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**Speech and Language Development in
Cognitively Delayed Children with Cochlear Implants¹**

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Abstract. The primary goal of this investigation was to examine the speech and language development of deaf children with cochlear implants who have additional impairments relative to that of their implanted peers without additional disabilities. A secondary goal was to address some limitations of previous investigations that have included multiple types of disabilities in their experimental groups by focusing on deaf children with cochlear implants who have a single additional impairment, mild cognitive delay. We retrospectively examined the speech and language development of 69 prelingually-deaf children. The experimental group consisted of 19 children with cognitive delays and no other disabilities. The control group consisted of 50 children who did not have cognitive delays or any other identified disability. The control group was stratified by primary communication mode: half used total communication and the other half used oral communication. Children were tested on a variety of standard speech and language measures and one test of auditory skill development at 6-month intervals. The results from each test were collapsed from blocks of two consecutive 6-month intervals to calculate group mean scores prior to implantation and at 1-year intervals post-implantation. The children with cognitive delays and those without such delays demonstrated significant improvement in their speech and language skills over time on every test administered. There was a trend for typically developing children to have better speech perception than children with cognitive delays, although these differences were not statistically significant. In contrast, typically developing children performed significantly better than those with cognitive delays on two of the three measures of receptive and expressive language. Finally, there was no significant difference between the groups of children on their parental reports of auditory skill development. The results suggest that deaf children with mild cognitive impairments benefit from cochlear implantation. Specifically, improvements are evident in their ability to perceive speech and their reception and use of language, although it may be reduced relative to their typically developing peers with cochlear implants, particularly in domains that require higher level skills such as sentence recognition and receptive and expressive language. These findings suggest that children with mild cognitive deficits be considered for cochlear implantation with less trepidation than has been the case in the past. Although their speech and language gains may be tempered by their cognitive abilities, these limitations do not appear to preclude benefit from stimulation from a cochlear implant on traditional measures of speech and language development.

Introduction

The first individual under 18 years of age to receive a cochlear implant in the United States did so in July 1980 (House & Berliner, 1991). During the following decade several hundred children received cochlear implants. It was not, however, until 1990 that the U.S. Food and Drug Administration approved their use in individuals aged 2 to 17 years. Since that time, candidacy criteria have broadened with improvements in technology and reports of increasingly positive changes in users' speech and language development. Currently, the criteria include children as young as 12 months of age with profound hearing loss and those with severe-to-profound hearing loss who are 24 months of age or older. Candidacy is based upon a number of factors including degree of hearing loss, demonstration of minimal benefit from amplification, enrollment in therapy that promotes auditory skill development, and no medical

contraindications. Children with additional disabilities are currently excluded from FDA clinical trials, but have been implanted with approved devices at Indiana University School of Medicine and at other centers around the country. Little is known about the effects of cochlear implantation on speech and language development in these children.

In their Annual Survey of Deaf and Hard-of-Hearing Children and Youth, the Gallaudet Research Institute reported that one-third of children in the United States with some degree of hearing loss have additional disabilities (Holden-Pitt & Diaz, 1998): 4% were blind or had an uncorrected vision problem; 4% had emotional/behavioral problems; 8% had mental retardation; and 9% had learning disabilities (the additional disability was not reported for 9% of the sample). In their study of 199 children with autism, Rosenhall, Nordin, Sandstrom, Ahlsen, and Gillburg (1999) reported a prevalence of profound bilateral hearing loss of 3.5%, higher than the 1% reported for the general population. Another investigation reported that 19% of children evaluated for cochlear implantation candidacy at Medizinische Hochschule Hannover had additional impairments (Lesinski, Hartrampf, Dahm, Bertram, & Lenarz, 1995). One only needs to spend a short amount of time in any pediatric center to recognize that hearing loss often coexists with other impairments, whether presenting as a syndrome or some other constellation of disabilities. These prevalence data indicate that cochlear implant centers will have a percentage of pediatric cochlear implant candidates with additional disabilities that may influence post-implant outcomes. Currently, there is no consensus on whether to implant children with multiple impairments. As with other children, this decision should be based upon the expected benefit. Although seemingly straightforward, the question of what constitutes benefit is a valid one, especially for this population. Is benefit cost effectiveness? Is it speech and language gains? Is it psychosocial development? Is it improved quality of life (which itself often is ill-defined)? Waltzman, Scalchunes, and Cohen (2000) raised this issue in their report of multiply impaired children with cochlear implants. They reported results on speech and language tests from children with cochlear implants who had a wide array of additional disabilities. Although the children with additional handicaps had lower scores on measures of speech and language development and fewer were able to complete the test measures relative to their implanted counterparts with no additional impairments, Waltzman et al. reported that the multiply impaired children demonstrated an increased “general connectedness to their environment” (p. 334), and improved auditory skills, communication skills, and social interactions. Even if it is determined that certain disabilities preclude benefit from a cochlear implant (however benefit is defined), there still will be a group of children implanted so early in life that additional disabilities may not yet be evident. In addressing this issue, it is important to understand the gains in speech and language development these children will experience if they are implanted and what types of measures will best reflect their performance.

Speech and Language Outcomes

A limited number of previous investigations have addressed some of these questions. Pyman, Blamey, Lacy, Clark, and Dowell (2000) retrospectively examined the speech and language development of 75 pediatric cochlear implant recipients. They were interested in determining whether the etiology of deafness was responsible for some of the variability in speech perception outcomes in children with cochlear implants. In addition to grouping by cause of deafness, the investigators classified the children according to whether each child showed evidence of motor and/or cognitive delays. Approximately one-third of the children with motor and/or cognitive delays were oral communicators and the remaining two-thirds were total communicators. This ratio was reversed for the children without additional impairments. Pyman et al. reported that although the cause of deafness had a relatively small influence on speech and language development, the presence of a motor and/or cognitive delay tended to slow aspects of speech and language development that employ higher level speech processing abilities. Specifically, the children with motor and/or cognitive delays (N=20) showed improvements in *detection* ability that were *similar in both degree and rate* as the children without additional impairments (N=55). However, on tests of

discrimination of suprasegmentals, vowels, and consonants [generally thought to be examples of mid-level speech processing (Aslin & Smith, 1988; Carney, 1991)], children with motor and/or cognitive delays demonstrated a *slower rate* of progress, but *reached the same final level* of functioning as the group without additional impairments after 4 years of implant use. Finally, on tests of high-level speech processing (open-set *word recognition*), the children with motor and/or cognitive delays showed *slower rates* of improvement and *never reached the level* of performance of the group without additional impairments, even after 4 years of implant use. Therefore, even though nearly 90% of the children with motor and/or cognitive delays could discriminate vowels and consonants at 4 years post-stimulation (a similar proportion of those without additional impairments could, as well), only approximately 60% could use this information to identify words in open-set sentences compared to more than 80% of those with no additional impairments.

