Published in final edited form as:

Ear Hear. 2008 August ; 29(4): 492–511. doi:10.1097/AUD.0b013e31816c409f.

An Exploratory Look at Pediatric Cochlear Implantation: Is Earliest Always Best?

Rachael Frush Holt[1] and **Mario A. Svirsky**[2]

[1]Department of Speech and Hearing Sciences, Indiana University, Bloomington, Indiana

[2]Department of Otolaryngology, New York University School of Medicine, New York, New York

Abstract

Objectives—Since the advent of cochlear implants, age at implantation has declined as investigators report greater benefit the younger a child is implanted. Infants younger than 12 mos currently are excluded from Food and Drug Administration clinical trials, but have been implanted with Food and Drug Administration-approved devices. With a chance that an infant without profound hearing loss could be implanted because of the limitations of the diagnostic measures used with this population and the potential for additional anesthetic risks to infants younger than 1-yr-old, it is prudent to evaluate benefit in the youngest cochlear implant recipients. The goals of this research were to investigate whether significant gains are made by children implanted before 1-yr-old relative to those implanted at later ages, while controlling for potential covariates, and whether there is behavioral evidence for sensitive periods in spoken language development. It was expected that children implanted before age 1 yr would have more advanced spoken language skills than children implanted at later ages; there would be a negative relationship between age at implantation and rate of spoken language development, allowing for an examination of the effects of sensitive periods in spoken language development; and these trends would remain despite accounting for participant characteristics and experiences that might influence spoken language outcomes.

Design—Ninety-six children with congenital profound sensorineural hearing loss bilaterally and no additional identified disabilities who were implanted before the age of 4 yrs were stratified into four groups based on age at implantation. Children's spoken language development was followed for at least 2 yrs after device activation. Spoken language scores and rate of development were evaluated along with four covariates (unaided pure-tone average, communication mode, gender, and estimated family income) as a function of age at implantation.

Results—In general, the developmental trajectories of children implanted earlier were significantly better than those of children implanted later. However, the advantage of implanting children before 1-yr old versus waiting until the child was between 1 and 2 yrs was small and only was evident in receptive language development, not expressive language or word recognition development. Age at implantation did not significantly influence the rate of the word recognition development, but did influence the rate of both receptive and expressive language acquisition:

Address for correspondence: Mario A. Svirsky, PhD, Department of Otolaryngology, New York University School of Medicine, 550 First Avenue, NBV-5E5, New York, NY 10016. Mario.Svirsky@med.nyu.edu.

children implanted earlier in life had faster rates of spoken language acquisition than children implanted later in life.

Conclusions—Although in general earlier cochlear implantation led to better outcomes, there were few differences in outcome between the small sample of six children implanted before 12 mos of age and those implanted at 13 to 24 mos. Significant performance differences remained among the other age groups despite accounting for potential confounds. Further, oral language development progressed faster in children implanted earlier rather than later in of life (up to age 4 yrs), whereas the rate of open-set speech recognition development was similar. Together, the results suggest that there is a sensitive period for spoken language during the first 4 yrs of life, but not necessarily for word recognition development during the same period.

Introduction

Pediatric cochlear implantation criteria have changed dramatically since 1980 when the first individual younger than 18 yrs received a cochlear implant (CI) (Eisenberg & House, 1982). In 1990, the US Food and Drug Administration (FDA) first approved cochlear implantation in children. At that time, criteria for implantation included bilateral profound deafness, age 2 yrs or older, and demonstration of little or no benefit from amplification (Staller, et al., 1991). Since that time, candidacy criteria have broadened to include children as young as 1 yr with profound hearing loss and some children have been implanted even before their first birthday.

Accumulating evidence suggests that better outcomes are achieved by congenitally deaf children who receive CIs earlier rather than later in life. Fryauf-Bertschy et al. (1997) reported that children who receive CIs between 2 and 5 yrs of age tend to have better open-set speech perception than children who receive one after 5 yrs of age. Tyler et al. (1997) reported higher speech recognition scores in children implanted before 4 yrs of age than those implanted after 4 yrs. Nikolopoulos et al. (1999) followed a group of 126 congenitally deaf CI users up to 4 yrs postimplantation. They reported a significant negative correlation between age at implantation and performance on closed- and open-set measures of speech perception, suggesting that earlier implantation results in better speech perception outcomes. Finally, in examining speech and language development of children implanted in the second, third, or fourth year of life, Svirsky et al. (2004) reported that children implanted before age 2 yrs had significant speech perception and language advantages over children implanted after 2 yrs of age. Although these data support an “earlier is better” approach, they also beg the question, How early is it appropriate to perform cochlear implantation?

