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Behavioral Inhibition and Audiological Outcomes in Prelingually Deaf Children with Cochlear Implants[1]

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Abstract. Cochlear Implants (CIs) enable many prelingually deaf children to acquire spoken language skills although individual speech and language outcomes are quite varied. Differences in a range of underlying cognitive skills may explain a portion of this variance. The aim of this study was to determine if variation in behavioral inhibition skills of prelingually deaf children were related to speech perception, language, vocabulary knowledge, and speech intelligibility outcome measures after implantation. We conducted a retrospective analysis of longitudinal data collected from prelingually and profoundly deaf children who used CIs. A continuous performance task that did not require any auditory processing was used to measure behavioral inhibition skills. Audiological outcomes based on a battery of speech and language measures were obtained from children after 1, 2, and 3 years of CI use. Compared to published norms, the children in our sample performed in the low-normal range and delay task performance improved as a function of chronological age as well as length of CI use. A correlational analysis revealed several significant associations between behavioral inhibition and receptive and expressive language, vocabulary knowledge, and speech intelligibility scores. Most of these relations remained significant even when the effects of length of CI use and chronological age were partialled out. These findings suggest that speech and language processing skills are closely related to the development of verbal encoding skills, subvocal rehearsal skills, and verbally mediated self-regulatory skills in prelingually deaf children with CIs.

Introduction

Cochlear Implants are sensory aids that have been approved for the treatment of profound prelingual deafness in children as young as one year of age (US FDA, 2000). Numerous studies of audiological outcomes in deaf children with CIs have consistently demonstrated benefit of a CI on the development of oral language abilities such as speech perception, expressive language, vocabulary knowledge, and speech intelligibility (Miyamoto, Svirsky, Kirk, Robbins, Todd, & Riley, 1997; Svirsky, Robbins, Kirk, Pisoni, & Miyamoto, 2000; Tyler, Fryauf-Bertschy, Kelsay, Gantz, Woodworth, & Parkinson, 1997). Despite the impressive results of CIs in many deaf children, the significant individual differences in audiological outcomes of prelingually deaf children remain a challenging clinical and theoretical problem (Blamey, Sarant, Paatsch, Barry, Bow, Wales, Wright, Psarros, Rattigan, & Tooher, 2001; Pisoni, Cleary, Geers, & Tobey, 2000; Sarant, Blamey, Dowell, Clark, & Gibson, 2001). Reported outcomes vary a great deal. Some children obtain age-appropriate speech while others vary from children who obtain age-appropriate speech and language skills with their CI to others who obtain little benefit other than an awareness of sound. Studies which have examined predictors of CI performance in children have reported effects of early implantation, communication mode, device type, and dynamic range (Geers, 2003; Geers, Brenner, & Davidson, 2003; Geers, Nicholas, & Sedey, 2003; Kirk, 2000; Tobey, Geers, Brenner, Altuna, & Gabbert, 2003). However, a significant portion of outcome variance still remains unexplained by these demographic and medical factors (Pisoni et al., 2000; Sarant et al., 2001).

Recently, a number of investigators have suggested that cognitive and behavioral factors may play a role in acquiring spoken language skills with a cochlear implant (Dawson, Busby, McKay, & Clark, 2002; Knutson, Ehlers, Wald, & Tyler, 2000a; Knutson, Ehlers, Wald, & Tyler, 2000b; Pisoni, 2000). Individual differences in attention, learning, memory, and executive function of deaf children with cochlear implants may explain a portion of the variance in audiological outcomes following implantation with a CI. Recent studies of working memory have demonstrated close relations between deaf children's ability to encode, temporarily store, and reproduce, sequential stimuli such as digit lists and their performance on a wide range of speech perception, production, and language tests (Burkholder & Pisoni, 2003; Dawson, Busby, McKay, & Clark, 2002; Pisoni, 2000; Pisoni & Cleary, 2003; Pisoni & Geers, 2000; Surowiecki, Sarant, Maruff, Blamey, Busby, & Clark, 2002). Other research has shown that a deaf child's ability to obtain benefit from audiovisual redundancy in a sequence memory task is closely related to their spoken language skills with a CI (Cleary, Pisoni, & Geers, 2001; Lachs, Pisoni, & Kirk, 2001).