In a smaller study mentioned earlier, Waltzman et al. (2000) surveyed speech perception abilities of 29 children with multiple disabilities who received cochlear implants. There was a wide array of disabilities and syndromes represented in the sample (such as, autism [N=1], attention deficit disorder [N=3], learning disability [N=2], gross motor delay or poor motor planning [N=7], reduced cognitive function or developmental delay [N=7], dyspraxia [N=1], CHARGE association [N=1], Waardenberg syndrome [N=1], Usher syndrome [N=1], etc.) and, we suppose for that reason, the results were not analyzed by type of disability. However, relative to a control group of children without additional disabilities, fewer of the multiply impaired children were able to complete the speech perception tests given and when they could, their average scores were lower than those of the children without additional impairments. Of their 29 multiply impaired participants, 7 were identified as being developmentally delayed or having reduced cognitive function. Not a single one of those with developmental delay or reduced cognitive function completed all seven of the speech perception tests given (including, auditory-only recognition of phonemes, words, and sentences in both closed- and open-set formats, and auditory-only discrimination of pattern perception from the Early Speech Perception test battery [Moog & Geers, 1990]). In fact, three of the children were unable to complete any of these tests over an 8-year testing period. A fourth only had scores reported for the Early Speech Perception Test (achieved only Level 1, No Pattern Perception) and the Glendonald Auditory Screening Procedure (Erber, 1982; 8% correct on open-set word recognition after 1 year of device use). Of these 4 with no or very minimal data, 3 used manual or total communication and 1 used oral communication. The remaining three participants with developmental delay were oral communicators. These three children scored between 70% and 100% correct on various tests of closed- and open-set word and sentence recognition after 4 years of implant use. Clearly, these seven children with developmental delay demonstrated a wide range of speech perception skills. The authors anecdotally pointed out that the children with multiple disabilities had increased social interaction and “connectedness” to the environment post-implantation. Also, although the rate of growth of auditory and linguistic skills was slower than that for deaf children without additional impairments, Waltzman et al. reported that the multiply handicapped children obtained demonstrable benefit from implantation, although there were no objective measures of this type of device benefit. Again, this raises the question of how to assess perceived benefit.

Hamzavi, Baumgartner, Egelierler, Franz, Schenk, and Gstoettner (2000) examined speech perception ability and general auditory behavior of 10 children with multiple handicaps who received cochlear implants. Their skills were assessed with the Evaluation of Auditory Responses to Speech (EARS) test battery in German. This battery includes closed-set word recognition, closed-set sentence recognition, and open-set word recognition tests. Prior to implantation, none of the children showed evidence of speech perception or production. Half the children achieved some level of word recognition and production after 3 years of implant use. The additional disabilities represented in the group that achieved some degree of speech recognition and production ranged from moderate learning difficulties (N=2), autism with mild intellectual deficit (N=1), hyperactivity with severe intellectual deficit (N=1),

and hemiparesis (N=1). Of the remaining 5 participants who did not demonstrate word recognition or production after 3 years of implant use, 4 demonstrated changes in their behavior: one, with severe intellectual deficit and severe psychomotor retardation, demonstrated differentiated phonation and searched for the source of detected sound in her environment; the second, who has severe learning difficulties and “low concentration,” was able to differentiate certain voices and utterances; a third, with severe motor retardation, had reduced lethargy and was able to associate speakers’ lip movements with phonation; and the fourth child, with psychomotor retardation and autism with stereotyped behavior, had improved phonation, reduced stereotyped character, was “happier with noise,” and liked to carry her processor with her. The tenth participant demonstrated no response to sound, even though it had been verified that the processor and internal device were both functioning. Similar to Waltzman et al.’s (2000) investigation, there was a rather wide range of speech and language outcomes in this population.

A recent case study by Fukuda et al. (2003) followed the language development of a single pediatric cochlear implant recipient with moderate developmental delay. This child underwent cochlear implant surgery at age 4;8 (years; months) after unsuccessfully wearing hearing aids for about 4 years. Prior to implantation this child communicated primarily through gestures and sign language. His pre-implant vocabulary included 144 words by gesture and 166 signs. After his implant was activated, he made substantial gains in a number of domains. His monosyllabic open-set, auditory-only word recognition scores increased from 0% pre-implant to 75% correct at 2 years post-stimulation. The authors reported that with only 15 months of implant use he could follow his teacher’s instructions in a noisy classroom and was beginning to understand simple conversations on the telephone in quiet environments. Although he had nearly 300 signs and gestures prior to implantation, he had no oral words. After 2 years of experience with his implant, he had an oral vocabulary of 692 words, and with just 10 months of implant use, was using three-word sentences. Finally, over the 15 months after his initial stimulation, his gross motor development increased by 24 months, his fine motor by 18 months, his social skills increased by 24 months, his self-help skills by 48 months, and his language by 36 months.

Each of these investigations has taken the field closer to understanding the speech, language, and general communicative behavior of children with multiple impairments who have cochlear implants. However, they are limited in their ability to guide us in determining whether children with multiple impairments benefit from cochlear implantation for at least two reasons. First, there are often too few participants to do any analyses beyond those that are descriptive in nature. Second, the array of disabilities represented in the samples is typically quite broad with each disability having different degrees of impact on communication. The current investigation was undertaken to try to control for these factors in order to better understand the abilities of multiply impaired children with cochlear implants and to better guide future studies.

Methods

Using a retrospective design, we analyzed the speech and language development of cochlear implanted children with and without cognitive delays. These children were followed in our laboratory at pseudo-regular 6-month intervals in which they completed a battery of speech and language measures at each visit.

Disability classification scheme

A number of investigators have attempted to separate hearing-impaired children with additional impairments into groups based on their additional disability. The rationale behind this categorization is that various additional impairments may differentially influence communication performance with a sensory aid. For example, the sequelae of autism affect communication differently than those of

blindness. In one effective categorization scheme, developed by Tharpe, Fino-Szumski, and Bess (2001), categories were chosen to be mutually exclusive and to be representative of those seen in audiology clinics. The classification scheme was used for a different purpose than ours (to examine hearing aid fitting practices for children with multiple impairments), but is believed to be relevant to our group of children with cochlear implants. The disability categories they used were: no disabilities other than speech and language disorders, vision impairment, mental retardation, physical impairment, and autism spectrum disorders. Although there are a number of ways to group disability categories, this method worked well for these investigators, and thus, we separated the children implanted at our center with multiple impairments into these categories. We quickly found that these categories are not mutually exclusive, in that some children had more than one additional disability that placed them into more than one category. A solution to this multiple category assignment follows.

One limitation of previous studies is that a range of additional disabilities was included in the experimental group, making it difficult to determine what aspects of each disability impact communication after cochlear implantation. Therefore, as a first step we wished to focus on a single disability, cognitive delay, in order to better isolate the effects of a specific disability on speech and language development in children with cochlear implants. Although this disability presents itself in various degrees of severity, we at least were able to focus on a single handicap known to impact communication. To address the multiple category assignment issue and to help control the range of disabilities included in the sample, we excluded implanted children with cognitive delays who had other diagnosed disabilities (e.g., vision loss, physical impairments, etc.).

