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The Ear Is Connected to the Brain: Some New Directions in the Study of Children with Cochlear Implants at Indiana University

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Abstract

Since the early 1980s, the DeVault Otologic Research Laboratory at the Indiana University School of Medicine has been on the forefront of research on speech and language outcomes in children with cochlear implants. This paper highlights work over the last decade that has moved beyond collecting speech and language outcome measures to focus more on investigating the underlying cognitive, social, and linguistic skills that predict speech and language outcomes. This recent work reflects our growing appreciation that early auditory deprivation can affect more than hearing and speech perception. The new directions include research on attention to speech, word learning, phonological development, social development, and neurocognitive processes. We have also expanded our subject populations to include infants and children with additional disabilities

Key Words: Auditory rehabilitation, cochlear implants, diagnostic techniques, pediatric audiology, speech perception

Abbreviations: AD = additional disabilities; BIT = Beginner's Intelligibility Test; BRIEF = Behavior Rating Inventory of Executive Function; EF = executive function; HVHP = Hybrid Visual Habituation Procedure; OT = Optimality Theory; PFAS = Pediatric Functional Assessment Scale; PUP = Prosodic Utterance Production; VHP = Visual Habituation Procedure; VPP = Visual Preference Procedure

"The ear is connected to the brain" has become a kind of mantra in the DeVault Otologic Research Laboratory at the Indiana University School of Medicine. This phrase reflects our growing acknowledgment that severe-to-profound deafness and associated interventions such as cochlear implantation affect not only hearing, speech perception, and spoken language development but also general neurocognitive and psychosocial development. The effects of early auditory deprivation and subsequent cochlear implantation on outcomes have been a focus of the laboratory since it was established by Richard Miyamoto in the early 1980s. The early days of the laboratory produced seminal work on speech perception and articulation skills of children with cochlear implants (Robbins et al, 1985; Miyamoto et al, 1986; Robbins et al, 1988; Carney et al, 1993; Miyamoto et al, 1989; Osberger, 1990). During the 1990s, the scope of the research expanded to include higher-level speech perception (e.g., lexical organization) and language outcomes, phonological development, and auditory working memory capacity (Kirk et al, 1995; Robbins, Osberger, et al, 1995; Kirk et al, 1997; Miyamoto et al, 1997; Chin et al, 2000; Chin and Pisoni, 2000; Kirk et al, 2000; Pisoni and Geers, 2000). Over the last decade, the scope of the research has expanded in several new directions. Our research

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program now includes younger cochlear implant recipients than before—often before 12 mo of age. It also now includes several investigations of cognitive and psychosocial development, executive function, and theory of mind.

This review of our work is organized into four parts: speech and language development, input and psychosocial development, neurocognitive processes, and development in children with additional disabilities. In some cases, we will discuss the methods that we selected or invented to study that domain. Our goal in writing this article is not only to provide a review of interesting recent findings in new domains from our research program but also to stimulate additional innovation by explaining why we view these new domains as important and describing our approach to investigating them.

SPEECH AND LANGUAGE DEVELOPMENT

Speech and language development has been a central focus of cochlear implant research teams since the beginning of the cochlear implant research field (Chin and Svirsky, 2006; Waltzman, 2006). Much of the research has employed established conventional clinical speech and language assessment tools—measures of vocabulary, spoken word recognition, articulation, and omnibus measures of receptive and expressive language—to investigate the effects of variables such as age at implantation, communication method, and amount of residual hearing on speech and language outcomes. These measures provide valuable information about the effects of demographic variables on speech and language outcomes. However, they do not provide much insight into the underlying processes of language development. To learn more about how these processes unfold and develop after cochlear implantation, much of our more recent work has involved measures of language processing, such as word learning, phonological coding, and lexical access. Also, we are investigating speech perception more thoroughly and at younger ages than our earlier research.

Speech Perception

As the age range for cochlear implantation has broadened to include infants as young as six months of age,[1] our research team has focused increasingly more attention on speech perception skills during infancy. This approach has involved implementing several experimental methodologies borrowed from fields within general developmental science and developing new methodologies. We have also broadened the types of speech perception skills we investigate to include attention to speech, audiovisual speech perception, and others.

Assessing speech perception skills in infants presents unique challenges. Infants are not able to follow verbal instructions, which precludes the use of many of the standard assessment tools commonly available. We addressed this challenge by borrowing and adapting methodologies used by developmental scientists to study speech perception and language development in typically developing infants with normal hearing (Golinkoff et al, 1987; Werker et al, 1998). In February 2001, we established the first infant speech perception and language laboratory for infants with cochlear implants. Since the founding of the Infant Laboratory, one of the primary topics of investigation has been infants' ability to discriminate speech sounds after implantation. We began investigating speech discrimination using a methodology commonly used by developmental scientists: a habituation/dishabituation procedure called the Visual Habituation Procedure (VHP).

The VHP relies on the process of habituation, a very basic response found in all animal species (Wood, 1969; Davis, 1970; Duerr and Quinn, 1982). During each trial of the VHP, the infant is presented with a repeating speech sound and a visual display of a checkerboard pattern; the trial continues until the infant looks away from the display for more than 1 sec. The trials continue until the infant's looking time across trials decreases to reach a habituation criterion. Then the infant is presented with the same checkerboard pattern for two more trials, once with the same speech sound and once with a novel speech sound. Infants who can discriminate the speech sounds will usually dishabituate to the novel speech sound (i.e., look longer).

The response to novelty in the VHP does not have to be conditioned. The only learning involved in this process is encoding. Other methodologies rely on the infant learning a contingent relationship between a change in a sound and the onset of a reinforcer and conditioning the infant to turn his or her head in response to the change in sounds (e.g., Kuhl, 1985; Werker et al, 1997; Eisenberg et al, 2004; Tsao et al, 2004). Those methodologies can provide an excellent assessment of infants' speech discrimination skills when infants are successfully conditioned, but it is often very difficult to condition infants to a change in sound (Werker et al, 1998).[2] Because of the high cognitive demands of conditioned head turn paradigms, we opted for using the less cognitively demanding VHP to assess speech discrimination.

An important limitation of the traditional VHP is that there are too few trials to determine with statistical reliability whether any individual infant shows discrimination for any particular contrasts. There is only one novel and one old trial. The reason for this is that the experience of a stimulus as novel is, by definition, a very transient phenomenon. In order to adapt the VHP so that it could provide information about individual infants, we added novel and old trials but modified two things to make the repetitions of the "novel" trials maintain their novelty. First, we presented fewer novel trials (4) than old trials (10), and the novel trials, rather than consisting of repetitions of the same speech sound, consisted of alternations of the novel and old speech sound. We called this methodology the Hybrid Visual Habituation Procedure (HVHP). Testing of the HVHP with normal-hearing infants indicated that it was reliable and more sensitive to basic discrimination abilities than several other variants of the VHP (Houston et al, 2007).