Compared to the recent literature on working memory, we know relatively little about the relations between CI outcomes and other cognitive functions. Several studies of sustained visual attention in deaf children with CIs have demonstrated an effect of CI experience, suggesting that this cognitive ability is closely tied to auditory and verbal language experience (Mitchell & Quittner, 1996; Quittner, Smith, Osberger, Mitchell, & Katz, 1994; Smith, Quittner, Osberger, & Miyamoto, 1998; Horn, Davis, Pisoni, & Miyamoto, under review). Only one of these studies actually examined relations between measures of sustained attention and audiological outcomes and this study did not find any significant correlations although children in this study had only used their CI for one year (Horn et al., under review). We know even less about executive functions in deaf children with cochlear implants. A recent study by Surowiecki et al. (2002) examined the relations between several measures of executive functions and CI outcomes in deaf children and did not find significant correlations, although the authors did not control for length of CI experience.

Knutson and colleagues investigated the relations between behavioral factors and audiological outcomes in deaf children with CIs (Knutson, Ehlers, Wald, & Tyler, 2000a; Knutson et al., 2000b). They tested children who had used their CIs for 36 months on the Child Behavior Checklist (CBCL) (Achenbach & Edelbrock, 1983; Achenbach & Edelbrock, 1986). This standardized parental report measures a variety of behavioral problems and produces composite standardized behavior scores for externalizing, internalizing, and total behavior problems. Externalizing behaviors on the CBCL include attention problems, impulsivity, aggression, and rule-breaking in contrast to internalizing behaviors which include withdrawal, depression, and anxiety. Knutson et al. reported that the externalizing and total problem composites were negatively correlated with performance on a range of speech perception and language tests (Knutson et al., 2000b). Their results suggested that a child's ability to control their behavior, particularly when behavioral inhibition was required, was related to their oral language skills.

The importance of Knutson et al.'s findings is magnified by a number of earlier studies which have found that deaf children and adults who use sign language appear to be more behaviorally impulsive than their peers (Altshuler, Deming, Vollenweider, Rainer, & Tendler, 1976; Chess & Fernandez, 1980; Kelly, Kelly, Jones, Moulton, Verhulst, & Bell, 1993; O'Brien, 1987). Therefore, it is critical to better understand how behavioral control, language development, and cochlear implant use interact during early development. However, we currently know little about the behavioral inhibition skills of deaf children who use CIs.

One aspect of behavioral control that has received some attention in the field of pediatric neuropsychology is response delay. This cognitive skill is thought to be distinct from the executive functions and is crucial for analysis and control of behavior (Barkley, 1997). Response delay includes related skills including inhibition of action, maintenance of a delayed state, and prevention of distraction from extemporaneous stimuli. These skills are critical for self-directed, goal-oriented behaviors. Impairments in behavioral inhibition and response delay are thought to be a central deficit in some types of Attention Deficit Hyperactivity Disorder (Barkley, 1997).

In the only study of response delay skills of deaf children with CIs to date, Mitchell and Quittner (1996) tested children on several continuous performance tasks. One of these tasks, the response delay task, required children to make serial key presses separated in time by a specific length. This length (4 seconds) was not explicitly stated but could be discerned from a visual counter which registered a point each time 4 or more seconds separated the key presses. Over a six minute period, children attempted to accumulate as many points as possible. Mitchell and Quittner found that deaf children's performance on this task was similar to age matched normal hearing children. The authors, therefore, concluded that behavioral inhibition skills of the deaf children with CIs were not impaired. However, the authors did not attempt to control for length of CI use and, therefore, their results cannot rule out the effect of auditory experience on response delay skills. Furthermore, Mitchell and Quittner did not examine relations between response delay skills and any outcome measures that assess oral language skills in this population.