The relationship between nonverbal IQ and language ability in normal-hearing children has been well established for some time (Wechsler, 1974; Zimmerman & Woo Sam, 1972). More recently, this relationship also has been demonstrated in children with cochlear implants (Dawson, Busby, McKay, & Clark, 2002; Geers, Brenner, Nicholas, Uchanski, Tye-Murray, & Tobey, 2002; Geers, Nicholas, & Sedey, 2003). Geers, Brenner, and Davidson (2003) showed a significant positive correlation between performance IQ and speech perception skills in children with cochlear implants, whereas Tobey, Geers, Brenner, Altuna, and Gabbert (2003) reported similar findings between performance IQ and speech production skills. These investigations indicate that there is a relationship between an individual's level of cognitive function and her/his performance on measures of speech and language whether or not she/he has normal hearing or is deaf with a cochlear implant. Our decision to focus on a subgroup of multiply impaired children, deaf children with cognitive delays, was based on this link and the previous reports that mental retardation or cognitive delay has postoperative effects on communication in children with cochlear implants (Hamzavi et al., 2000; Lesinski et al., 1995; Pyman et al., 2000; Waltzman et al., 2000).

Participants

Sixty-nine children with cochlear implants were identified for inclusion in the analysis. All the children had an onset of deafness by 2.5 years of age and used current cochlear implant devices and speech processing strategies. All but one of the children were implanted with their device before 5 years of age. The child implanted outside this age range received his device at age 5;11 and was included in the experimental group.

The participants were separated into two groups, with the second group stratified by communication mode (oral or total). The experimental group (N=19) consisted of hearing-impaired children with cochlear implants who were identified as developmentally delayed by a psychologist and/or scored at least 1 standard deviation below the mean on one or more of the following tests of cognitive functioning, and who did not have a diagnosis of any other impairment. Specifically, we included children who scored at least 1 standard deviation below the mean on the Wechsler Preschool and Primary

Scale of Intelligence-III (WPPSI-III; Wechsler, 2002), the Developmental Assessment of Young Children (DAYC; Voress & Maddox, 1998), the Bayley Scales of Infant Development - II (Bayley, 1993), or the Stanford-Binet Intelligence Scales – 4th edition (Thorndike, Hagen, & Sattler, 1986). The WPPSI reliably measures intellectual abilities of children down to age 2;6. The DAYC can be used to identify children birth through 5;11 with possible delays in the domains of cognition, communication, social-emotional development, physical development, and adaptive behavior. The Bayley is used to evaluate the developmental status of children as young as 1 month of age. Finally, the Stanford-Binet Intelligence Scales test intelligence and cognitive abilities in adults and children down to age 2 years. The control group consisted of hearing-impaired children with cochlear implants who scored within 1 standard deviation of the mean on any of these tests and who did not have a diagnosis of any impairment. Those children identified as controls were further stratified by primary mode of communication: oral communicators (N=25) or total communicators (N=25). The feature of total communication that distinguishes it from oral communication is that oral speech is combined with signing in English word order (otherwise known as Signed Exact English). Oral communication does not use any signing. The control group was stratified by primary mode of communication because it has consistently been shown that it influences cochlear implantation outcome (i.e., Kirk, Miyamoto, Ying, Perdeu, & Zuganelis, 2002; Osberger et al., 1991; Sommers, 1991).

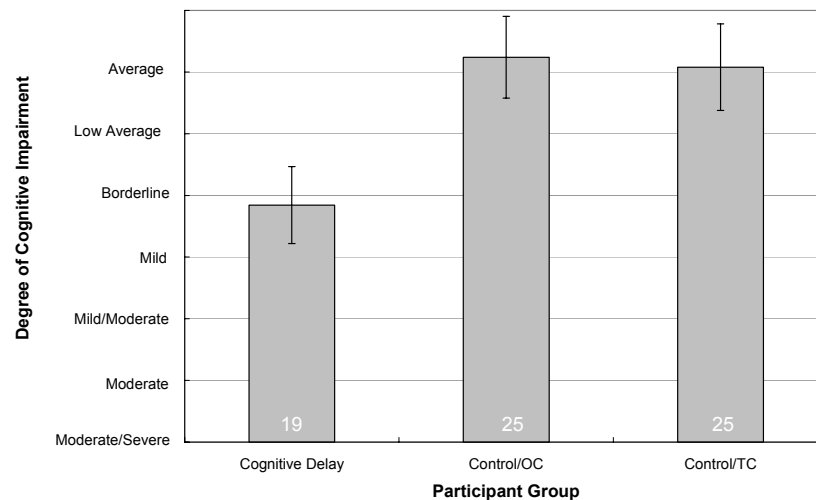


Figure 1. Mean degree of cognitive impairment and +1 standard deviation. The far left bar indicates the experimental group of children with cognitive delays, the middle bar indicates the control group of typically developing children who use oral communication, and the far right bar indicates the control group of typically developing children who use total communication. The numbers on each bar indicate the number of children in each group.

The mean estimate of cognitive functioning and +1 standard deviation for each group are displayed in Figure 1. The numbers on each bar indicate the number of participants included in each group. The oral communicators with no additional disabilities and the total communicators with no additional disabilities both had mean estimates of cognitive functioning in the average range. The average estimate of cognitive function for the children with cognitive delays was in the borderline to mildly impaired range. The variance in cognitive functioning across groups was similar. The child with the most severe cognitive impairment in the sample had a mild to moderate degree of cognitive impairment. Clearly, our group of cognitively impaired children did not have severe degrees of cognitive disability. We had children implanted at our center with more severe cognitive impairments. However, these children had other disabilities. Thus, they were not included in the experimental group because we wanted as homogenous groups as possible.

Participant Demographics

Table 1 displays pertinent demographic information for the participants. Specifically, mean and +/- 1 standard deviation data (in parentheses) are displayed for each group's age at onset of deafness, age at implantation, and duration of deafness prior to implantation. The proportion of children who used oral communication and the proportion of children who used total communication are also shown. The children in the experimental group were implanted about 1 year later than were those in the control group who used oral communication, and about 6 months later than were those in the control group who used total communication. Therefore, the children in the experimental group were chronologically older at each testing interval than were the children in the control group. Because age at onset was similar across groups, they also had longer periods of deafness without electrical stimulation relative to the children in the control group. A slightly larger proportion of the children with cognitive delays used total communication than used oral communication (58% total versus 42% oral communication).

Group	N	Age at onset of deafness (months)	Age at implantation (months)	Duration of deafness (months)	Proportion of oral communicators (percent)	Proportion of total communicators (percent)
Cognitive Delay	19	0.00 (0.00)	37.68 (14.64)	37.68 (14.64)	42%	58%
Control Oral Communicators	25	0.00 (0.00)	25.72 (12.79)	25.72 (12.79)	100%	0%
Control Total Communicators	25	1.88 (6.02)	31.96 (13.62)	30.08 (13.04)	0%	100%

Table 1. Demographic information for the experimental group and the two control groups.

Test Battery

Test of Auditory Skill Development. The Infant-Toddler Meaningful Auditory Integration Scale (IT-MAIS; Zimmerman-Phillips & Osberger, 1997) is a tester-administered parental/caregiver questionnaire. The questions are intended to prompt answers that functionally describe a child's use of her/his sensory aid(s) and her/his auditory behavior in response to speech and environmental sounds. Scores range from 0 to 40, with higher scores reflecting more advanced auditory skill development.