We have now used the HVHP to investigate discrimination of several speech contrasts in infants with cochlear implants as well as those with milder degrees of hearing loss and those with no hearing loss (Horn, Houston, et al, 2007; Houston et al, 2007). We found that the HVHP is a reliable and robust tool for infant speech discrimination when the sound contrast is easy (i.e., involving changes in multiple features—"seepug" versus "boodup," for example). We have had mixed results with more difficult phonetic contrasts (i.e., single-feature contrasts). On the one hand, we have preliminary findings suggesting that with more difficult contrasts, performance on the task predicts later measures of vocabulary development, suggesting predictive validity (J.Y. Ting, D.M. Houston, R. Holt, and R.T. Miyamoto, unpublished data). On the other hand, we also found that when the HVHP is used for more difficult contrasts it shows poorer test-retest reliability.

In order for a test to be clinically useful, it needs to be both reliable and valid. Thus, we are still working on improving this methodology. One direction we are taking is combining looking time measures with measures of heart rate. Infants' heart rate decelerates when they go from a state of inattention to sustained attention (Richards, 1988). Thus, infants' heart rate should decelerate when they notice and attend to a change in a stimulus, independently of whether they orient to the stimulus. By collecting both voluntary (orienting) and involuntary (heart rate deceleration) responses, we hope to develop more sensitive and more reliable measures of speech discrimination that can be used with infants of varying cognitive abilities.

Audiovisual Speech Perception

Another aspect of speech perception we are exploring is audiovisual speech perception. The ability to integrate auditory and visual information is an important aspect of speech perception for listeners with normal hearing and may be even more important for those with hearing loss. In the past few years, we have investigated audiovisual speech perception skills in children who have profound hearing loss prior to learning spoken language.

In one study, we investigated the development of audiovisual speech perception in profoundly deaf children prior to implantation up to 5 yr postimplantation (Bergeson et al, 2005). The children were administered the Common Phrases test of sentence comprehension (Robbins, Renshaw, et al, 1995) in three presentation conditions: auditory alone, visual alone, and audiovisual. As expected, children improved on this test across time and improved more in the auditory-alone and audiovisual conditions as compared to the visual-alone condition. We also found that children enrolled in oral communication education environments outperformed children in total communication environments in all conditions, even at the pre-implantation period. Moreover, pre-implantation lipreading skills were significantly correlated with performance on speech perception outcome measures 3 yr postimplantation. The results suggest that very early audiovisual perception abilities play a role in the development of spoken language.

These findings point to the importance of audiovisual speech perception at young ages. We have since conducted several studies on various aspects of audiovisual speech perception in infants who receive cochlear implants. In one study, we presented infants with videos of static or dynamic (i.e., talking) faces either accompanied by speech or presented in silence (Ting and Bergeson, 2008). Six- to 13-mo-olds with normal hearing preferred to watch the dynamic-speech face the most and the static-silent face the least. Infants with cochlear implants, on the other hand, did not begin to show an audiovisual speech preference until approximately 1 yr following implantation suggesting a delay in audiovisual integration skills.

Despite the lack of initial preference for audiovisual over visual-alone stimuli, it is possible that infants with cochlear implants are still capable of integrating auditory and visual information. In a study of audiovisual speech integration using the Visual Preference Procedure (VPP), we presented infants with the same woman's face on two sides of a large-screen television monitor (Bergeson, Houston, et al, 2010). On one side, the talker repeated the word "back" and on the other the talker repeated the word "judge." We then presented the auditory word matched with only one of the faces and measured infants' looking time to the matching versus mismatching face. Five- to 13-mo-olds with normal hearing looked significantly longer at the matching face during the first block of trials but looked equally at the two faces during the second block of trials. Surprisingly, the infants with cochlear implants (13–38 mo of age; 1–24 mo of implant use) displayed exactly the opposite pattern. They looked equally at the two faces during the first block of trials but looked longer at the matching face during the second block. This suggests that audiovisual integration is a more effortful and less automatic behavior for infants who have experienced a period of auditory deprivation prior to receiving cochlear implants.

Recall that older pediatric implant users performed better on a test of audiovisual sentence comprehension if they had been in an oral communication rather than a total communication environment, even before receiving their cochlear implants. This finding, along with a qualitatively different pattern of performance on the audiovisual speech integration test by infants with implants, highlights the importance of early linguistic experience not only on audiovisual speech perception but also on general cognitive processing. One reason that children in oral communication environments might outperform children in total communication environments is competition for limited attentional and cognitive resources. That is, manual communication does not specify the same underlying articulatory gestures of the talker as compared to auditory or lipreading cues. We designed a study to begin to tease apart the competition effects of simultaneous auditory-oral and manual communication in infants under simulated conditions of hearing loss (i.e., noise) (Ting et al, 2011). Using the VPP, we familiarized 8.5-mo-old normal-hearing infants with repetitions of single words in either a speech-only or a speech + sign condition. We then presented the infants with speech-only passages, two of which contained the words presented in the familiarization phase, and two of which contained new words. Infants exposed to the speech-only condition, but not the speech + sign condition, looked at the familiar word passages significantly longer than the nonfamiliar word passages. This finding suggests that experience with total communication may have negative effects such as competition for processing resources, which potentially affect a wide range of spoken language outcomes in infants with hearing loss. However, more research is needed to determine the extent of such effects and how they affect infants and children with hearing loss rather than normal-hearing infants with simulated hearing loss, particularly those children who may have extensive experience with fluent total communication or those children who receive little benefit with hearing aids or cochlear implants.

Sensitivity to Lexical Stress

Most studies of speech discrimination focus on phonological contrasts that are meaningful for differentiating words. However, there are many other types of speech information that infants must discriminate and identify for reasons other than recognizing words. Sensitivity to lexical stress,[3] for example, is important for segmenting words from the context of fluent speech—at least in English (Jusczyk et al, 1999). In collaboration with Liat Kishon-Rabin and Osnat Segall at Tel-Aviv University, we are investigating the effects of language experience on lexical stress discrimination in both infants with normal hearing and deaf infants with cochlear implants. Words in modern Hebrew tend to end with a stressed syllable; words in English tend to begin with a stressed syllable. We have found that Hebrew-learning and English-learning infants with normal hearing perform differently on tests of lexical stress discrimination when using the same stimuli and same methodology, suggesting that the language input affects performance. By investigating stress discrimination in infants with cochlear implants, we can determine if language input affects their speech perception skills the way it does in infants without hearing loss. Moreover, sensitivity to the predominant stress pattern of the native language may predict language outcomes. We expect that sensitivity to the native stress pattern will correlate with vocabulary development because segmentation is thought to play a foundational role in spoken word learning.

Spoken Word Learning

As mentioned earlier, an exciting direction that our laboratory has taken is to move beyond collecting conventional clinical outcome measures to obtain additional measures of processing and learning. Investigating learning is important because it is a more direct measure of the basic capabilities of children than conventional endpoint outcome measures, which assess what children have learned already. Assessing children's ability to learn is important for determining children's ongoing needs. If educational needs were determined by outcome measures only, then children who scored within normal ranges might mistakenly be assumed to have learning abilities within normal ranges and have clinical services discontinued. This is a problem because the child may still have more difficulty learning than children with normal hearing and keep up only because of the additional services (e.g., intensive therapy, FM system, etc.).