We designed the current study to test the hypothesis that response delay skills would be related to oral language outcomes in deaf children with CIs. We also investigated whether response delay skills improved as a function of CI use when we controlled for chronological age. Finally, we used standardized scores to investigate whether the response delay skills of these children were comparable to a large national sample of normal hearing children. To assess response delay skills, we used a laboratory measure of response delay skills identical to the one employed by Mitchell and Quittner (1996).

Experiment

Methods

Subjects. All participants were part of an ongoing longitudinal study of the development of oral speech and language in prelingually deaf children with CIs at the Indiana University School of Medicine Cochlear Implant Program. A total of 47 prelingually deaf children (profoundly deaf by 2.5 years old) who received CIs by 9 years of age were included in this retrospective analysis. All children used Nucleus 22 devices. Mean age of implantation was 4.8 years old. Twenty-two children used OC (immersed in a training program using oral communication only) and 25 used TC (a program in which both oral and manual language are used) at the time of testing. Over 65% had an unknown cause of hearing loss, presumed to be congenital. The most common acquired etiology was meningitis. A subset of children (n=18) had pre-implant non-verbal IQ scores (WISC or WPPSI) in their charts. The mean standardized IQ score was 104.1 (sd=15.28).

Children were tested once every 6-12 months from before implantation to three years post-implantation. Interval data were collapsed into one of four intervals: pre-implant, one year post-implant, two years post-implant, and three years post-implant. Not all children were tested at each interval, as is common in clinical populations such as this, creating missing data cells. Missing data occurred for several reasons. First, some children moved away from the Indianapolis area after implantation and were unable to continue with the study. Second, because our clinical participants are given a large number of tests during each visit, they are often too tired or not cooperative enough to complete all testing procedures.

Procedures. The Preschool Delay task (Gordon, McClure, & Aylward, 1996) is recommended for use in normal hearing children from 3-5 years of age. The test apparatus consists of a customized mechanical button box with one blue key located centrally below an LED display. The apparatus is capable of running a number of neuropsychological tests and is used to measure response delay and behavioral inhibitory skills (Gordon et al., 1996). The delay task is a 6 minute test in which children are asked to press a key and then wait for some period of time before pressing the button again. Each time the child presses the button after waiting four seconds, they receive a point which is shown on the display screen. The number of points a child receives over 6 minutes is divided by the total number of key presses to compute the “error-ratio” (ER) which is the primary dependent measure on this task. Children receive no feedback regarding their performance other than the point counter. Thus, it is up to the subject to figure out how long they have to wait in order to get a point.

Five speech and language outcome measures were used in the correlational analyses. Open set speech perception was measured using the Phonetically Balanced Kindergarten (PBK) test (Haskins, 1949). Children hear a spoken word and are asked to repeat the word aloud to the examiner. This test is administered using a live voice and is scored by word and phoneme. The words used in this test are phonetically balanced, English monosyllables.

Sentence comprehension was measured with the Common Phrases test (Osberger, Miyamoto, Zimmerman-Philips, Kemink, Stroer, Firszt, & Novak, 1991). The child is asked to repeat a sentence or follow a command and his response is scored as correct or incorrect. Sentences are administered in auditory (CPA), visual (CPV), and auditory plus visual modalities (CPAV) in separate conditions. We analyzed scores from all three presentation formats.

Vocabulary knowledge was assessed with the Peabody Picture Vocabulary (PPVT) Test (Dunn & Dunn, 1997). This widely used test is a closed set, forced choice task. At our center, each vocabulary item is presented live either orally or manually with Signed Exact English depending on the individual child’s preferred mode of communication. The child then chooses from four pictures, one of which correctly corresponds to the meaning of the word.

The Reynell Developmental Language Scales 3rd edition (Reynell & Huntley, 1985) was administered to assess both receptive (RDLS-R) and expressive (RDLS-E) language skills. The receptive scales measure 10 different skills including word recognition, sentence comprehension, and verbal comprehension of ideational content. The expressive language scales assess skills such as spontaneous production of speech and picture description. Like the PPVT, the RDLS was administered in the child’s preferred mode of communication.