Tests of Word and Sentence Recognition Presented Auditory-only. The Grammatical Analysis of Elicited Language – Pre-Sentence Level Test (GAEL-P; Moog, Kozak, & Geers, 1983) was adapted to be used as a closed-set speech perception measure. Children are familiarized with all 30 test objects in an auditory-plus-visual mode. During testing, target words are presented live-voice through the auditory channel only. During each trial, children are presented with four objects, one of which is the target word. They are required to respond by indicating the object that corresponds to the target. These responses are scored by percent correct. Chance performance is 25% correct.

The Mr. Potato Head Task (Robbins, 1994) is presented via live voice and is scored for both word and sentence correct. Mr. Potato Head is a children's toy that consists of a "potato" body, body parts, and accessories. The approximately 20 body parts and accessories can be physically manipulated and attached to the "potato" body. During the task, children are given auditory-only, sentence-length instructions on how to assemble the toy. For example, "Give him some green shoes." Two scores are derived from performance on this task: sentence and word correct scores. The sentence score is based on the number of commands (out of 20) correctly carried out, whereas the word score is based on the number of key words (out of 20) identified (regardless of whether the full command was carried out or not). For example, the child would get the word correct if she/he picked up the green shoes, but would not get the sentence correct if she/he did not put them on the "potato" body in the example above. The key word task is considered closed-set because the child could select 1 of the 20 body parts or accessories by chance; therefore, chance performance is 5% correct. The sentence-level task is considered open-set, because the child cannot correctly carry out the command merely by chance.

Test of Multimodal Word Recognition. The Pediatric Speech Intelligibility Test (PSI; Jerger, Lewis, Hawkins, & Jerger, 1980) was modified and used as a closed-set word and sentence recognition test. Both portions use plates that contain six pictures. The child is to point to the picture of either the target word or the picture that the target sentence described. Few data were collected on sentence recognition. Therefore, only word recognition data will be presented. For the purpose of testing multimodal speech perception, the PSI is given in three modes: auditory-only, visual-only, and auditory-plus-visual. The amount of auditory-plus-visual enhancement is the difference between percent correct in the auditory-plus-visual condition and either the auditory-only or the visual-only conditions.

Tests of Receptive and Expressive Language. The Peabody Picture Vocabulary Test, Third Edition (PPVT-III; Dunn & Dunn, 1997) measures receptive vocabulary development and is given in each child's primary mode of communication (total or oral). This means that the stimuli are presented in the auditory-plus-visual mode for the children who use oral communication and in the auditory plus sign mode for children who use total communication. This is in contrast to the speech perception tests, where all the children are presented the stimuli in the auditory-only mode, in which no signing or visual cues are used. The PPVT-III requires the child to correctly point to one of four line drawings in response to a spoken word (for an oral communicator) or a spoken and signed word (for a total communicator). A receptive vocabulary age is derived and is then converted into a receptive language quotient (receptive vocabulary age divided by chronological age). A receptive language quotient of 1.0 indicates that the child's language age and her/his chronological age are equal. In other words, her/his receptive language skills are appropriate for her/his age. Receptive language quotients below 1.0 indicate that her/his receptive language ability lags behind what would be expected based on her/his chronological age; receptive language quotients greater than 1.0 indicate that her/his receptive language skills exceed what is expected based on her/his chronological age.

The Reynell Developmental Language Scales (RDLS; Reynell & Huntley, 1985) assesses receptive and expressive language abilities separately. Both the receptive and the expressive portions of the RDLS are administered in the child's preferred communication mode (total or oral communication). The receptive portion has 62 items arranged into 10 sections and requires the child to comprehend a hierarchy of language structures ranging from identifying named objects to inferencing and vocabulary/grammar. The expressive language portion assesses the child's ability to express a hierarchy of language structures ranging from object labeling to complex instructions through the use of 62 items in 10 sections. As with the PPVT, the receptive and expressive vocabulary ages are derived and converted into receptive and expressive language quotients, respectively (receptive/expressive vocabulary age divided by chronological age).

Procedure

Children were administered the test battery prior to implantation and at approximately regular 6-month intervals after the cochlear implant was first stimulated. Not every child in each group was tested with every speech and language test at every test interval, due to time constraints, lack of ability to maintain attention for all the tests, or missed appointments. All tests were administered and scored by licensed speech-language pathologists with training in working with deaf children with cochlear implants. Testing was conducted in a quiet room using live-voice presented at approximately 70 dB SPL. Again, the speech perception tests were administered in an auditory-only modality, whereas the language tests were administered with auditory and visual cues (for those who use oral communication) or with auditory, visual, and sign cues (for those who use total communication). In contrast to test administration, test *instruction* for both speech perception and language tests was carried out in the child's primary mode of communication. Finally, signed and spoken responses were accepted for all the tests given.

Results

To increase power we collapsed data from blocks of two consecutive 6-month intervals and reported mean scores by year, because not all children were tested at every 6-month interval on every test. If a child were only tested once in a 1-year span, we used that test score in calculating mean performance; if a child were tested twice, we used the latter score in our calculations. In doing this, we were able to include more data points per interval. All the figures displayed in this section have similar layouts. Each figure or panel in a figure displays data for a single test condition. Mean group data and +1 standard deviation are displayed in histograms: black-filled bars indicate the performance of the experimental group made up of the children who are cognitively delayed; the unfilled bars indicate the performance of the children without additional impairments (control group) who use oral communication; and the gray-filled bars indicate the performance of the children without additional impairments (control group) who use total communication. Each series of bars are displayed in 1-year intervals beginning with the pre-implantation scores (0 years of device use) and continuing up to 3 years of device use, depending on the test. The numbers on each bar indicate the number of participants tested from that particular group for the given 1-year interval.

For each test given, the data were entered into a two-way Analysis of Variance (ANOVA) with one repeated measure. The between-participant factor was participant group (Cognitive Delay, Control/OC, Control/TC) and the within-participant factor was years of use (0, 1, 2, and 3, depending on the amount of data collected at each interval).

Auditory Skill Development

Infant-Toddler Meaningful Auditory Integration Scale. Figure 2 displays average group data for the IT-MAIS. Prior to implantation all groups performed similarly. After 1 year of implant use, all three groups demonstrated significant improvement in auditory skill development [$F(1, 28) = 67.67, p < 0.001$], with the children without additional impairments who use total communication performing similarly to those who were cognitively impaired. The children without additional impairments who use oral communication outperformed those who use total communication and the children with cognitive delays. However, these differences were not statistically significant. These results indicate that parents of implanted children with cognitive delays and those of implanted children without such delays report similar levels of auditory skills in their children prior to implantation and 1 year after device use.