One of the directions we have taken toward understanding the basic learning abilities of children with cochlear implants is to investigate word-learning skills in preschool-age children and in toddlers and infants. In our first study on word learning, we found that preschool-age children from excellent oral rehabilitation programs performed much more poorly on a word-learning task than age-matched children with normal hearing (Houston et al, 2005). The only children with cochlear implants that performed similarly to age-matched children with normal hearing were the two (out of 24) who received their implants under 1 yr of age. This finding led to a more systematic investigation of very early implantation on word-learning skills. We used the Intermodal Preferential Looking Paradigm (IPLP) (Golinkoff et al, 1987) to investigate word learning in children who received cochlear implants between 6 and 24 mo of age and who had 12 to 18 mo of implant experience. We found that children who received implants before 12 mo of age showed similar performance to age-matched children with normal hearing, whereas children who received implants between 12 and 24 mo did not (Houston et al, 2012). Moreover, we found that performance on the word-learning task predicted later vocabulary level but not speech perception outcomes, suggesting that word-learning abilities are an important foundational skill needed for later language development.

Speech Production and Intelligibility

Speech Production

Areas of speech production most commonly addressed have been articulation/phonology and speech intelligibility. Our earliest research addressed the efficacy of cochlear implants, concentrating on the measurement of speech production skills before and after cochlear implant surgery and on comparisons of speech produced by people using cochlear implants, conventional hearing aids, or tactile aids (Osberger et al, 1991; Osberger et al, 1993; Osberger et al, 1994). Results from these studies established that cochlear implants have no deleterious effects on speech production and that, given comparable hearing thresholds, cochlear implants offer more speech production benefits than conventional hearing aids (for all but those with the most residual hearing) and tactile aids.

Our more recent research on speech production still addresses speech intelligibility, but rather than examining exclusively surface articulation and phonological characteristics, we have begun to examine deeper aspects of phonological organization and structure, incorporating methods from theoretical linguistics (e.g., Chin, 2002; Kim and Chin, 2008; Sanders and Chin, 2009). Additionally, we have begun to make explicit and detailed comparisons of the speech production of children with cochlear implants with that of children and adults with normal hearing (Chin et al, 2003; Chin and Krug, 2004). This is due to the fact that, overall, speech production by children with implants has vastly improved over the years, due to such factors as younger ages at time of implantation surgery and newer speech processing technology.

Phonological Organization after Cochlear Implantation

Detailed examinations of phonological organization have been directed toward two main purposes. First, these studies investigate the robustness of language acquisition in cases where the input is degraded beyond the point at which most children acquire their native language. This research addresses the theoretical question of which aspects of a phonological system are highly dependent on specific input and which are relatively independent of specific input from the surrounding language. Second, these studies help to identify specific problematic areas in phonological organization that can be addressed during remediation. This addresses the clinical question of which phonological patterns, particularly error patterns, are common to either children generally or children with cochlear implants specifically, and which reflect idiosyncratic organization by individual children who use cochlear implants or subgroups of these children.

A basic organizational characteristic of any phonological system is the inventory of sound segments (consonants and vowels) that are used to construct larger units that convey meaning. These inventories of sound segments differ from one phonological system to another, and they serve as an important characteristic that defines each system as a potentially unique one. In addition, the inventory of sound segments forms the basis for higher-level analyses of any phonological system. These higher analyses include phonotactic regularities, that is, constraints on permissible sequences of segments in syllables, morphemes, and words. For clinical populations, they also include analyses of production error patterns with respect to an ambient system.

Analyses of consonant inventories have been a common and important approach to assessing clinical and developing phonological systems. We found that the consonant inventories of children with cochlear implants were not simply subsets of the ambient inventory but, rather, were unique to individual systems, with not only missing segments but also additional segments with respect to the ambient inventory (Chin, 2003). Moreover, qualitative differences were observed in the consonant inventories of children who used total communication and the children who used oral communication. Inventories of oral communication users tended to contain more English segments (e.g., alveolar fricatives, velar stops, velar nasals) than did the inventories of total communication users. Conversely, specific non-English segments, such as uvular stops, tended to occur in the inventories of total communication users more than in inventories of oral communication users.

A further difference between communication modes, this time in the phonotactic realm, was found in realizations of initial consonant clusters (Chin and Finnegan, 2002). In a group of 12 children, 48% of attempted clusters were produced correctly; however, the 6 children who used oral communication produced 75% of their clusters correctly, whereas the 6 who used total communication produced just 21% of their clusters correctly. Across the two groups, the patterns of consonant cluster realizations were similar and were also similar to patterns observed in children with normal hearing. Specifically, realizations of two-segment clusters were either one or two segments (i.e., there were no null onsets). Single-segment realizations generally retained the less sonorant of the two consonants, similar to what has been observed for children with normal hearing. Two-segment realizations, in which either or both of the sounds could be in error, generally reflected the overall realization patterns for the constituent singletons. Approximately 7% of the realizations exhibited epenthetic vowels, indicating knowledge of the constituent segments but lack of ability to realize them within a single onset.

The results from our analyses of consonant inventories (Chin, 2003) and consonant clusters (Chin and Finnegan, 2002) indicated an advantage of oral communication over total communication for acquisition of these two aspects of the English phonological system. In both cases, the phonological systems of children who used oral communication more closely resembled the ambient system (English) than those of children who used total communication. It is possible that the source of these differences is the relative proportion of resources that each group is able to devote to learning the details of ambientlike articulation (in the case of inventories) and of phonotactic sequencing (in the case of clusters).

Further investigation of initial consonant clusters was conducted within the framework of Optimality Theory (OT; Prince and Smolensky, 2004). A commonly cited characteristic of the language of children with cochlear implants is the large amount of variability. This is usually noted for ranges of single-index outcome measures, and various sources of this variability are adduced, such as age at implantation, duration of device use, socioeconomic status, and so forth. Observed variability in phonological analyses such as the ones conducted in our laboratory are somewhat different in being qualitative rather than quantitative. Some advantages of Optimality Theory (OT) in assessing this variability are its concentration on discovering a phonological source of phonological variability and its attribution of variability to (in principle) a single source: differences in constraint rankings (see below). Thus, what appears to be widespread random variability is actually tightly constrained, lawful variability when examined more closely within the context of the analytical approach.

