivers received monetary compensation.

Normal-hearing children. Thirty-one typically developing NH children (14 females) were recruited through Indiana University's "Kid Information Database" and through the Life Education and Resource Home Schooling Network of Bloomington, IN. All children were native speakers of English; parental reports indicated no history of a hearing loss, speech impairment, or cognitive or motor disorder. Table 1 summarizes the demographic characteristics of these children. For their participation, children received a small toy and their parents received monetary compensation.

Covariates

Two measures of verbal ability were collected, verbal short-term memory capacity and vocabulary development. In addition to chronological age, these two measures were treated as covariates in order to ensure that any group differences obtained on the nonverbal cognition measures were in fact not due to differences in underlying verbal abilities.

Verbal short-term memory. The forward and backward digit span task of the Wechsler Intelligence Scale–Third Edition (WISC–III) was used (Wechsler, 1991). In the forward digit span task, subjects were presented with lists of pre-recorded spoken digits with lengths (2–10) that became progressively longer. The subjects' task was to repeat each sequence aloud. In the backwards digit span task, subjects were also presented with lists of spoken digits with lengths that became progressively longer, but they were asked to repeat the sequence in reverse order.

Digits were played through an Advent AV570 loudspeaker at 65 dB; the child's responses were recorded by a desk-mounted microphone and scored for accuracy offline. Subjects received a point for each list that they correctly recalled in each digit span task. When a child incorrectly recalled both lists at a given list length, testing ended.

Generally, the forward digit span task is thought to reflect the involvement of processes that maintain and store verbal items in short-term memory for a brief period of time, whereas the backward digit span task reflects the operation of controlled attention and higher-level executive processes that manipulate and process the verbal items held in immediate memory (Rosen & Engle, 1997).

Vocabulary development. The Peabody Picture Vocabulary Test (PPVT) (3rd edition) is a standard measure of vocabulary development for ages 2 years and up (Dunn & Dunn, 1997). In this task, participants are shown four pictures on a single trial. They are prompted with a particular English word and then asked to pick the picture that most accurately depicts the word. For each child, a scaled score is derived based on comparison with a large normative sample.

Nonverbal Cognition: Experimental Measures

Several subtests from the neuropsychological instrument known as the NEPSY were used as experimental measures (Korkman, Kirk, & Kemp, 1998). The NEPSY is a clinical neuropsychological test battery designed to assess children's development (ages 3–12 years old) in several cognitive domains including attention/executive functions, visual-spatial processing, sensorimotor functions, and memory and learning (for review, see: Ahmad & Warriner, 2001). We used four NEPSY subtests that were chosen to focus on several aspects of nonverbal cognition (motor sequencing, response inhibition, tactile perception, and visual-motor integration) plus a visual-spatial learning and memory task from the Children's Memory Scale (CMS; Cohen, 1997). These tasks are described in more detail below.

Motor sequencing. The NEPSY "fingertip tapping" subtest (Korkman et al., 1998) was used as a measure of basic sensorimotor sequencing skill and finger dexterity. As per standard procedure, children were instructed to tap the index finger against the thumb 32 times as quickly as possible for each hand separately ("repetitive tapping" task). Children were also instructed to tap the fingers sequentially against the thumb from index to little finger as quickly as possible for each hand separately ("sequential tapping" task). Scores were derived based on the amount of time taken to complete each tapping task without errors. The scaled score was an aggregate of performance on all four tasks.

Tactile perception. The NEPSY "finger discrimination" task (Korkman et al., 1998) was used as a measure of tactile perception. As per standard procedure, on each trial the experimenter touched one or two fingers of the child's hand (testing preferred and non-preferred hands on separate trials), which the child cannot see due to an occluder. The child was instructed to indicate which finger or fingers were touched. All children received two scores, reflecting performance on preferred and non-preferred hands. Note that only the untransformed raw scores are reported because a derivation of scaled scores was not available in the NEPSY manual for this subtest.