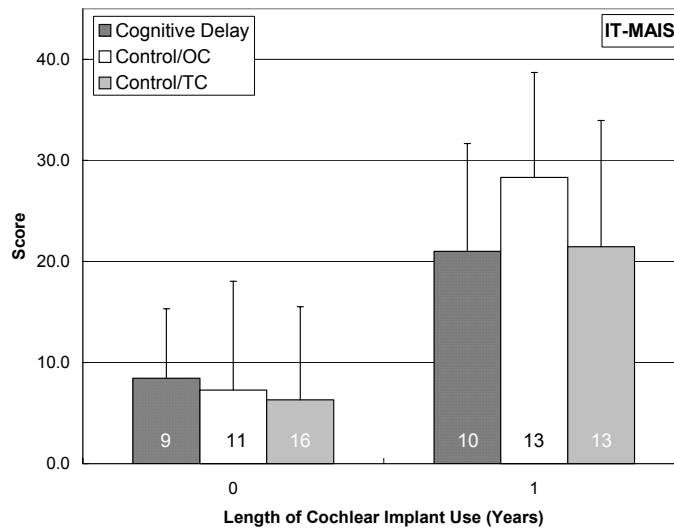


Figure 2. Mean group scores and +1 standard deviation on the IT-MAIS. The dark gray-filled bars represent the performance of the children with cognitive delays, the unfilled bars represent the performance of the typically developing children who use oral communication, and the light gray-filled bars represent of the typically developing children who use total communication. The numbers on each bar indicate the number of children tested in each group.

Word and Sentence Recognition Presented Auditory-only

The Grammatical Analysis of Elicited Language – Pre-Sentence Level Test. The top panel of Figure 3 displays average group performance on the GAEL-P. Prior to implantation all the groups scored near chance (25% correct). Performance for all three groups significantly improved over two years of device use [$F(1, 24) = 52.68, p < 0.001$]. After 2 years of experience with a cochlear implant, the average score for all the groups was near 50% correct. The small difference in performance between the groups was not statistically significantly.

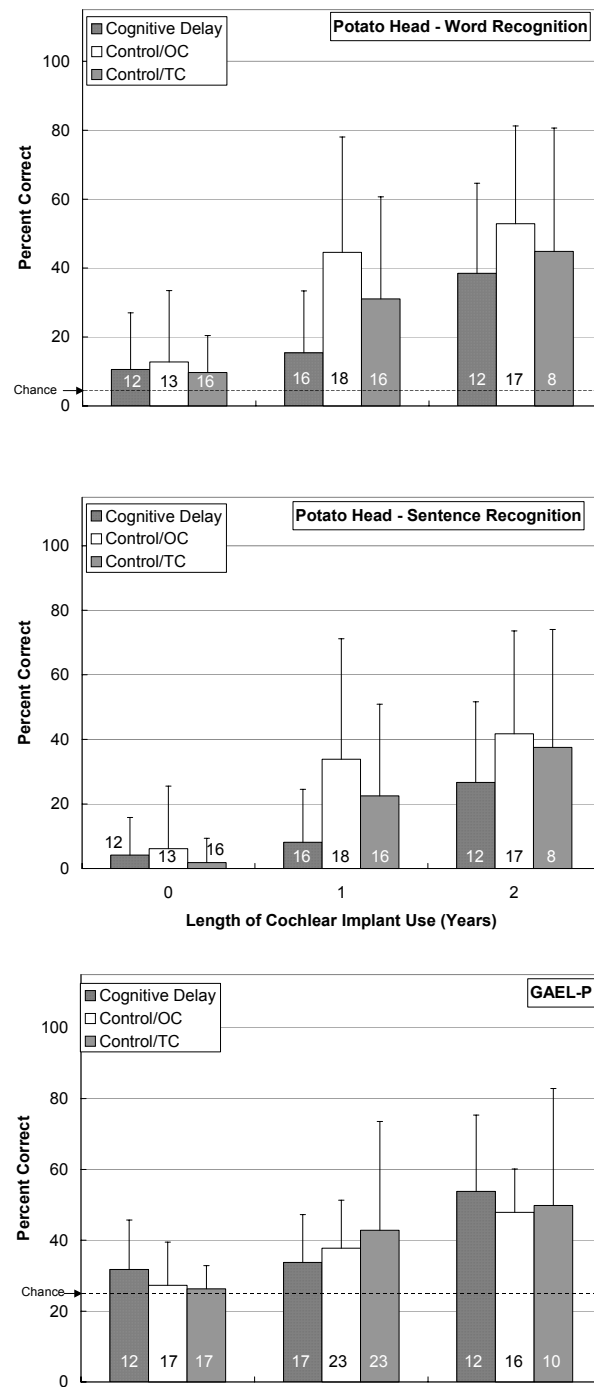


Figure 3. Mean group performance and +1 standard deviation on auditory-only tests of word and sentence recognition. The top panel displays word recognition performance on the GAEL-P, the middle panel displays word recognition performance on the Mr. Potato Head Task, and the bottom panel displays sentence recognition performance on the Mr. Potato Head Task. The dark gray-filled bars represent the performance of the children with cognitive delays, the unfilled bars represent the performance of the typically developing children who use oral communication, and the light gray-filled bars represent of the typically developing children who use total communication. The numbers on each bar indicate the number of children tested in each group.

Mr. Potato Head Task. Average group performance on the word recognition portion of the Mr. Potato Head Task is shown in the middle panel of Figure 3. All three groups performed slightly above chance prior to implantation and performance for all three groups significantly improved over 2 years of device use [$F(1, 22) = 74.13, p < 0.001$]. After 1 year of cochlear implant experience, there was a trend for the children without cognitive delays who use oral communication to perform better than their counterparts who use total communication and far better than those with cognitive delays. By 2 years of device use, the gap in performance between the groups began to close, although there was still a trend for children without cognitive delays who use oral communication to outperform those who use total communication, who in turn perform better than the children with cognitive delays. The difference in performance between the groups on the word recognition portion of the Mr. Potato Head Task was not statistically significant. Average group performance on the sentence recognition portion of the Mr. Potato Head Task is displayed in the bottom panel of Figure 3. Sentence recognition ability was poorer than word recognition ability for all three groups. Prior to implantation all the groups performed similarly and significantly improved through 2 years of device use [$F(1, 22) = 56.06, p < 0.001$]. Similar to their performance on the word recognition portion of the task, there was a trend for children without additional impairments who use oral communication to have higher sentence recognition scores than did those who use total communication or who are cognitively impaired. The children with cognitive delays performed more poorly as a group than did the typically developing children who use either oral or total communication. Although this difference did not reach levels of statistical significance, there was an interaction between length of device use and participant group [$F(2, 34) = 3.52, p = 0.04$].

These results indicate that children with and without cognitive delays improved in their word recognition skills over a 2-year period after implantation and did so at nearly similar rates. Further, for sentence recognition, there was an interaction between whether a cognitive delay exists and length of device use, with the improvement in sentence recognition occurring later for the children in the cognitively impaired group than it did for the typically developing children.