OT is a nonderivational theory of phonology that defines relations between underlying representations ("inputs") and surface representations ("outputs"). Given a specific input, the OT grammar determines which of a set of potential outputs is an optimal one, based on the satisfaction of a set of universal constraints and a language-specific ranking of those constraints. "Faithfulness" constraints require that outputs preserve the properties of their corresponding inputs; "markedness" constraints require that outputs meet specified criteria of well-formedness. Consonant clusters violate a universal markedness constraint against complex outputs, but reduced cluster realizations violate a faithfulness constraint that outputs must resemble inputs. This tension between markedness and faithfulness constraints occurs throughout the phonologies of linguistic systems, and no less so in the systems of children who use cochlear implants (Chin, 2006, 2008). These conflicts are resolved by differential rankings of the constraints. Thus, in children's systems, which evidence reduction, the markedness constraint (against output clusters) is ranked more highly than the faithfulness constraint (prohibiting reduced outputs). Conversely, in mature systems, which evidence correct clusters, the faithfulness constraint (preserving clusters) is more highly ranked than the markedness constraint (against output clusters). Longitudinal analysis of clusters within this paradigm shows that at early stages, markedness constraints are ranked higher than faithfulness constraints. Subsequently, as both children and systems mature, markedness constraints are demoted so that faithfulness constraints are ranked higher. This general pattern of development, originally observed in children with normal hearing, applied equally as well for children with cochlear implants.

Also within an OT framework, we examined realization patterns for stop consonants (Chin, 2002) and consonant strengthening versus weakening (Kim and Chin, 2008). Realization patterns for stop consonants reflected the same types of differences between users of oral communication and users of total communication as for the larger consonantal inventories: total communication users tended to have fewer ambient stops and more nonambient ones. As with consonant clusters, differential constraint rankings explained differences between children in their realization patterns.

Analysis of specific speech errors is another important aspect of assessing children's speech production. As mentioned previously, detailed phonological analyses permit us to determine where specific differences between a children's system and the ambient system occur. Of course, these differences are perceived by listeners as speech errors, but important questions in this regard are whether errors are systematic or random, pervasive or idiosyncratic, principled or unprincipled. For example, manner of articulation errors for consonants are either strengthening (producing more consonantal, more occlusive segments; e.g., fricatives become stops) or weakening (producing less consonantal, less occlusive segments; e.g., fricatives become glides). Our examination of these processes in children with cochlear implants (Kim and Chin, 2008) showed that strengthening processes in children with implants were related to overall developmental patterns and reflected universal implications and markedness. Conversely, weakening processes tended to be more context sensitive and related to minimization of articulatory effort. An optimality theoretical analysis again revealed patterns similar to those displayed by children with normal hearing.

Speech Intelligibility after Cochlear Implantation

Speech intelligibility is important in assessing speech production in children with cochlear implants because it directly addresses the communicative function of language, unlike assessment of phonetic inventories, consonant clusters, strengthening and weakening patterns, and the like. To measure speech intelligibility, we have used the Beginner's Intelligibility Test (BIT) (Osberger et al, 1994), an imitative task with a transcription scoring procedure developed in our laboratory for use with children with cochlear implants. Using this measure, we established that improvements in speech intelligibility over time for children using cochlear implants were greater than would be expected for children using conventional hearing aids (Svirsky et al, 2000). We also determined that receiving cochlear implants early has a significant positive effect on the development of speech intelligibility, implying that the ability to develop intelligible speech declines as children mature without the benefit of good auditory input (Svirsky et al, 2007). However, we also determined that on the whole, children with cochlear implants are significantly less intelligible than children with normal hearing at both the same chronological age and the same hearing age (Chin et al, 2003). Furthermore, whereas children with normal hearing reach near-ceiling levels of speech intelligibility around the age of 4 yr, the development of intelligible speech in children with cochlear implants is considerably more gradual, with no ceiling being reached at chronological age 4 yr or hearing age 4 yr. Finally, when we compared speech intelligibility as measured by the BIT with the abilities to perceive and produce contrasts in minimal pairs, we found that contrastive perception and production were both correlated with overall speech intelligibility (Chin et al, 2001). This indicates that the ability to produce intelligible words in connected speech is related to the ability to contrast the consonants and vowels that make up those words.

Speech intelligibility is also affected by segmental and suprasegmental characteristics, including prosody. We have recently begun to explore the relationship between spoken intelligibility and prosody production of prelingually deafened children who use cochlear implants (Phan et al, 2011). We administered the BIT and the Prosodic Utterance Production (PUP) task to 6- to 10-yr-old children who had used a cochlear implant for 3–8 yr, and to a group of 4- to 14-yr-old children with normal hearing. We then asked a panel of naïve adult listeners to rate the intelligibility of the words in the BIT sentences and to identify the PUP sentences as one of four grammatical or emotional moods (declarative, interrogative, happy, or sad). The adults also rated how well they thought each child conveyed the designated mood in the PUP sentences. Not surprisingly, the children with normal hearing performed better on both the intelligibility and prosody tasks than the children with cochlear implants. Analyses of the mood ratings, however, revealed a significant difference between the two groups of children only for the interrogative mood category. In fact, both groups of children conveyed certain moods at least as well as the adult model speaker. This suggests that children who use cochlear implants are capable of perceiving and producing pitch and rhythm cues commonly associated with emotional mood, although they still have difficulty producing rising pitch at the end of questions.

Music Production

The two underlying features of speech prosody, pitch and duration, are also key components of music. Although children with cochlear implants have difficulty perceiving and producing musical melody (e.g., Nakata et al, 2006; Vongpaisal et al, 2006; Xu et al, 2009), they can discriminate pitch intervals in a nonmusical context (Vongpaisal et al, 2006). To get a sense of the potential relation between prosody and music production in children with cochlear implants, we asked the same groups of children to perform a series of melodic contours (up, down, up-down, down-up) and to sing the familiar song "Happy Birthday" (Bergeson, Chin, et al, 2010). We found smoother pitch progressions and more accurate pitch direction for children with normal hearing than for the children with cochlear implants. However, all children showed similar patterns of performance across the melodic contour categories, for example, performing the "up" contours more accurately than the "down" contours, similar to previous research on lexical tone production (Han et al, 2007; Zhou and Xu, 2008) and English intonation production (Peng et al, 2008). Moreover, children with cochlear implants produced melodic contours with a greater pitch range than children with normal hearing. These results were surprising given that previous studies had found compressed pitch ranges in songs produced by children with cochlear implants (Nakata et al, 2006; Xu et al, 2009).

For the song production task, we found more accurate pitch direction and pitch intervals for children with normal hearing as compared to children with cochlear implants. However, when we charted pitch accuracy across the duration of the song, we found that children with cochlear implants started off their songs well but then became much less accurate than children with normal hearing. This suggests there may be a neuro-cognitive component (e.g., working memory capacity) to the decreased song production abilities for children with cochlear implants.

Overall, these findings suggest that pediatric cochlear implant users have difficulty producing prosody in a variety of contexts. However, implant users did not have uniformly inaccurate pitch contours. Although the cochlear implant processing strategies do not code pitch very accurately, the fact that children with cochlear implants can produce some accurate pitch contours and can discriminate pitch intervals in a nonmusical context (as shown in Vongpaisal et al, 2006) suggests that there may be another explanation. It is possible that a period of auditory deprivation prior to implantation leads to the underdevelopment of systems in the brain typically associated with prosody and music perception and production.