Response inhibition. The NEPSY "knock and tap" subtest (Korkman et al., 1998) was used as a measure of manual response inhibition. On this subtest, the child learns a set of appropriate motor behaviors in response to the examiner's motor behavior. For instance, if the experimenter knocked on the table the child was required to tap on the table with his preferred hand; and if the experimenter tapped on the table, the child was to knock on the table. Later in the subtest, a new set of rules was introduced to the child: if the experimenter hit the side of her fist on the table, the child was to knock on the table, and if she knocked the child was to hit the side of his fist on the table. And if the experimenter tapped on the table, the child was to do nothing. The number of correct responses out of a total possible raw score of thirty trials was recorded. Note that only the untransformed raw scores are here reported because a derivation of scaled scores was not available in the NEPSY manual for this subtest.

Visual-motor integration. The NEPSY "design copy" task (Korkman et al., 1998) was used as a measure of visual-motor integration and visual-spatial processing. As per standard procedure, children were given a packet of 18 geometric designs and asked to copy each design in the space provided. The child was not allowed to erase any mistakes made, and was not allowed to turn the paper while drawing. Each design was scored on a four-point scale taking into consideration things such as angle, completeness, and proportion. Each of the child's design was scored based on standardized criteria.

Visual-spatial learning and memory. The "dot locations" subtest of the Children's Memory Scale (CMS; Cohen, 1997) was used as a measure of nonverbal visual-spatial learning and memory. As per standard procedure, the children were shown a picture of six blue dots inside a large white background. The dot pattern was presented to the child for five seconds before being taken out of sight. The child was then asked to reproduce the dot pattern from memory by placing six blue chips onto a 3×4 grid. The child was allowed to place the chips on the grid in any order and no time restriction was imposed. The final pattern produced by the child was recorded and no feedback was given on the child's performance. The chips were then cleared from the child's grid, and the same dot pattern was shown to the child again for five seconds and then taken out of view. The child was asked again to reproduce the dot pattern from memory. This process was repeated a third time, resulting in a total of three "learning" trials in which the same dot pattern was used. Next, a trial of red dots was presented and the child was asked to reproduce it. The red dot trial was not scored, but rather served as a distracter. The child was then asked to recall from memory the initial blue dot pattern that had been presented three times ("short delay" trial). At the conclusion of the experiment (after a delay of approximately 30 minutes), the child was asked once more to reproduce the blue dot pattern from memory ("long delay" trial). The child's pattern reproductions were scored based on total number of chips placed correctly on the grid. Therefore, a child could receive a total raw score of up to 24 points for the immediate recall section, and up to six points for the delayed recall portion of the task. As per standard procedure, the raw scores were then converted into three separate scaled scores, taking into account the age of the child: visual-spatial learning score (sum of scores on trials 1–3); visual-spatial "total" score (sum of scores on trials 1–3 plus short delay trial); visual-spatial "long delay" score (score on the long delay trial).

Language Outcomes

As part of the CI children's regular annual visits to the Department of Otolaryngology, 18 of the 24 children were assessed on three core subtests of the Clinical Evaluation of Language Fundamentals, 4th Edition (CELF–4), an assessment tool for diagnosing language disorders in children aged 5 to 21 years (Semel, Wiig, & Secord, 2003). The remaining six children did not have scores available because they were below the minimum age requirement for the CELF–4 at the time of testing.

These three subtests of the CELF–4 measure aspects of general language ability: Concepts and Following Directions (C&FD), Formulated Sentences (FS), and Recalling Sentences (RS) (see Paslawski, 2005, for a review and description of all subtests). For each child, a single combined scaled score ("Core Language Score") is derived based on performance on these three tasks, where the age-scaled mean score is 100.

In order to assess whether individual differences on any of our experimental measures were associated with language outcome as measured by the Core Language Score of the CELF–4, we calculated partial correlations while controlling for the common variance associated with chronological age, forward and backward digit spans (i.e., "total" digit span, which is a sum of the scores on forward and backward spans), and vocabulary scores. For all correlations, raw scores were used. In order to reduce the undesirable effect that one or two unusually low or high performing children might have on the validity of the correlation results, outliers for each task, defined as +/–2 standard deviations from the mean, were not included, resulting in two CELF–4 scores not being included in the final analysis.

RESULTS

Covariates

As Table 1 shows, the NH children's chronological age was well-matched relative to the CI children. However, the NH children exceeded the CI children on their forward and backward digit spans and receptive vocabulary scores. For the main analyses reported below, these three dependent measures were treated as covariates, in order to ensure that any group differences obtained on the nonverbal cognition measures were in fact not due to differences in verbal short-term memory capacity or vocabulary development.