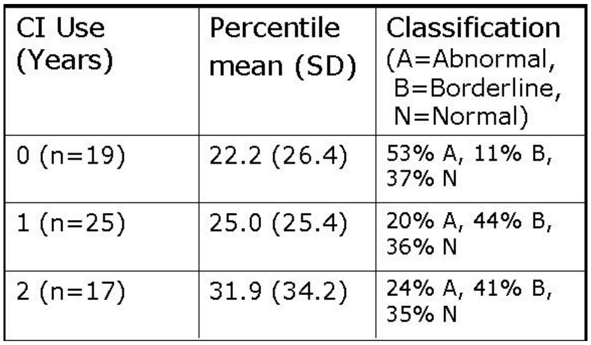
Speech intelligibility was assessed using the Beginner’s Intelligibility Test (BIT) developed by Osberger, Robbins, Todd, and Riley (1994). Audio recordings were made of children repeating a list of 10 sentences presented to them in the auditory modality by a clinician. These recordings were then played back to 3 naïve adult listeners who were asked to transcribe what they thought the children were saying. Intelligibility scores were based on the number of words correctly transcribed by the adult listeners.

Each of these tests, including the delay task, were administered in an Otolaryngology clinical setting by licensed health professionals who had received special training in working with deaf children who used CIs.

Results

Delay task performance of prelingually deaf children with CIs compared to a normative sample. Normative data for the delay task performance are available for children aged 3-5 years old. Therefore, many of the children in our sample were too old to derive percentile or standard scores from these norms. Therefore, we calculated normative percentile scores from the CPT raw scores only for children who were 6 years of age or younger at the time of testing. Table 1 shows the percentile means for ER at each interval along with the number of subjects who could be normed at each interval. This subset of our sample performed in the low normal range on this task compared to the normative sample. The highest mean percentile scores were found in children who had used their CI for two years (32nd percentile). However, even after two years of implant use, about 1/3 of the children performed in the "abnormal range" according to the published norms (Gordon et al., 1996). We did not perform statistical analyses using these percentile scores because these scores may not have been representative of the entire sample of children.

Effects of chronological age and length of CI use. As mentioned previously, because our subjects were drawn from a larger clinical population enrolled in a long-term longitudinal study of CI outcomes, there were missing data cells for different children at each test interval. A traditional repeated measures ANOVA would, therefore, eliminate data from any child who was not tested at each interval. Such an analysis can lead to skewed results as well as underestimates of variability (Schafer & Graham, 2002). To avoid this problem, we analyzed our data using the SAS Mixed Procedure (Wolfinger & Chang, 1995). The SAS Mixed Procedure utilizes a maximum-likelihood estimation method to create a model without eliminating any participants (Schafer & Graham, 2002). In this manner, systematic selection biases can be avoided and data from all children, even those who were not tested at each interval, could be included in the statistical model.



CI Use (Years)	Percentile mean (SD)	Classification (A=Abnormal, B=Borderline, N=Normal)
0 (n=19)	22.2 (26.4)	53% A, 11% B, 37% N
1 (n=25)	25.0 (25.4)	20% A, 44% B, 36% N
2 (n=17)	31.9 (34.2)	24% A, 41% B, 35% N

Table 1. Mean percentile error ratio on the delay task at each interval of CI use. Clinical classifications based on the normative sample tested by Gordon et al. are provided as well.

The results obtained using the SAS mixed model revealed a significant effect of chronological age on delay ER ($F(1,46.7) = 11.02$, $p=0.0018$). We also found an effect of length of CI use on delay ER ($F(2,50.1) = 4.66$, $p=0.014$), when chronological age was controlled. Our analysis also revealed a significant interaction between chronological age and length of CI use ($F(2,52.1) = 3.51$, $p=0.037$). Figure 1 displays the individual ER scores as a function of chronological age at CI activation and length of implant use. Examination of this figure shows that at pre-implant and 1 year post-implant intervals, delay task performance improved as a function of chronological age. However, at two years after implantation, mean ER decreased with chronological age. Children who were younger at time of implantation (and younger over the three-year testing period) showed more improvement in ER with CI use than children who were older at implantation.