Multimodal Word Recognition

Pediatric Speech Intelligibility Test – Word Recognition. Group data on the auditory-only condition of the PSI are displayed in the top panel of Figure 4. Prior to implantation, none of the children with cognitive delays were able to identify any of the words presented on the PSI in the auditory-only modality, while some of the children without additional impairments could. After 1 year of device use, children with cognitive delays were able to correctly identify 13% of the words presented on average, while the children without cognitive delays who used oral communication could correctly identify 52% of the words on average. The mean score on the PSI presented auditory-only for the typically developing children who used total communication was 27% words correct. After 2 years of device use, the children with cognitive delays improved to over 60% correct on average, whereas the typically developing children who use oral communication scored 93% correct on average. The typically developing children who use total communication improved in the second year of device use, but to a much smaller degree: their average performance was just under 40% correct, although only 4 children were tested at this interval from this group. The variance in performance was quite large for all the groups and by 2 years of device use, few children were tested. For these reasons there was no significant difference in performance between the groups, although all groups improved significantly over time in their auditory-only word recognition on the PSI [$F(1,12) = 55.90, p < 0.000$].

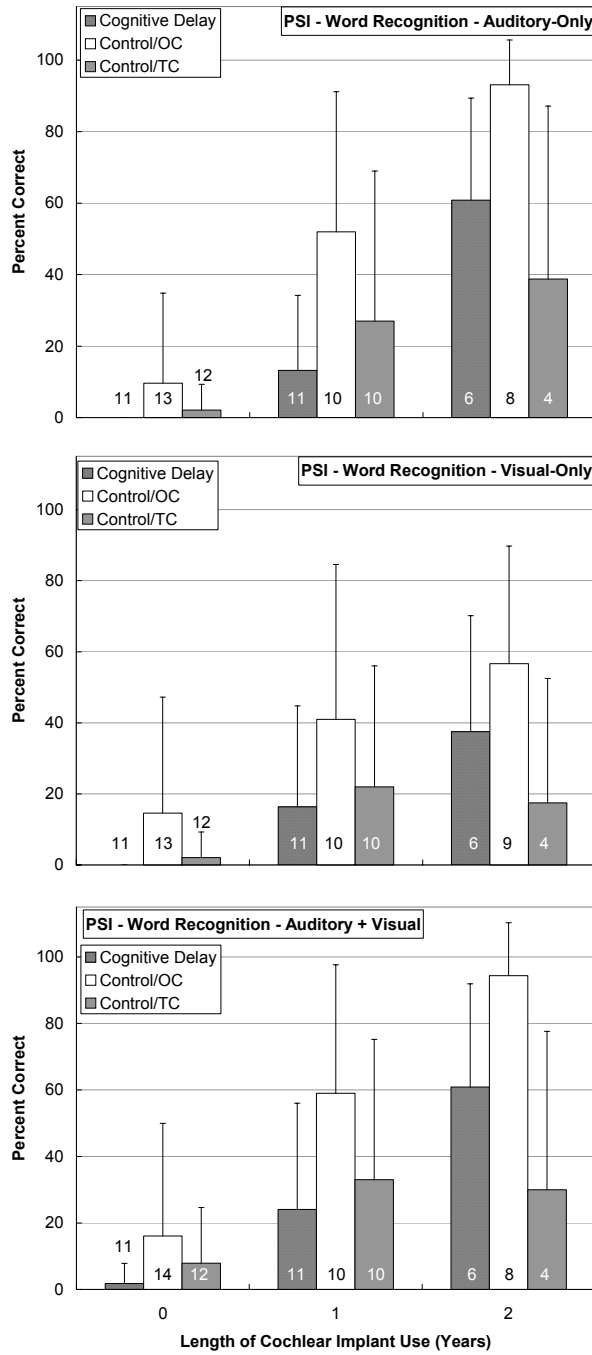


Figure 4. Mean group performance and +1 standard deviation on multimodal tests of word recognition. The top panel displays auditory-only performance on the PSI, the middle panel displays visual-only performance on the PSI, and the bottom panel displays auditory-plus-visual performance on the PSI. The dark gray-filled bars represent the performance of the children with cognitive delays, the unfilled bars represent the performance of the typically developing children who use oral communication, and the light gray-filled bars represent of the typically developing children who use total communication. The numbers on each bar indicate the number of children tested in each group.

Average group data for PSI presented in the visual-only mode are displayed in the middle panel of Figure 4. The pattern of data is similar to the auditory-only condition. However, the overall performance was poorer in the visual-only relative to the auditory-only mode. Again, performance improved significantly over time [$F(1,13)=15.76$, $p=0.002$], with no significant difference between groups.

Finally, average group data for PSI in the auditory-plus-visual mode are displayed in the bottom panel of Figure 4. Again, performance for all groups improved significantly over time [$F(1,12) = 39.73$, $p < 0.000$], with no significant difference between groups. Prior to implantation, all three groups showed a slight to modest improvement in word recognition with the presentation of both auditory and visual information relative to the auditory- or visual-only conditions (between 2 – 6% improvement in scores). After 1 year of cochlear implant use, having access to both auditory and visual cues had a greater impact on the children's performance than it did prior to cochlear implantation. Relative to the auditory-only condition, average scores in the auditory-plus-visual mode increased by 11% for the children with cognitive delays, 7% for the typically developing children who use oral communication, and 6% for the typically developing children who use total communication. By 2 years of device use, the children with cognitive delays and those without delays who use oral communication did not show any auditory-plus-visual gain (meaning that their performance was no better in the auditory-plus-visual condition than in the auditory-only condition). Finally, only four typically developing children who use total communication were tested after 2 years of device use. The average score for these children actually decreased by 8% with the combination of auditory and visual information relative to when only auditory information was available. Few children were tested during the second year of device use on the PSI in part because the test was designed for use with very young children and most of the children had outgrown it by 2 years post-implantation.

These results suggest that after 1 year of experience with a cochlear implant, all three groups were able to take advantage of auditory-plus-visual gain to some extent, but that by 2 years of experience this advantage disappears. Further, although the large variability in the data obscures any significant differences between groups, there is a trend for typically developing children who use oral communication to score higher on tests of multimodal word recognition than do typically developing children who use total communication or children with cognitive delays. Children with cognitive delays fared similarly to typically developing children who use total communication on this multimodal closed-set word recognition test.

Receptive and Expressive Vocabulary

Peabody Picture Vocabulary Test, Third Edition. The top panel of Figure 5 displays average group language quotient data on the PPVT. Recall that a language quotient is an index of how appropriate a child's language skills are for her/his chronological age. Language quotients of 1.0 indicate that a child's language skills are appropriate for her/his age, whereas those below 1.0 indicate that her/his language skills lag behind what is appropriate for her/his chronological age, and those above 1.0 indicate that they exceed those appropriate for her/his chronological age. Also, recall that the PPVT was given in each child's primary mode of communication, such that the children who use oral communication received the stimuli in the auditory and visual modes, whereas the children who use total communication received them in the auditory, visual, and sign modes. Further, children were allowed to give oral or signed responses. Performance across groups tended to be quite similar through 1 year of device use with all groups performing well below what would be expected based on their chronological ages. All groups' performances improved significantly over time [$F(1,20) = 13.90$, $p = 0.001$], indicating that their receptive vocabulary increased at a rate beyond what would be expected simply by typical development. After 2 years of device use, the group of typically developing children who use total communication

began to separate from the other children and really stood out after 3 years of device use with language quotients approaching 1.0. This difference between groups was significant [$F(2,20) = 7.16, p = 0.005$]. A planned post-hoc least significant difference test indicated that the typically developing children who use total communication scored significantly higher than those that use oral communication ($p = 0.001$) and the children with cognitive delays ($p = 0.04$). There was no significant difference in receptive vocabulary recognition between the typically developing children who use oral communication and the children with cognitive delays.