Nonverbal cognition: Experimental measures. Table 2 shows a summary of the scores for each of the five experimental tasks (eight measures total, due to several of the tasks having multiple scores). Whenever possible, scaled scores are reported and one-sample *t*-tests were conducted to compare each score to the respective normed baseline score (10). We report untransformed raw scores for the finger discrimination and knock and tap subtests because the NEPSY manual does not provide scaled scores on these tasks. The table also shows the results of an independent-samples *t*-test comparing performance between the two groups on each measure.

For the NH children, in all cases where scaled scores are available, their scores were at the expected level, or, for three of the measures, they were actually significantly better than the norms: design copy, visual-spatial (total), and visual-spatial (long delay). On the other hand,

TABLE 2
Experimental Measures: Group Means and Comparison to Norms

	CI Children				*NH Children*				*Group Cmp*
Measure	*n*	*M*	*SD*	*t*	*n*	*M*	*SD*	*t*	*t*
FingTap	23	8.8	2.6	−2.1*	31	10.2	2.3	0.5	2.07*
FingDis-P	24	15.8	1.7	—	31	16.6	1.6	—	2.9
FingDis-NP	24	15.9	2.1	—	31	16.3	2.1	—	0.36
Knock-Tap	24	28.2	2.20	—	31	28.7	1.07	—	1.45
Design Copy	23	10.2	2.1	0.4	31	12.1	3.2	3.6**	2.48*
VisSp-Lrn	24	10.8	3.3	1.2	31	10.5	3.2	0.8	−0.43
VisSp-Ttl	24	11.4	3.1	2.2*	31	11.4	3.2	2.5*	0.00
VisSp-Del	24	10.8	2.9	1.4	31	11.5	2.2	3.8**	0.99

All scores are scaled scores except for FingDis-P, FingDis-NP, and Knock-Tap, which are raw scores. In the cases where scaled scores were available, one-sample *t*-tests were conducted to compare to norms (scaled score = 10). VisSp-Lrn: visual-spatial learning score (sum of scores on trials 1–3) from the Children's Memory Scale; VisSp-Ttl: visual-spatial "total" score (sum of scores on trials 1–3 plus short delay trial) from the Children's Memory Scale; VisSp-Del: visual-spatial "long delay" score from the Children's Memory Scale; FingTap: fingertip tapping score from the NEPSY; FingDis-P: finger discrimination preferred hand score from the NEPSY; FingDis-NP: finger discrimination non-preferred hand score from the NEPSY; Design Copy: from the NEPSY; Knock-Tap: from the NEPSY.

$^{*}p < .05$, $^{**}p < .01$.

the CI children scored significantly better than the expected norm value on only one measure, visual-spatial (total) and they scored significantly lower than norms for one other measure, fingertip tapping. In terms of group comparisons, the independent-samples *t*-tests revealed significant differences between the two groups only on fingertip tapping and design copying.

Because the fingertip tapping scaled score is derived from a combination of four raw scores—repetitive tapping (preferred and non-preferred hands) and sequential tapping (preferred and non-preferred hands), it is informative to assess performance on each of the four component tasks separately. We conducted analyses of covariance (ANCOVA) to assess group differences on the four fingertip tapping scores while controlling for the common variance associated with chronological age (CA), chronological age plus forward digit span (CA+FD), chronological age plus backward digit span (CA+BD), and chronological age plus vocabulary scores (CA+PPVT). The results of the group comparisons revealed that although the CI children were numerically slower than the NH children on all measures of fingertip tapping, they were significantly slower specifically on the repetitive tapping task with their non-preferred hand only, a finding that holds for all covariates.

In sum, the results on these experimental tasks revealed that deaf children with cochlear implants performed at age-appropriate levels on visual-spatial learning and memory, tactile discrimination, response inhibition (knock and tap), and visual-motor integration (design copy). Although the age-matched NH group of children showed slightly better performance than the CI children on design copying, this difference was due to the NH children performing better than expected for their age. The one measure that the CI children clearly showed a disturbance or delay on was the fingertip tapping task, as compared both to normative scores as well as the NH age-matched control group. This disturbance on the motor sequencing task remained robust even when effects of verbal working memory and vocabulary development were controlled.