Attention to Speech

Acquiring phonology, learning words, and tuning one's perceptual system to the properties of the ambient language all involve learning processes. And one of the best predictors of whether a person will learn something is the degree to which that person attends to the learning situation. However, very little is known about the extent to which attention is important for acquiring language in children with normal hearing—much less children with hearing loss. We do know that infants with normal hearing naturally attend to speech and even show specific preferences for speech over similarly complex nonspeech sounds at birth (Vouloumanos and Werker, 2007), but the role that these kinds of preferences play in language development is not clear (Jusczyk, 1997). If having regular and sustained attention to speech is important for language acquisition, then the degree to which deaf infants and children attend to speech after cochlear implantation is an important topic in its own right to investigate (Houston and Bergeson, forthcoming).

Over the last decade, we have been investigating deaf infants' sustained attention to various types of speech sounds after cochlear implantation compared to their age-matched peers with normal hearing. In our first experiment, we assessed attention to simple repeating speech sounds (e.g., "hop hop hop") produced in an adult-directed manner. Our dependent measure for sustained attention to speech was the different amounts of time infants spent looking at a checkerboard pattern while hearing a repeated speech sound versus hearing nothing. We assessed infants' attention to the repeating speech sounds at regular intervals from 1 day to 18 mo after implantation. At all post-CI intervals, infants showed significantly less sustained attention to speech than 6- and 9-mo-olds with normal hearing (Houston et al, 2003). When compared to their chronological age-matched peers, however, attention to speech by infants with cochlear implants as compared to peers with normal hearing changed as a function of amount of CI experience. During the earliest intervals (1 day to 1 mo) attention to speech by infants with cochlear implants was less than their peers with normal hearing. However, at later intervals (2 mo to 18 mo), their attention to speech was similar in both groups (Houston, 2009).

The findings that after a few months of cochlear implant experience, deaf infants' attention to speech is similar to their chronological age-matched peers but reduced compared to their hearing age-matched peers with normal hearing raise several questions about whether attending to speech like hearing age-matched peers is necessary to develop early speech perception skills. Preliminary analyses show a relationship between attention to speech on this task at 6 mo after implantation and spoken word recognition performance 2 to 3 yr after implantation, suggesting that greater attention to speech may facilitate acquiring better speech perception skills (Houston, 2009).

These findings led to a subsequent study in which we investigated the role of speech mode on infants' attention to speech after implantation (Bergeson et al, 2012). Infants were presented with three types of trials: infant-directed speech, adult-directed speech, and silence. At early post-cochlear implant intervals (i.e., before 1 yr of CI experience), infants showed no looking-time preferences for any of the trial types. After 1 yr of cochlear implant experience, infants did show a preference for infant-directed over adult-directed speech and silence but, unlike the previous study, these infants never showed a looking-time preference for adult-directed speech over silence. Moreover, their preference patterns differed from their chronological age-matched peers with normal hearing even after 6 mo of experience with their cochlear implants. The differences in results between the two studies may be due partly to differences in experimental design: three trial types versus two. Another reason may have to do with the fact that the stimuli in the latter experiment consisted of meaningful natural phrases (e.g., "Hello.... How are you today?"), which may have maintained normal-hearing infants' attention to speech more than implanted deaf infants because they may have been better able to extract meaning from the phrases.

These findings also led us to investigate deaf infants' visual attention skills. Preliminary findings so far suggest that deaf infants do not show reduced visual attention compared to chronologically age-matched peers with normal hearing, suggesting that the effects of early auditory deprivation on sustained attention may be limited to the auditory modality. Taken together, the findings suggest that at least some deaf infants show different patterns of attention to speech than infants with normal hearing, even after 6 mo or more of cochlear implant experience, and that attention to speech is related to speech perception outcomes.

Attending to speech may contribute to language development in multiple ways. The findings discussed in this section focused on how attending to speech may directly affect the quantity and/or quality of speech information encoded. But attending to speech may also help shape children's linguistic environment: attending to speech may reinforce communication to the child, which can further influence the child's language development and social interactions. The issue of the nature of the input to infants and children with cochlear implants is the focus of the next section.

SOCIAL DEVELOPMENT

Human infants are born into a richly structured sociocultural environment of caregivers, objects, and routines that create affordances for the emergence of uniquely human capacities such as spoken language (Tomasello, 1992). In addition, typically developing infants develop social-cognitive abilities such as social imitation, shared gaze, and joint attention that allow them to "tune in" to others starting from the first hours of life. From a sociocultural perspective of development, human cognition is socially—and linguistically—mediated through interactions with others using cultural artifacts within socially shared events that make up everyday life (Vygotsky, 1978). From birth the social and linguistic environments of children with normal hearing and a child's active participation in social exchanges are integral to language and vocabulary acquisition (Akhtar et al, 2001; Tomasello, 2003), sociocognitive development (Racine and Carpendale, 2007; Ontai and Thompson, 2008), and autobiographical memory (Fivush and Nelson, 2004) and form the foundation of the child's communicative competence (Tomasello, 1992). Infants with hearing loss experience a period of degraded and limited auditory access to social linguistic interactions, and a lack of opportunity to actively participate in social linguistic interactions likely to impact language acquisition and communicative competence. Thus, to better understand the variability in language outcomes following cochlear implantation, we are now conducting research on the input infants and toddlers receive from their parents before and after implantation, family environment, and the social/cognitive development of children with cochlear implants and how these factors influence language development (Bergeson et al, 2006; Peters and Beer, 2011; Bergeson, 2011; Kondaurova and Bergeson, 2011b; Frush Holt et al, 2012).

Parent-Child Interactions

One way in which caregivers can provide linguistic scaffolding for their infants and young children is to make use of acoustic attributes that attract their children's attention to speech, highlight important linguistic constructs, and simplify the speech input according to their children's cognitive skills. Caregivers around the world naturally provide such features when speaking to infants with normal hearing (relative to their speech to adults). This type of scaffolding might be particularly important for infants and young children with profound hearing loss with cochlear implants. Although pediatric implant users' speech perception skills are greatly improved over those of children with profound deafness who use hearing aids, the auditory input they receive is still degraded in terms of encoding of pitch and voice quality, two important features of infant-directed speech. It is possible that infants with cochlear implants will therefore pay less attention to speech during mother-child interactions, which could result in mothers' decreased use of infant-directed speech features.

We have been recording the interactions of normal-hearing mothers and their infants with profound hearing loss and varying experience with cochlear implants, as well as mothers' speech to other adults (baseline control). We have examined the suprasegmental, segmental, and linguistic features of the maternal speech across several studies. Rather than decrease their use of infant-directed speech registers when interacting with infants with cochlear implants, the mothers in our studies actually exaggerate suprasegmental speech features such as pitch height and segmental speech features such as vowel space in their speech to their infants as compared to speech to adults (Bergeson et al, 2006; Dilley and Bergeson, 2010; Bergeson, 2011; Kondaurova and Bergeson, 2011a, 2011b). Importantly, mothers tailor their use of these types of speech cues according to their infants' hearing experience in addition to chronological age (and presumably cognitive abilities). Our findings suggest that mothers are, in fact, using some level of linguistic scaffolding to help their young infants and children with cochlear implants develop spoken language skills.