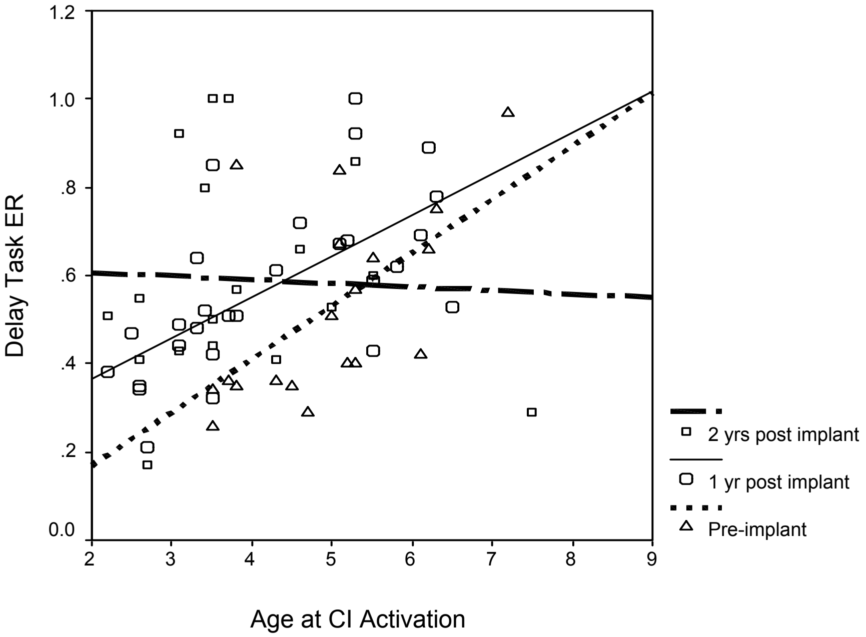


Figure 1. Delay task error ratio as a function of chronological age of CI activation and length of CI use.

This interaction should be interpreted with caution, due to a possible selection bias in the older children. As mentioned previously, the Preschool Delay task is clinically recommended for children 3-5 years old. For older children, the Gordon Diagnostic System offers a harder version of the delay task. Some of the older children in this study were unable to complete the harder delay task and, therefore, the easier task was administered. Thus, these delayed children may have been over-represented among our oldest children (particularly the child who was implanted at 7.5 years old and had an ER of 0.2). Nevertheless, the main effect of length of CI use on delay ER demonstrates that, overall, children's performance on this task improved with CI experience.

Correlations between delay task and speech and language outcome measures A total of 35 children in our sample had delay task scores obtained after 1 to 3 years of CI use. For children who had delay ER scores at more than one interval, we used only their post-implant year 1 scores for correlation purposes. The number of children who had scores on both the delay task and the outcome measures differed across post-implant intervals and outcome measures. Therefore, we report the sample size of each correlation.

Bivariate correlations were computed between delay task ER and each of the outcome measures at 1, 2, and 3 years post-implantation. A summary of these results is displayed in Table 2 which includes each individual Pearson correlation, p value, and sample size. We found significant and positive correlations between ER and vocabulary (PPVT), expressive language (RDLS-E), receptive language (RDLS-R), and speech intelligibility (BIT) scores at all three post-implant outcome intervals. As shown in Table 2, these correlations were strong and most were significant at a level of $p<0.01$. In contrast, no significant correlations were found between delay task ER and open set speech perception scores (PBK) or sentence comprehension scores (CPA, CPV, CPAV).