Relation between experimental measures and language outcomes. We next examined the CI group's language outcome as measured by the CELF–4. Not unexpectedly, their group mean Core Language Score (86.89) was significantly lower than the age-scaled mean of 100, $t(17) = 7.8$, $p < .001$, indicating a substantial language delay. Determining whether particular underlying neurocognitive abilities can help explain why some children do well with their implant while others continue to struggle has become a major research goal (Pisoni et al., 2008). Although most previous work has focused on the role played by conventional demographic characteristics such as age at onset of deafness, length of auditory deprivation, age at implantation, and early linguistic experience (e.g., Kirk, Pisoni, & Miyamoto, 2000), comparatively little research has investigated more basic elementary cognitive and neuropsychological components. Previous work suggests there may be close links between nonverbal cognitive abilities and language development, with nonverbal abilities able to predict early language problems in typically developing children (Hayiou-Thomas, Harlaar, Dale, & Plomin, 2006; Oliver, Dale, & Plomin, 2004; Viding et al., 2003) and influencing language outcomes in deaf children with CIs (Dawson et al., 2002; Geers, 2002; Geers & Moog, 1987; Geers, Nicholas, & Sedey, 2003).

To investigate the possible association between nonverbal cognition and language outcomes, we computed partial correlations between the experimental measures and the CELF–4 Core Language Score, while controlling for the common variance associated with chronological age, with outliers excluded (when outliers were included in the correlation analyses, no significant results were obtained with any of the measures, although the overall trends are the same as reported below). With outliers excluded, the results revealed that only sequential fingertip tapping ($r = –0.53$, $p < .05$) and visual-spatial learning and memory ($r = 0.53$, $p < .05$) were significantly correlated with language outcomes. Sequential fingertip tapping was negatively correlated with language outcome, indicating that children who were slower on the sequential fingertip tapping task showed lower language scores as measured by the CELF–4. When the common variance associated with forward and backward digit spans were controlled, the correlation with visual-spatial learning and memory no longer reached significance. On the other hand, the correlation between sequential fingertip tapping and language outcome still remained significant even when controlling for digit spans ($r = -0.57$, $p < .05$). Thus, language outcome appears to be at least partially associated with the development of nonverbal visual-spatial memory abilities (see Dawson et al., 2002); however, this association may be due to shared variance between visual-spatial and verbal memory. In contrast, the association between sequential fingertip tapping and the CELF–4 remained significant even while controlling for the common variance associated with verbal short-term memory.

Together, these findings suggest that out of the nonverbal measures used here, only motor sequencing skills—over and above effects of verbal memory, vocabulary development, chronological age, and age at implantation—are associated with language outcomes in the CI children.

GENERAL DISCUSSION

The present findings can be summarized as follows. First, out of the experimental tasks used here, the only one that the deaf children with CIs showed a clear disturbance on was motor sequencing. They performed at or near age-typical levels on tactile perception (finger discrimination), response inhibition (knock and tap), visual-motor integration (design copy), and visual-spatial learning and memory. Second, performance on motor sequencing was significantly correlated with language outcome in the CI children.

Motor Sequencing Disturbance

A number of previous studies have found disturbances in various aspects of motor performance in deaf children (e.g., Savelsbergh et al., 1991; Schlumberger et al., 2004; Wiegersma & Van der Velde, 1983). Many of the tasks used in prior studies could be considered to be relatively complex because they involved the coordination of multiple movements (e.g., ball-catching ability) (Savelsbergh et al., 1991) and dynamic motor coordination (Schlumberger et al., 2004). On the other hand, consistent with the current findings, Schlumberger et al. (2004) also found a disturbance (i.e., significantly *slower* performance) in deaf children with CIs on more basic tasks of repetitive sequential movements: alternating heel–toe movements on the floor and using one hand to repetitively tap the knee.