Relationship between Input and Speech/ Language Outcomes

It is one thing to provide the exaggerated cues for infants to attract their attention to speech and to encourage them to attend to important linguistic features, but it is quite another to determine whether use of the infant-directed speech register is causally related to infants' development of spoken language skills. In fact, there is accumulating evidence that maternal infant-directed speech is linked to increased speech perception skills and cognitive abilities such as learning associations in infants with normal hearing (Kaplan et al, 2002; Liu et al, 2003). In addition to the quality of maternal input, the quantity of maternal speech input also seems to have strong and positive effects on children's later vocabulary and language skills (e.g., Hart and Risley, 1995; Hurtado et al, 2008).

To determine whether individual features of maternal speech input are related to the development of spoken language abilities in children with cochlear implants, we have carried out correlation analyses between mothers' speech features and the performance of infants with implants in the word-learning task described above. In a previous study of maternal speech to infants with cochlear implants, we found increased use of word and utterance repetition relative to adult-directed speech, similar to mothers' speech to infants with normal hearing (Bergeson, 2011). We predicted that factors such as the quantity of the input (i.e., number of words mothers used during the recorded interactions) and mothers' use of word and utterance repetition might influence infants' ability to learn novel words. In fact, only maternal utterance repetition was found to be significantly positively correlated with word-learning performance in infants at 12–18 mo postimplantation (Bergeson et al, 2011). In other words, infants with implants who were better word learners also had mothers who repeated various utterances (e.g., “Look at the fish!”) several times when interacting with them at earlier ages. It is possible that word repetition and input quantity are related to vocabulary acquisition rather than online word learning tasks.

Taken together, the studies of mother-infant interaction are consistent with the hypothesis that mothers and children must both be active participants in dynamic and reciprocal social exchanges to develop spoken language and vocabulary skills. Although young infants and children with hearing loss are at risk of degraded auditory access to social linguistic interactions, mothers in our studies seem to be responding in a sensitive manner, tailoring their speech registers to the infants' and children's developing auditory abilities. Thus, not only are these infants and children receiving the social-emotional benefits of infant-directed speech, known especially for its highly affective qualities, but they are also receiving the social-linguistic benefits of rich interactions with their caregivers.

Sociocognitive Development

Sociocognitive development encompasses both a child's ability to reason about behavior by considering the thoughts, beliefs, desires, and intentions of others, and a child's ability to understand and predict emotion (de Rosnay and Hughes, 2006). For children with normal hearing, opportunities from birth to listen to and participate in everyday conversations with siblings, parents, and friends provide a rich social linguistic environment that supports the child's developing understanding of mind and emotions. Congenitally deaf children, however, have both impoverished social and linguistic environments. It is therefore important to understand the effects that these atypical environments may have on children's sociocognitive development prior to and after receiving a cochlear implant. One objective of our research is to identify new methods and measures of sociocognitive understanding that will assess success and benefit with a cochlear implant for individual children and will help explain the individual differences in speech and language outcomes that are common in children with cochlear implants.

Peer Conversations

As a natural extension of our mother-infant research, we are also investigating the bidirectional relationship between sociocognitive development and language development in preschool-age children with cochlear implants to determine their impact on communicative and social competency. To do so, we compared qualitative and quantitative aspects of conversations of four deaf peer dyads to four normally hearing peer dyads during undirected play (Beer, 2008). We assessed the *connectedness* of alternating exchanges between peers, which provided a measure of the connectivity between the two children. These data give us valuable new information about how well the speakers are “tuned in” to one another, which requires general knowledge about the desires, beliefs, and intentions of one's interlocutor and linguistic ability to negotiate an activity using this knowledge. Connectedness of conversation is related to the development of social understanding in typically developing preschoolers because it is positively correlated with false belief performance and affective perspective taking. In addition, we calculated the amount of mental state talk children used, which is correlated with theory of mind performance in children with normal hearing. We found that deaf peer dyads engaged in fewer total exchanges and fewer verbalized connected exchanges on average than normally hearing dyads. In addition, both groups referred to mental states most often within connected turns although deaf peers had fewer total references to mental states than normally hearing peers. These preliminary findings suggest that deaf children who use spoken language may have difficulty establishing and maintaining perspectively rich connected conversation with a peer—a necessary precursor to more sophisticated linguistic interactions that require collaborative co-construction among conversational partners. These differences may explain some of the delays reported in more distal outcomes related to hearing loss such as theory of mind (ToM) and emotion understanding observed in deaf children (Peterson and Siegal, 1999; Peterson, 2004).

Social Understanding and Social Competence

Building on our preliminary investigation of communicative competence we have expanded our research tools to include measures of social cognition, executive function (EF), and social competence in children with hearing loss. Research with normal-hearing children provides evidence for the proposal that executive ability and theory of mind understanding are fundamentally linked in development and that executive control is a necessary but not sufficient contributor to children's understanding of theory of mind (Carlson et al, 2004). Recent research has found that children with hearing loss experience delays in social understanding (i.e., false belief performance, emotion understanding) and particular aspects of executive function (i.e., inhibition, behavior regulation, working memory) that may result from limited access to conversations due to a period of auditory deprivation and accompanied language delay (Peterson, 2004; Beer et al, 2012). We are administering several measures of EF (inhibition, working memory, shifting) and social understanding (i.e., diverse desires, knowledge access, false belief, hidden emotions, emotion identification with auditory-only and visual-only cues) in order to understand the relations between EF and social understanding in deaf children, which is complicated by their delay in language. Furthermore, we are assessing the implications that a delay in social understanding and EF may have on social competency (e.g., problem behaviors, internalizing, externalizing) and academic performance as measured by parent and teacher report. Clinically, it is important to understand which children are at high risk for developmental delays and what variables predict risk or resilience, as such knowledge guides intervention strategies. It is also important to understand whether intervention in one domain is likely to have cascading positive consequences in other domains.

NEUROCOGNITIVE PROCESSES

Recent theoretical work in speech perception and spoken language development suggests that both domain-general and domain-specific cognitive processes are recruited (Ullman, 2004; Behme and Deacon, 2008; Conway and Pisoni, 2008). What this means is that language processing may be at least partially subserved by the same underlying attentional and neurocognitive mechanisms that are involved in other cognitive domains. Moreover, early auditory deprivation may have a modality-specific effect on these processing operations (i.e., affecting the general cognitive processing of auditory input only) or have a modality-general effect on processing (i.e., affecting the cognitive processing of both auditory and visual inputs). We adopt a general working hypothesis that deaf children with cochlear implants may experience other neural, cognitive, and affective sequelae of early auditory deprivation combined with a delay in language prior to implantation (Pisoni et al, 2010). A child's performance with a cochlear implant may reflect variation in domain-general neurocognitive processes underlying speech and language processing. In order to test this hypothesis, we have expanded our traditional clinical battery of speech and language measures to include measures of executive-organizational-integrative (EOI) abilities: working memory, rapid efficient phonological processing, concentration and inhibition, and organization-integration—information processing measures that assess how well a child uses the limited and degraded sensory information obtained from the implant (Pisoni, 2000). Speech and language processing is highly dependent on these domain-general neurocognitive processes.