Reynell Developmental Language Scales. The middle panel of Figure 5 displays average group language quotient data on the receptive portion of the RDLS. Preoperatively, all three groups performed similarly and well below what would be expected based on their chronological ages. Receptive language performance for all the groups improved significantly over time [$F(1,28) = 28.73, p < 0.000$], indicating that after implantation their receptive language skills improved at a faster rate than typical development predicts. Further, the difference between the groups approached significance [$F(2,28) = 2.677, p = 0.086$]. After using a cochlear implant for 1 year, the receptive language skills of the typically developing children who use oral communication were superior to those of the typically developing children who use total communication and to those of the children with cognitive delays. By 2 years of device use, the children in the control group scored similarly regardless of if they used oral or total communication and the children with cognitive delays had slightly lower language quotients on average than did the children with no additional impairments. After 3 years of cochlear implant use, the children without additional impairments who use total communication had an average language quotient greater than 0.7, whereas that for the children without additional impairments who use oral communication was just below 0.6, and that for the cognitively impaired children was just above 0.4. However, the number of typically developing children tested who use total communication was quite small ($N=3$). One reason for this is that the RDLS can be used with normally hearing children through age 6 and just past that age with children who have hearing loss. Many of the children had outgrown the test due to their age and/or language proficiency. Therefore, it is possible that the children who were tested at the later intervals in the groups with high attrition (e.g., the control group of total and control group of oral communication users) were those who were lower performers relative to those who were not tested. However, there were no significant differences between the average language quotients of the typically developing children at earlier test intervals who were not tested at later intervals relative to those who were tested at later intervals.

Average group language quotient data for the expressive portion of the RDLS are displayed in the bottom panel of Figure 5. The typically developing children who use oral communication had average expressive language skills that were closer to their chronological ages at each testing interval than did the typically developing children who use total communication or those who have cognitive delays. This was especially marked prior to implantation and after 1 year of device use. At years 2 and 3 after implantation, the typically developing children who use total communication and the children with cognitive delays began to show an increase in their expressive language skills at a faster rate than would be expected by typical development. Further, the total and oral communicators in the control group had similar average language quotients after 2 years of cochlear implant experience. No average expressive language gains beyond those seen during typical development were shown by the children without additional impairments who use oral communication, as evidenced by their average language quotient staying rather constant across testing intervals. When all four testing intervals were included in the ANOVA, the number of participants who were tested in each interval dropped to such a low number that neither of the main effects or the interaction was significant. Again, attrition in the later years of device

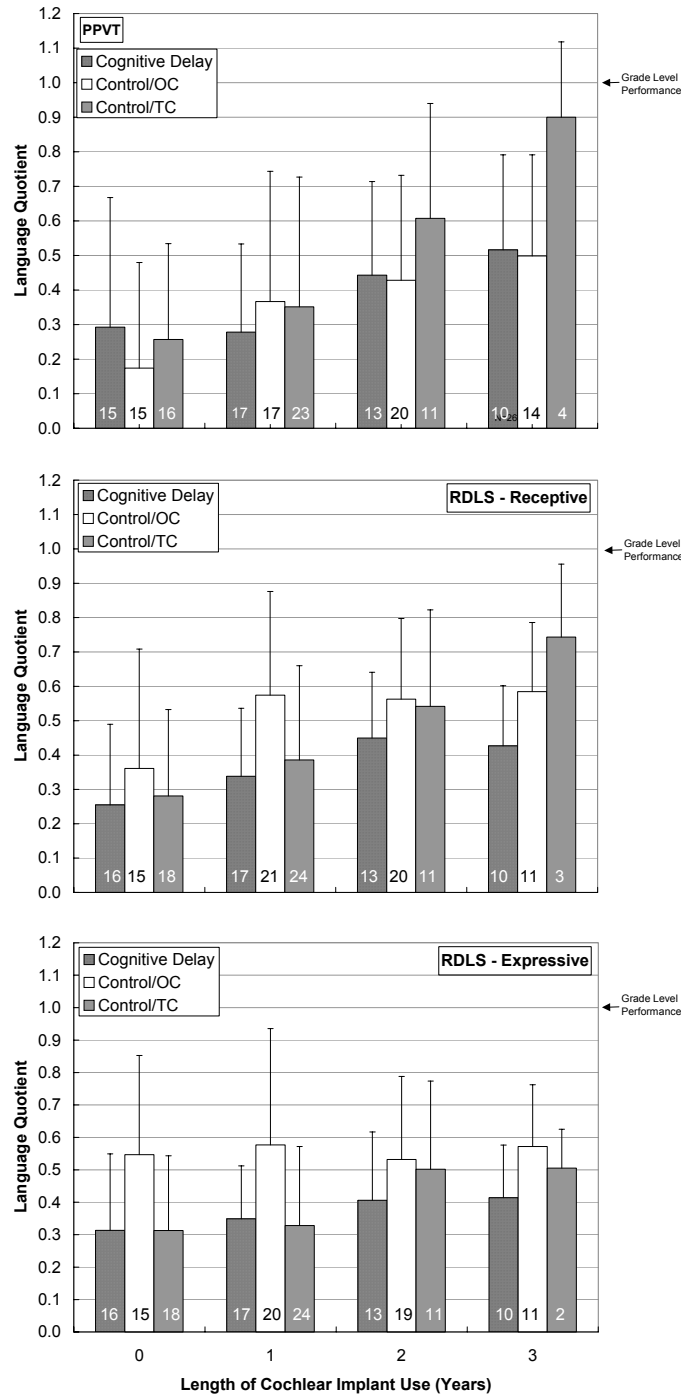


Figure 5. Mean group performance and +1 standard deviation on measures of receptive and expressive language. The top panel displays receptive vocabulary performance on the PPVT, the middle panel displays receptive language performance on the RDLS, and the bottom panel displays expressive language performance on the RDLS. The dark gray-filled bars represent the performance of the children with cognitive delays, the unfilled bars represent the performance of the typically developing children who use oral communication, and the light gray-filled bars represent of the typically developing children who use total communication. The numbers on each bar indicate the number of children tested in each group.

use is most likely due many of the typically developing children (those in the control group) not being administered the test due to their age and/or language proficiency. When the data from the third year of implant use are excluded, length of device use had a significant influence on expressive language skills in all the groups [$F(1,28) = 8.841, p = 0.006$], as did participant group [$F(2,28) = 4.841, p = 0.016$]. A planned post-hoc least significant difference test showed that the average expressive language quotient of the typically developing children who use oral communication was significantly higher than that of the children with cognitive delays ($p = 0.005$). There was no significant difference in performance between the typically developing children who use total communication and the children with cognitive delays, nor was there an effect of communication mode for the typically developing children.