Likewise, the group of CI children assessed here performed significantly slower on the NEPSY fingertip tapping task, which requires both simple repetitive and more complex sequential tapping movements. Although the overall scaled score was significantly lower in these children, which takes into account all four sub-tests of the fingertip tapping task (repetitive tapping and sequential tapping for both preferred and non-preferred hands), follow-up analyses on the raw untransformed scores indicated that the CI children were worse specifically on the repetitive tapping task using the non-preferred hand. This task involves repeatedly tapping the index finger against the pad of the thumb as quickly as possible.

That the motor sequencing deficit occurred specifically in the non-preferred hand suggests a possible disturbance with fine motor control in the non-dominant (right) hemisphere (all but two of the CI children were right-handed; reanalyzing the fingertip tapping data after removing the two left-handers did not modify the main effects). Previous work suggests that deafness results in a change to the normal pattern of cortical hemispheric specialization for language and visual-spatial processing (Marcotte & LaBarba, 1987; Neville et al., 1998; Wolff & Thatcher, 1990). For instance, deaf native signers of American Sign Language show an anomalous pattern of right-hemisphere brain activity when reading English (Neville et al., 1998), presumably because of a reliance on the right hemisphere for visual-form processing. As suggested by the current findings, it may be that these hemispheric changes resulting from deafness also affect the organization of neural circuits related to fine motor control in the non-dominant hemisphere, as evidenced by slower fingertip tapping performance.

The deaf children in the present study were unimpaired on all of the other experimental tasks. Although a null effect cannot conclusively demonstrate that impairments do not exist, due to possible lack of sensitivity with the measures, this finding is consistent with previous work showing equivalent or in some cases even superior performance on visual-spatial processing in the deaf compared to hearing populations (see Hauser, Cohen, Dye, & Bavelier, 2007).

Future research must determine the role that communication mode plays in the development of sequencing skills, if any: for instance, would the sequencing deficits observed here extend to deaf children who use a sign language as their primary mode of communication, such as deaf native signers (Bavelier et al., 2006)? Previous work does in fact suggest that signing deaf children also may have disturbances with (sequential) motor functions (e.g., Savelsbergh et al., 1991;

Wiegersma & Van der Velde, 1983). In addition, future work should further explore possible disturbances to cortical (motor) function in the right hemisphere, and the possible connection to other findings showing cortical reorganization in the deaf (e.g., Neville et al., 1998).

Of the experimental tasks used here, the fingertip tapping task alone appears to be an index of general sequencing skill or perhaps fine motor control specifically. Combined with previous work examining aspects of visual and visual-motor sequencing skills in the deaf (Horn, Davis, Pisoni, & Miyamoto, 2005; Horn et al., 2006; Rileigh & Odom, 1972; Schlumberger et al., 2004; Todman & Seedhouse, 1994), the current results are consistent with the hypothesis that a period of auditory deprivation may lead to secondary disturbances to aspects of nonverbal cognition, especially skills related to the representation and organization of sequences of visual-motor or motor patterns.

Synchrony Between Motor and Language Development

The second major finding of this paper is that performance on the finger sequencing task (specifically, the sequential tapping subtask) was significantly correlated with a global clinical measure of language development in the CI children. In typically developing children, motor and language milestones are closely coupled and tend to occur in synchrony (Lenneberg, 1967; Molfese & Betz, 1987; Siegel et al., 1982). In fact, motor control and coordination have been found to be strongly associated with various language measures in infants, children, and adults (Carello et al., 2002; Ejiri & Mastaka, 2001; Iverson & Fagan, 2004). For example, Carello et al. (2002) used a sequential fingertip tapping task that was not unlike the one used in the current study, and found that performance was associated with phonological decoding (i.e., reading) abilities in normal adults. Reading impairments due to developmental dyslexia are also associated with difficulties with motor timing and coordination (Wolff, 1999; Wolff, Michel, Ovrut, & Drake, 1990). In addition, children with specific language impairment (SLI) have been found to perform more poorly than age-matched controls on tasks involving motor control (Powell & Bishop, 1992); twin studies in which one or both twins have SLI have revealed a genetic link between language, motor skills, and working memory impairment (Bishop, 2000). Taken together, these findings point toward a close coupling between the development of motor skills and language processing, implying that successful language acquisition in part relies on intact motor skill development.