Outcome Measure*	Post-implant yr 1	Post-implant yr 2	Post-implant yr 3
Speech perception			
PBK words	NS	NS	NS
PBK phonemes	NS	NS	NS
Sentence comprehension			
CP auditory	NS	NS	NS
CP visual	NS	NS	0.51 (22)
CP audiovisual	NS	NS	NS
Language & Vocabulary			
PPVT	**0.69 (34) ++**	**0.62 (35) ++**	**0.55 (31)**
Reyn receptive	**0.60 (32) +**	**0.64 (29) ++**	**0.61 (27) ++**
Reyn expressive	**0.54 (32) +**	**0.61 (25)**	0.46 (27)
Speech Intelligibility			
BIT	**0.42 (30) +**	**0.38 (32)**	0.40 (24)

*. All outcome measures were in % correct except for PPVT (raw scores) and Reynell (age equivalent scores in months)
NS: non-significant bivariate correlation ($p>0.05$)
Normal face type: significant bivariate correlation ($p<0.05$)
Bold face type: significant bivariate correlation ($p<0.01$)
++. significant with length of CI use and chronological age partialled out ($p<0.01$)
+. significant with length of CI use and chronological age partialled out ($p<0.05$)

Table 2. Bivariate and partial correlations between delay task scores and outcome measures.

For those bivariate correlations which were significant, we also conducted a series of partial correlations to control for chronological age and length of CI use at time of delay task administration. The results shown in Table 2 reveals that many of these partial correlations were also significant at the $p<0.01$ level and several others at the $p<0.05$ level. For the outcome measures obtained after 1 year of use, partial correlations were significant between delay ER and PPVT, RDLS-R, RDLS-E, and BIT measures. For outcomes obtained after 2 or 3 years of CI use, a number of these relations no longer reached statistical significance. This was not surprising since most of the delay ER scores were obtained after 1 year of CI use. For PPVT at year 2, and RDLS-R at years 2 and 3, the partial correlations with delay ER remained significant.

Discussion

The results of our analysis of the Gordon Delay Task revealed several new findings regarding the behavioral inhibition skills of prelingually deaf children who use CIs. First, a large number of deaf children displayed behavioral inhibition skills which were atypical when compared to the results of a normative sample reported by Gordon et al. (1996). This finding contrasts with the earlier results of Mitchell and Quittner (1996) who tested an older population of children on a similar delay task and found no differences in performance between normal-hearing children and prelingually deaf children with CIs. The difference between the current and previous findings may be due to the fact that the participants in our study were several years younger on average than the children tested by Mitchell and Quittner. Measures obtained from the Gordon Delay Task may not have been sensitive enough to detect differences in behavioral inhibition ability between older deaf and normal hearing children. Furthermore, most of the participants in our study had used their CI for only one year while Mitchell and Quittner included children who had used their implant for as long as six years. It is possible that the deaf children Mitchell and Quittner tested caught up to normal hearing children in their behavioral inhibition skills due to these extra years of CI use. However, additional research needs to be done in this area to better understand this issue.

We also found that delay task performance increased with length of CI use as well as chronological age. These two results suggest that CI use in prelingually deaf children facilitates the development of behavioral inhibition skills. The skills used in the Gordon Preschool Delay Task may indeed include some language related skills as discussed below. In addition, we found an interaction between length of CI use and chronological age suggesting that the older children did not demonstrate the same degree of improvement with CI use as did younger children in our study. This interaction may represent a sampling artifact in our study design.

Finally, our analysis of the delay task revealed several significant correlations between behavioral inhibition skills and vocabulary knowledge, language, and speech intelligibility scores at one year post implantation. Several of these relations also remained significant after two or three years of CI use. One explanation of these findings is that performance on the response delay task reflects behavioral control skills, which play a significant role in real world language learning. Our findings along with the results of Knutson et al. (2000a, b) provide some converging support for this hypothesis, although prospective, longitudinal studies are needed to directly assess the possible causal relationship between behavioral control abilities and oral language development in deaf children with CIs. There is an extensive literature showing that the difficulties faced by children with ADHD, including impulsivity, have a significantly negative impact on scholastic performance (Lamminmaki, Ahonen, Narhi, Lyytinent, & de Barra, 1995) and many children with ADHD also exhibit delays in language and math abilities (Barkley, 1990, 1997).