Modality-Specific Cognitive Processes

In the late 1990s, David Pisoni and others in the laboratory began measuring forward and backward auditory digit spans to obtain process measures of immediate memory capacity and auditory working memory (Pisoni and Geers, 2000). Compared to age-matched children with normal hearing, they found that children with cochlear implants had shorter forward and backward digit spans, slower verbal rehearsal speeds, and slower scanning and retrieval speeds of items from the lists of digits in short-term memory (Burkholder and Pisoni, 2003; Pisoni and Cleary, 2003; Pisoni and Cleary, 2004). In addition, better performance on these measures was positively correlated with measures of spoken word recognition. Together these findings suggest fundamental limitations in processing capacity of working memory, less robust perceptual encoding, and slower active maintenance and retrieval of phonological representations in working memory. These early studies provided some of the first evidence of disturbances in basic elementary neurocognitive processes related to language processing in children with cochlear implants.

More recent work has measured change in immediate memory capacity and working memory (as measured by digit span) and verbal rehearsal speed—two core elementary neurocognitive measures of information processing required of all speech and language outcome measures—after long-term cochlear implant use, and the relations between these changes and children's performance on several traditional speech and language assessments (Pisoni et al, 2011). In a sample of 112 cochlear implant users tested at age 8–9 and then again 10 yr later, there was a greater tendency for improvement in digits forward (immediate memory capacity) than in digits backward (working memory and executive control) for many but not all children. Scores on digits forward were also strongly associated with performance on speech and language measures in high school, whereas digits backward scores were correlated only with higher-order global measures of language such as spoken language comprehension and reading. In contrast, verbal rehearsal speed increased for almost every child between the elementary school evaluation and the high school evaluation, and scores at both times were strongly intercorrelated. Furthermore, verbal rehearsal speed at elementary school was strongly correlated with several speech and language measures at high school. The objective of this research on working memory capacity is to identify the core neurocognitive processes that underlie speech and language development after cochlear implantation, not only to explain the great amount of variability in outcomes but to identify children who may be at high risk for poor outcomes at an early age, and to design and implement novel and individualized interventions and treatment throughout the school years for children with cochlear implants (Kronenberger et al, 2011).

Modality-General Cognitive Processes

The finding that language skills were correlated with domain-general neurocognitive processes in the auditory modality (i.e., auditory memory) led to investigating the hypothesis that early auditory deprivation leads to modality-general disturbances. To investigate the possibility of a modality-general impact of auditory deprivation in children with cochlear implants and to ultimately identify pre-implant predictors of outcome that do not require hearing and audition, our laboratory has investigated the development of skills outside the auditory domain such sequence memory and learning, visual attention, visual-motor integration, and motor skills.

Sequence Memory and Learning

Using a modified version of the popular Simon sequence game, children were asked to reproduce sequences of spoken color names (auditory), colored lights (visual), and color names combined with colored lights (auditory + visual) by touching the corresponding colored panels on the Simon game. Children with cochlear implants had shorter sequence spans in all three conditions compared to children with normal hearing, and one-third failed to show any sequence repetition or learning effects at all (Cleary and Pisoni, 2001; Pisoni and Cleary, 2004). In addition, children with cochlear implants displayed a reversal of the "modality effect," showing longer memory spans for visual sequences than auditory sequences. The results of these sequence memory and learning studies suggest that a period of auditory deprivation affects both the neural processes involved in learning and memory as well as the neurocognitive processes used to encode and maintain sensory information *in both auditory and visual domains*.

Further evidence of modality- and domain-general effects of auditory deprivation comes from a series of studies in our laboratory using nonauditory sequencing abilities (motor and visual) in deaf children with cochlear implants. When asked to demonstrate learning of a visual sequence or to reproduce a series of finger taps, children with cochlear implants performed more poorly than a control group of children with normal hearing of the same age, suggesting that auditory deprivation may not only affect hearing and speech perception but also cognitive abilities related to perceiving and producing sequential information (Conway, Karpicke, et al, 2011; Conway, Pisoni, et al, 2011). A current investigation is exploring the time course for the effects of auditory deprivation on visual sequence learning by investigating visual sequence learning in deaf infants before and after cochlear implantation.

The findings on sequence memory learning have important implications for language development after cochlear implantation. A child with a cochlear implant may be able to perceive auditory input provided by the implant but may have difficulty encoding, processing, and learning aspects of language that rely on sequential regularities such as phonological and grammatical sequencing. Indeed, several studies in our laboratory have discovered close links between visual sequence learning and language processing in normal-hearing adults (Conway et al, 2007), children with cochlear implants (Conway, Pisoni, et al, 2011), and normal-hearing infants (Shafto et al, 2012).

Visual Attention

Using a continuous performance task during which children must sustain visual attention and respond only when they detect a target stimulus, we also found that some children with cochlear implants showed atypical visual attention skills compared to normally hearing children (Horn et al, 2005). However, visual attention began to improve after 12 mo of implant use due to increased perceptual sensitivity for distinguishing targets from the nontargets.

Visual-Motor Integration

In another study, we also examined visual-motor integration in children with cochlear implants to assess the hypothesis that this perceptual motor skill would contribute to the development of speech and language after implantation (Horn, Fagan, et al, 2007). Results indicated that implant users with at least 2 yr of implant experience were delayed compared to the published norms on both a visual-motor task that required them to copy increasingly complex two-dimensional figures and a timed maze tracing task. In addition, scores on the copying task were correlated with speech perception and working memory.

Motor Skills

Finally, we examined motor skills of prelingually deaf children using the Vineland Adaptive Behavior Scales (Horn et al, 2006). Findings indicated divergence in the development of fine and gross motor skills: Older children showed more advanced gross motor skills but less advanced fine motor skills pre-implant than younger children. In addition, children with more advanced pre-implant fine motor skills showed more progress in receptive and expressive language postimplant than children with less advanced fine motor skills.

This line of research strongly supports the hypothesis that early auditory experience can impact the development of cognitive processes and motor skills that are not specific to audition or spoken language. Furthermore, the significant relationships detected among visual motor integration, fine-motor skills, and tests of language and speech perception suggest that these areas may be coordinated in development and/or may share underlying cortical processing resources (Luria, 1973). Overall these findings have provided us with new avenues for understanding individual differences in performance with a cochlear implant as well as providing novel pre-implant predictors of performance.