Recent work also points toward an association between motor development and language outcomes in children with CIs. Several longitudinal studies have compared motor assessments made before the child received an implant to the child's audiologic outcome measures post-implantation. These studies have found that children who present with higher motor scores do better on assessments of language, vocabulary, and spoken word recognition than children with lower motor scores (Broesterhuizen, 1997; Horn et al., 2005b). In addition, Horn et al. (2006) found that fine, but not gross, motor abilities were strongly correlated with expressive and receptive language abilities. Furthermore, performance on a visual-motor implicit sequence learning task was significantly correlated with aspects of spoken language development in deaf children with CIs (Conway, Pisoni, Anaya, Karpicke, & Henning, 2011). Broesterhuizen (1997) concluded that the development of speech and language skills in the deaf depend on abilities related to learning and producing sequential movements of the hand and mouth.

In the current study, it was the more complex sequential tapping task that was significantly correlated with language outcomes, not the repetitive tapping task that the CI children had the most difficulty with. The sequential tapping task is regarded as a more complex motor movement measure, requiring the child to tap the fingers sequentially against the thumb from index finger to little finger as quickly as possible. Children who were slower on this task showed lower language outcome measures, even after controlling for the effects of auditory-verbal working memory and vocabulary development. Because spoken language consists of a complex pattern of linguistic units (phonemes, syllables, words) occurring in temporal sequence (Lashley, 1951), it perhaps makes sense that the more complex finger sequencing task, rather than the simple repetitive tapping task, was most closely associated with language outcomes. Indeed, the ability to (implicitly) learn complex sequential patterns may be an important prerequisite for the development of language processing skills (Cleeremans, Destrebecqz, & Boyer, 1998; Conway, Karpicke, & Pisoni, 2007; Conway & Pisoni, 2008; Ullman, 2004).

The association observed between motor sequencing skill and language outcome—aside from its theoretical implications regarding the underpinnings of language development—is also of interest clinically because it helps to explain the enormous variability observed in language outcomes following cochlear implantation. Although a CI has been shown to be an efficacious medical prosthesis for developing spoken language skills (Gates & Miyamoto, 2003), as noted by both the National Institutes of Health (NIH) in a 1995 consensus statement on CIs in adults and children (NIH Consensus Statement, 1995) and a recent report by the American Speech-Language-Hearing Association (2004), an enormous degree of variation in outcome and benefit exists. It is likely that a very large amount of this unexplained variance is due to underlying basic neurocognitive factors that have gone largely unexamined in this clinical population, such as fundamental processes of learning, memory, and visual-motor development (Pisoni, 2000; Pisoni et al., 2008; Pisoni et al., 2010). The current findings thus point toward fine motor sequencing as being one such underlying neurocognitive factor that influences language outcome in this population.

Deafness, Motor Sequencing, and the Prefrontal Cortex

Why might deafness result in fine motor sequencing disturbances? One possibility is that the underlying cause of deafness may include disturbances to other parts of the brain and body not directly related to the hearing mechanism per se. However, this appears unlikely as the majority of children in this study did not present with any obvious impairments or damage aside from hearing loss. Another possibility is that delays in spoken language development cause the observed motor skill disturbances. Wiegersma and Van der Velde (1983) suggested that verbal rehearsal (i.e., “internal speech” strategies) may aid in the learning of new complex movements. However, this may also be improbable in the current case because it does not seem likely that children would rely on inner speech in order to perform the fingertip tapping tasks. For instance, there is evidence that children within the age ranges of our participants do not spontaneously engage in verbal rehearsal strategies (Ornstein, Naus, & Liberty, 1975; Naus, Ornstein, & Aivano, 1977).

A third possibility is that a period of auditory deprivation, rather than language delay, contributes to motor sequencing deficits. Because sound is inherently a temporally arrayed signal, it is possible that a lack of exposure to sequential input during early development results in the atypical development of neurocognitive processes having to do with the encoding and representation of sequential patterns. Under this view, sound may provide a kind of "scaffolding" (i.e., supporting framework) for processing serial input; therefore auditory deprivation could have negative consequences for the encoding, representation, and reproduction of serial patterns regardless of sensory modality (Conway et al., 2009). Findings showing the presence of a sequential processing disturbance in the deaf in different domains (Conway et al., in press; Pisoni, Conway, Kronenberger, Henning, & Anaya, 2010; Rileigh & Odom, 1972; Todman and Seedhouse, 1994)—rather than a disturbance solely with motor or verbal processing skills—lends support to this scaffolding hypothesis.