Children younger than 12 mos currently are excluded from FDA clinical trials, but have been implanted with FDA-approved devices at centers around the United States, Europe, and Australia. Few investigations have been performed on this population of implant recipients. In a recent report, Dettman et al. (2007) compared rates of receptive and expressive language development in infants implanted between 6 and 12 mos of age (N = 11) and children implanted between 13 and 24 mos (N = 36) who completed all six subscales of the Rossetti Infant-Toddler Language Scale (Rossetti, 1990). Even when children with cognitive delays were excluded from the analysis (which included six children from the older age-at-

implantation group), infants implanted during the first year of life had significantly faster rates of receptive (1.12) and expressive (1.01) language development than the children implanted in the second year of life (receptive: 0.78; expressive: 0.73). Using a clinician-administered parental report of auditory skill development (the Infant-Toddler Meaningful Auditory Integration Scale; Zimmerman-Phillips & Osberger, 1997), Waltzman and Roland (2005) reported gains on the magnitude of 30 points of a maximum of 40 points in infants implanted before 12 mos of age who had used their devices for 6 mos. Although the infants were not compared with children implanted at older ages, large gains were reported by parents of these very early implanted infants.

At least two issues are particularly relevant when considering the risks of cochlear implantation in infants younger than 1 yr. First, is there a chance that an infant without profound hearing loss could be implanted because of the limitations of the diagnostic measures used with this population? Second, are there additional anesthetic risks to infants younger than 1 yr relative to older children?

Identifying Hearing Loss in Infants: Sensitivity and Specificity Issues

Behavioral audiometric testing is the gold standard for measuring hearing sensitivity (e.g., Widen, et al., 2000). Behavioral testing with infants is conducted clinically using visual reinforcement audiometry (VRA). The procedure reinforces head-turn responses in infants in response to a signal (narrow-band noise, pure tones, or speech). The procedure has been used reliably with typically developing children as young as 5 to 6 mos (Moore, et al., 1977). VRA is not appropriate for infants younger than about 5.5 mos because they do not make directed head turns toward sound sources (Clifton, et al., 1981). Some infants are delayed in their development and are unable to complete VRA at 6 mos of age. For example, prematurely born infants tend to perform better on VRA if testing is conducted when they are 6 to 8 mos corrected age (Moore, et al., 1992; Widen, 1990) and some infants with neurodevelopmental deficits are unable to complete VRA altogether (e.g., Norton, et al., 2000). Therefore, there are infants in our population of interest who will be unable to complete VRA. In this situation, audiologists must rely on objective measures of auditory function.

Three objective measures of auditory function are typically used in clinical settings: evoked otoacoustic emissions testing (OAE), auditory brainstem response testing (ABR), and auditory steady-state response testing (ASSR). Each of these measures assesses specific portions of the auditory system rather than the auditory pathway as a whole. Further, none of these measures have perfect sensitivity and specificity. In other words, each measure will, at times, fail to identify an individual with a hearing loss when in fact she/he has one and will incorrectly indicate a hearing loss in an individual who has normal hearing sensitivity. For our purposes, the issue is less one of identifying impaired versus normal hearing, but rather one of identifying profound hearing loss versus not profound hearing loss (normal hearing or hearing loss that is mild to severe in degree). Specificity issues are of primary concern in this line of research, because we are interested in assessing the risk of implanting infants lacking bilateral profound sensorineural hearing loss.