These findings suggest that even with a rather large amount of variability across participants, these tests of receptive and expressive vocabulary and language are sensitive to differences between children with cochlear implants who have cognitive impairments and those who do not (PPVT and RDLS), and between typically developing children with cochlear implants who use different communication modes (PPVT only).

Discussion

The purpose of this investigation was to determine whether there are differences in the speech and language performance of deaf children with cochlear implants who have cognitive delays and deaf children with cochlear implants who are otherwise typically developing. The motivation for this work primarily comes from two observations: 1) nearly one-third of children with hearing loss in the United States have additional disabilities (Holden-Pitt & Diaz, 1998) and 2) there is no consensus on whether to implant children with severe-to-profound sensorineural hearing loss who also have other impairments. Previous attempts to address this question are limited due to few participants included in the sample and the inclusion of many disabilities that differentially impact communication. To better control for these factors, we used relatively strict inclusion criteria: onset of deafness before age 2.5 years and implanted with a current device and speech processing strategy before 5 years of age (except one child in the experimental group who was implanted at 5;11). More importantly, the experimental group included children who scored more than 1 standard deviation below the mean on tests of cognitive function and/or were identified by a psychologist as developmentally delayed and were not identified with any other disability (N=19). The control group consisted of children who scored within 1 standard deviation of the mean on tests of cognitive function and had no disability other than deafness (N=50). These strict criteria were meant to help create more homogenous groups than were used in previous studies. Even with these strict criteria, we still encountered a large amount of intersubject variability.

Although there was a trend for parents of typically developing children who use oral communication to report higher levels of auditory skill development on the IT-MAIS than typically developing children who use total communication and children with cognitive delays, these differences were not significant. These results suggest that parents of children with and without cognitive delays report similar levels of early developing auditory skills in their children prior to implantation and after 1 year of device use.

Implanted children with and without mild cognitive delays showed significant gains over time on all the speech and language tests administered. Although there was a trend for children with cognitive delays to have lower scores on tests of auditory-only word recognition than typically developing children, no significant differences were found between the groups, as measured by the GAEL-P and the Mr. Potato Head Task. This suggests that children with and without mild cognitive delays show similar improvements in word recognition skills over a 2-year period after implantation. A slightly different outcome was found for auditory-only sentence recognition. There was an interaction between the

presence of a cognitive impairment and length of device use, such that children with mild cognitive delays require more experience with their cochlear implants to achieve sentence recognition scores similar to typically developing children with cochlear implants.

On tests of word recognition presented in the auditory-only, visual-only, and auditory-plus-visual modalities, we did not find any significant differences in performance between the children with cognitive delays and those who are typically developing or between typically developing children who use different communication modes. However, it is likely that this was due to the rather high degree of variability across participants. There was a trend for typically developing children who use oral communication to have higher word recognition scores in any given modality on the PSI than their counterparts who use total communication and children with mild cognitive delays. The later two groups performed similarly on the PSI. All the groups of children demonstrated some degree of auditory-plus-visual gain after 1 year of device use, but did not show evidence of it after 2 years of device use. The lack of auditory-plus-visual gain in all the groups during the second year of device use is due in part to the large gains in auditory-only word recognition.

In contrast to the speech perception measures, significant differences were found between children who have cognitive delays and children without such delays on tests of receptive and productive language skills and receptive vocabulary skills. Although all the groups had significantly improved receptive vocabulary skills on the PPVT after cochlear implantation, the typically developing children who use total communication had significantly higher average receptive vocabulary language quotients than did those who use oral communication and children with cognitive delays. There were no significant differences in receptive vocabulary language skills between the children with cognitive delays and children who are typically developing who use oral communication. In contrast, there were no group differences found on the receptive language portion of the RDLS (although they were approaching levels of significance). An explanation for this discrepancy lies in the observation that the iconic nature of the vocabulary tested in the PPVT may give children who use total communication an advantage over those who use oral communication. An alternative explanation is that the RDLS is designed for use with normal-hearing children under age 6 and with children slightly older than age 6 with hearing loss. Therefore, it is possible that the children who were tested at the later intervals in the control group of oral and total communication users, where there was high attrition, were lower performers relative to those who were not tested. Although the average language quotients at early test intervals were similar between children who were tested at later intervals and those who were not tested at later intervals, it is still possible that the children who were not tested at later intervals indeed had outgrown the tests and only the low performers were being tested. Test materials that can be used with a wider age range and skill level are needed to follow children longitudinally to address this alternative explanation.

The typically developing children who use oral communication had significantly better expressive language skills than did the children with cognitive delays out through 2 years of device use. Although the difference between the typically developing children who use oral versus total communication appears to be large, especially after 1 year of device use, it failed to reach significance on post-hoc analysis. These findings suggest that even with a rather large amount of variability across participants, these tests of receptive vocabulary and receptive and expressive language are sensitive to differences between children with cochlear implants who have cognitive impairments and those who do not, and that the PPVT (a test of receptive vocabulary) is sensitive to differences between typically developing children with cochlear implants who use different communication modes.

These results indicate that children with mild cognitive impairments benefit from cochlear implants in their ability to perceive speech and their reception and use of language, although it may be reduced relative to their typically developing peers with cochlear implants to some extent, particularly in

domains that require higher level skills, such as sentence recognition and receptive and expressive language. Although Pyman et al. (2000) used different tests, our results are consistent with theirs which propose that children with motor and/or cognitive delays tended to have slower development of certain aspects speech and language that employ higher level speech processing abilities. Further, our results slightly differ from Waltzman et al. (2000), who reported that the children with multiple impairments who received cochlear implants were less likely to be able to complete tests of speech and language and when they could had lower scores on average than typically developing children with cochlear implants. Although there was a tendency for this in our population of children, for the most part, the differences between groups failed to reach significance. Again, Waltzman et al.'s sample included children with a wide range of disabilities, whereas our experimental group only included children with mild cognitive delays and no other disabilities, making it difficult to compare our results with theirs.

In summary, our results suggest that deaf children with mild cognitive deficits be considered for cochlear implantation with less trepidation than has occurred up until this point. Although their speech and language gains may be tempered by their cognitive abilities, these limitations do not appear to preclude benefit from a cochlear implant on measures of speech and language development traditionally used with typically developing populations.

Future directions for research in this area include determining whether children with more severe degrees of cognitive impairment show similar trends in their speech and language development or if there is a critical level of cognitive ability needed to derive benefit from the stimulation received by a cochlear implant. There is a need to determine whether certain types of therapy may help children with mild cognitive impairments use the information they receive from their cochlear implants for higher level abilities, such as sentence recognition and reception and use of language. There is also a need to determine the impact of other disabilities, such as autism, low vision, physical impairments, and combinations of disabilities on speech and language development in deaf children with cochlear implants. Additionally, more work may be needed to help better define what benefit is in populations that may not demonstrate gains on traditional measures of speech and language development. Finally, longitudinal testing with materials suitable for wider age and skill ranges may help detect finer differences between implanted children with and without cognitive delays.

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