Fuster (2001) has argued that the prefrontal cortex (PFC) is critically involved in the temporal organization of behavior, including representing, formulating, and planning sequences of thought and action. For any complex sequential skill or behavior, the PFC is thought to be intimately involved because it allows for the integration of sensory cues with cognitive actions across time. Under this view, the PFC is important for any kind of sequencing or temporal functions (Conway & Pisoni, 2008), ranging from higher level planning, executive memory, and language processing, to more elementary abilities such as the sequential movements required for the fingertip tapping task. The PFC has many interconnections with other sensory, motor, and subcortical regions, making it an ideal candidate for more abstract, domain-general aspects of cognitive sequencing function (Miller & Cohen, 2001).

Whereas certain subcortical structures such as the basal ganglia also appear important for sequencing skills (Middleton & Strick, 2000; Seger, 2006), there is evidence that the PFC and basal ganglia contribute to sequencing skills in different ways. The PFC may be important for learning *new* sequences while the basal ganglia only becomes active once the sequences become well-practiced (Fuster, 2001); alternatively, the basal ganglia may contribute to reinforcement learning while the cortex is specialized to handle unsupervised learning situations (Doya, 1999).

For these reasons, if deaf children do indeed show delays on nonverbal sequencing abilities, disturbances to neural circuits in the PFC might be a possible explanation. Consistent with this hypothesis, electrophysiological data suggest that deaf children compared to hearing peers show decreased cerebral maturation in left fronto-temporal regions and bilateral frontal regions (Wolff & Thatcher, 1990). This finding in turn could be due to fewer neural projections out of auditory cortex in the deaf (Emmorey, Allen, Bruss, Schenker, & Damasio, 2003)—presumably including connectivity to the PFC. A lack of auditory input due to deafness therefore appears to reduce auditory-frontal connectivity, fundamentally altering the neural organization of the frontal lobe and crucially, the prefrontal cortex. Delayed cortical maturation in this region could have significant effects on the development of cognitive and motor sequencing skills used in language and other aspects of cognitive processing. If this account is correct, then other neurocognitive functions attributed to the frontal lobe ought to also be disturbed in hearing-impaired populations as well—such as executive functions, which include domain-general control processes such as working memory capacity, behavioral regulation, and goal-oriented behaviors. In fact, recent evidence suggests this is the case (see relevant chapters in Marschark & Hauser, 2008). These new findings suggest that a period of profound deafness may affect not only nonverbal sequencing skills, but also frontal lobe-related neurocognitive abilities more generally, including executive functions and cognitive control processes.

Future research might fruitfully address what could be regarded as limitations with the present study. For instance, our current sample of CI children had an imbalance of males ($N = 15$) to females ($N = 9$). Because gender can have a myriad of effects on cognitive, linguistic, and neural development (e.g., Reiss, Abrams, Singer, Ross, & Denckla, 1996) it will be important to control for gender differences in future studies. In addition, although the current study used a fairly diverse set of nonverbal measures that indicated a selective disturbance on nonverbal sequencing skill, much research remains to verify and clarify this proposal. For instance, are there some aspects of sequential processing that are more affected than others? And finally, all of these questions will likely benefit from careful use of neuroimaging and electrophysiological methods to explore the complex web of effects of auditory deprivation, linguistic development, and neurocognitive reorganization.

In sum, the present study provided an opportunity to examine the development of nonverbal, non-auditory abilities in deaf children with cochlear implants. The findings suggest that a period of auditory deprivation early in development has repercussions for cognitive functioning far beyond the obvious hearing- and language-related effects. Our findings indicate that although deaf children with CIs show age-typical levels of performance on nonverbal cognitive abilities related to visual-spatial processing, visual-motor integration, and sensory discrimination, they appear to be impaired on fine motor sequencing skills. Furthermore, motor sequencing skills were closely associated with language outcomes in these children, suggesting that individual variability on sequencing functions is closely coupled to language acquisition processes and therefore may help explain the enormous variation in speech and language outcomes and benefit observed in this clinical population.

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