Executive Function and Cognitive Control

In addition to performance-based measures of executive function such as digit span, spatial span, Stroop, and planning, we have also used a parent report measure of executive function called the Behavior Rating Inventory of Executive Function (BRIEF) and BRIEF-P (Preschool version) that can be used from age 2 to 18. The BRIEF is used to assess problem behaviors related to executive function as exhibited in everyday life. T-scores indicate elevated (T-score ≥60) and clinically elevated (T-score ≥65) domains of executive function. Data from our laboratory indicate that some children with cochlear implants are delayed in some, but not all, areas of executive function. We found that children with cochlear implants have significantly elevated scores on working memory, inhibition, and behavior regulation compared to peers with normal hearing. In addition, we found that children who score above the median on the BRIEF have significantly poorer performance hearing sentences in noise (but not quiet) and on measures of general language ability (but not vocabulary) compared to children who score below the median. This research suggests that problems with working memory may selectively impact tasks with high cognitive load. In addition, a period of auditory deprivation and language delay experienced by children with CIs may impact their ability to control and monitor attention and regulate their behavior during tasks that require high cognitive resources and focused attention (Beer et al, 2009, 2011).

Working Memory Training

Recently, our laboratory completed a feasibility study using the Cogmed Working Memory Training program, a computer-based program designed to improve working memory and executive functioning (Kronenberger et al, 2011). Deaf children with cochlear implants who had average to below-average working memory participated in 25 training sessions over a period of 5 wk. Results demonstrated acceptable feasibility based on parent reports and significant improvement on measures of verbal and nonverbal working memory capacity, and real world working memory. Novel process-based intervention is a promising area for future research in our laboratory for selectively modifying the underlying neurocognitive processes affected by deafness that underlie speech and spoken language outcomes in deaf children with cochlear implants.

CHILDREN WITH ADDITIONAL DISABILITIES

Children with deafness and additional disabilities (AD) constitute a significant proportion of the deaf pediatric population and pose unique challenges to the cochlear implant team. A third or more of deaf children in the United States are believed to have at least one additional disability (Holden-Pitt and Diaz, 1998). Evaluation and rehabilitation after cochlear implantation in children with AD is complex due to the heterogeneity of this group of children, the lack of appropriate assessment tools, and different expectations of implantation by families and clinicians caring for multiply handicapped deaf children.

Recent findings suggest that although some children with AD do make progress in auditory skills development and language, they do so at a much slower rate than children without AD. Moreover, many children with significant developmental delays may make no progress on conventional speech and language assessments. With regard to pre-implant predictors of benefit in children with AD, both nonverbal IQ and the degree of developmental delay have been successful in predicting speech intelligibility, language, and auditory skills, more so than age at implantation or aided thresholds (Waltzman et al, 2000; Edwards, 2007; Meinzen-Derr et al, 2011).

At our cochlear implant center, AD is not contraindicative of cochlear implantation; as a result, we have had the opportunity to follow a large cohort of children with AD for the past 5 yr. In a study comparing deaf children with cochlear implants who have a mild cognitive delay to children with cochlear implants who are otherwise typically developing, Holt and Kirk (2005) found that both groups of children made significant progress in speech and language after 1 yr of device use but that the group of children with a cognitive delay made significantly less progress than the group without a cognitive delay, particularly in areas that require higher-level linguistic skills such as expressive and receptive language and sentence recognition. In a more recent study of 31 children presenting with a variety of additional handicapping conditions such as cerebral palsy, CHARGE syndrome, blindness, and various additional syndromic conditions, we found that after 12 mo of implant use, children's functional auditory skills and receptive language increased significantly and that children made 1 yr of progress in 1 yr's time in socialization and daily living skills (Beer et al, 2010).

Finally, in an effort to predict progress after implantation in children with AD we are developing a new scale, the Pediatric Functional Assessment Scale (PFAS), which can be used across all handicapping conditions to assess the impact of a child's impairment on five domains of adaptive functioning: self-care, motor, speech-language, socio-emotional, and cognitive domains. Ratings of severe (1), moderate (2), and mild (3) impact on functioning in each of these domains are obtained by using pre-implant data from parent questionnaires, developmental assessments, and reports from a speech-language pathologist. Scores on the PFAS range from 5 to 15 with lower scores indicating a more severe impact on adaptive functioning. We applied the PFAS retrospectively to a cohort of 30 cochlear implant recipients with one or more additional complicating diagnoses who are being followed at our center (Harris et al, 2010). The mean PFAS score for this cohort was 9 (SD = ±3). A median split analysis showed high-functional impact (low PFAS score) to be associated with poorer performance as indexed by the functional auditory skills at baseline assessment and both 6 and 12 mo postimplant. Moreover, performance on the Adaptive Behavior Composite Score of the Vineland-II was correlated with PFAS at baseline, pre-implant, and 6 mo postimplant, which indicates that the PFAS is tapping into adaptive functioning behaviors.

Although these initial findings are encouraging, the data are limited in a number of ways. First, several children were unable to complete the speech perception and language tests that require higher level responses. Second, the tests that assess lower level auditory skills were not sensitive enough to detect the incremental progress over time that is typical of these children. Finally, although clinicians, parents, and speech-language therapists all report anecdotally that access to sound provided by the implant provides improvements in quality of life, increased environmental sound awareness, and increased connectedness to family members, we do not have sufficient data to support these conclusions because these domains were not assessed. We are presently expanding our assessment battery for children with AD to include these measures.

SUMMARY

The field of cochlear implantation is still very young, and the research questions have evolved rapidly. In just three decades scientists have gone from wondering if cochlear implants would provide any significant gains in access to sound, to wondering how well users could hear and perceive speech with them, to studying the effects of early auditory deprivation and subsequent implantation on linguistic, social, and neurocognitive processes underlying spoken language development. In this article we have summarized some of the recent work in our research laboratory at Indiana University that reflects this evolution and described our rationale for pursuing these particular lines of research. Through this work we are making steady progress toward understanding the complex interactions between the cochlear-implanted ear and the developing brain in infants and children.

NOTES

1. The Food and Drug Administration (FDA) approves cochlear implantation in children 12 mo old and older. However, several centers including ours provide cochlear implants at earlier ages "off-label" when there is strong evidence that an infant is profoundly deaf and not progressing in his or her speech and hearing development with hearing aids.
2. Conditioning an infant to detect a change in sound is much more difficult than conditioning an infant to detect the presence of a sound, as is the case with visual reinforcement audiometry. Attempts were made to implement a conditioned head turn procedure for speech discrimination. Our experience was that many infants with cochlear implants did not learn the contingency between a change in sound and the reinforcer. The methodology has been reported to be more successful with infants and toddlers with milder degrees of hearing loss (Eisenberg et al, 2004).
3. Lexical stress refers to the distribution of stressed and unstressed syllables in a word. For example, words like *doctor* and *candle* have word-initial stress whereas words like *guitar* and *surprise* have word-final stress.

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