Another explanation of our current findings is that the relations observed between response delay and language outcomes in our sample of children reflect underlying language related skills such as internal speech, sub-vocal verbal rehearsal, and counting which are used by children as a strategy to perform the delay task. For instance, one possible strategy would be to press the button, then count to oneself for a few seconds, and then press the button again. This sequence would be repeated until the score counter registered a point, indicating that the subject had waited a sufficient length of time. Thereafter, the subject would simply count the required number of seconds before each response, thereby maximizing the ER.

Clearly, such a strategy would require some degree of sophistication in counting, and internal speech mechanisms which utilize subvocal verbal rehearsal strategies. Several related findings in the literature suggest that deaf children may not be able to take advantage of subvocal rehearsal strategies during tasks in which it would be beneficial to do so. For example, in a recent study, Pisoni & Cleary (2003) measured the immediate memory capacity for spoken lists of digits in normal hearing children and in deaf children with cochlear implants and reported a main effect of hearing status and an interaction between hearing status and order of recall. The normal hearing children showed longer digit spans overall compared to deaf children with CIs. When asked to recall the sequence in the same order as presented (forward digit span) the normal hearing children showed greater digit spans compared to the condition that required recall of the sequence in the backwards order (backward digit span). This effect in normal hearing children was taken as evidence that normal hearing children were able to benefit from subvocal verbal rehearsal strategies to maintain a longer digit sequence in working memory when temporal sequence did not have to be manipulated. In contrast, the deaf children showed significantly smaller differences between spans obtained in the forward and backward conditions, suggesting that these children did not utilize subvocal rehearsal strategies successfully.

Other studies have demonstrated that the atypical recall capacity of deaf children with CIs is not limited to tasks involving lists of auditory stimuli. Indeed, immediate recall for sequences of visual stimuli which can be verbally encoded such as colored lights (Cleary et al., 2001; Dawson et al., 2002) have also been found to be atypical compared to age matched, normal hearing peers. This finding lends further support to the hypothesis that deaf children with cochlear implants are atypical in their capacity to encode, manipulate and store stimuli which can be represented phonologically or subvocally (Pisoni & Cleary, 2002).

Finally, there is now strong evidence that subvocal rehearsal abilities are related to open set speech perception, vocabulary knowledge, expressive and receptive language, speech intelligibility, and speaking rates in prelingually deaf children with CIs (Burkholder & Pisoni, 2003; Dawson et al., 2002; Pisoni & Cleary, 2002; Pisoni & Geers, 2000). The present results obtained with the response delay task suggest that subvocal rehearsal abilities are closely tied to other traditional measures of CI benefit. However, relations between delay ER and other process measures known to reflect subvocal rehearsal (such as forward-backward digit span) need to be investigated in future studies of deaf children with CIs.

In summary, the present results provide several new findings regarding the relations between the cognitive processes involved in behavioral inhibition and audiological outcomes in prelingually deaf children with CIs. A period of early auditory deprivation prior to implantation may have specific cognitive effects on deaf children, which may impact their ability to successfully acquire an oral language with a CI. However, further work is needed in order to understand precisely how early auditory deprivation affects the development of behavioral inhibition and how language skills and response delay skills are related. For instance we do not know whether auditory experience has an effect on behavioral inhibition skills through remodeling of language processes per se, or whether there are more specific effects of audition on cortical areas responsible for self-regulatory behavior mediated by pre-frontal cortex, anterior and posterior cingulated gyrus, and other areas (Barkley, 1997; Rubia, Overmeyer, Taylor, Brammer, Williams, Simmons, Andrew, & Bullmore, 2000). Future research on neuropsychological functions of deaf children who use CIs is needed to answer this and other related questions. Furthermore, neuropsychological testing of these children may prove useful as a clinical tool in assessing outcomes and benefit from a CI in this unique population of children.

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