OAEs, whether transient evoked (TEOAE) or distortion product (DPOAEs), are believed to measure the nonlinear response properties of the cochlea, specifically the outer hair cell system. The underlying assumption regarding the usefulness of OAEs for identifying hearing loss is that many losses that are mild to moderately severe in degree involve damage to the outer hair cells. Gorga et al. (1997, 1999) measured DPOAEs in 1267 ears of 806 participants who ranged in age from 1.3 to 96.5 yrs and compared the results with each listener's behavioral thresholds. In the earlier investigation, Gorga et al. (1997) examined hearing level as a function DPOAE/noise ratio (the difference between the amplitude of the emission and the level of the noise at a given frequency) for each participant at interoctave frequencies between 0.75 and 8 kHz. DPOAE measurements were better at separating impaired ears from normal ones at mid and high frequencies than low frequencies. However, there was a large amount of overlap between these populations: depending on frequency, participants with profound hearing losses (>90 dBHL) had DPOAE/noise ratios between approximately −10 and +10 dB, as did individuals with moderate to severe losses; listeners with mild hearing losses had DPOAE/noise ratios between approximately −15 and +30 dB and those with normal hearing had ratios ranging from approximately 0 to +40 dB. If a strict criterion of +9 dB DPOAE/noise ratio were used, at least one listener at 2 kHz, five listeners at 3 kHz, seven listeners at 4 kHz, and three listeners at 6 kHz with profound hearing loss would be missed (identified as having normal hearing) and many more listeners with normal hearing would be mislabeled as having hearing loss, particularly at 2 kHz. In a follow-up investigation, Gorga et al. (1999) reported that using a multivariate analysis (using combinations of multiple frequencies) resulted in better sensitivity and specificity than using the more typical univariate analysis (using a single frequency at a time), particularly for listeners with severe to profound hearing loss. These data suggest that impaired and normal-hearing populations' OAE results overlap and that although sensitivity and specificity can be improved with different analysis tools, OAEs are not perfect for identifying hearing loss, even those that are profound in degree.

Another large-scale study on the predictive value of objective measures specifically assessed infants, our population of interest. Norton et al. (2000) compared the performance of TEOAEs, DPOAEs, and ABRs assessed neonatally for predicting hearing status at 8 to 12 mos corrected age. Infants with reliable VRA results (95.6% of those who returned for follow-up VRA testing) were included in the sample (N = 2995). Minimum response levels at 1, 2, and 4 kHz and speech awareness thresholds (SATs) from VRA testing were used as gold standards. TEOAEs and DPOAEs (using moderate-level stimuli) were nearly equivalent in predicting auditory status. In contrast, ABRs resulted in similar performance as OAEs for predicting auditory status at 2 kHz, 4 kHz, and for SAT, but ABRs had more predictive value than OAEs for thresholds at 1 kHz. In other words, neonatal OAE results (of either type) were related to hearing sensitivity at 2 and 4 kHz and to SATs at 8 to 12 mos corrected age, whereas neonatal ABR results were related to hearing sensitivity at all frequencies tests (1, 2, and 4 kHz) and to SATs at 8 to 12 mos corrected age. ABRs involve recording electrophysiological responses to brief stimuli, typically clicks or tone bursts. The responses are small in amplitude and thus are averaged over many sweeps. Recording a response relies on neural synchrony to some degree and click-evoked ABRs typically reflect hearing sensitivity between 2 and 4 kHz better than at lower or higher frequencies. No conditions for

any objective measure resulted in false positive rates of zero, meaning that some normal ears did not produce measurable TEOAE, DPOAE, and/or ABR responses (Norton et al.).

ASSR is a relatively new clinical tool in which electrophysiological recordings are made from individuals in response to pure-tone stimuli that are amplitude-, frequency-, or both amplitude- and frequency-modulated. ASSR is intended to address some of the shortcomings of ABR. For example, frequency-specific ABR recordings to stimuli such as tone bursts alone or tone bursts combined with noise maskers are less robust than click-evoked responses in quiet, making it challenging to obtain frequency-specific information from ABR recordings. Also, it is believed that ASSRs can be recorded over a wider dynamic range than ABRs, because ASSR stimuli can be presented at effectively higher levels than those used to elicit ABRs because of the use of longer sinusoidal stimuli in ASSR versus the brief clicks used in ABR testing. Finally, ASSR software uses statistical criteria for determining whether a response exists, and thus does not rely on the expertise of the observer in identifying waveforms embedded in the noise of ongoing brain and muscle activity.

Rance et al. (1998) compared hearing threshold estimates from click-evoked ABR, ASSR, and behavioral testing in 108 infants and young children ranging in age from 1 to 49 mos at the time of ASSR testing. No participant had repeatable waveforms to ABR testing at the limits of the equipment, 100 dBnHL. As expected, behavioral hearing thresholds in the low frequencies correlated poorly with the ABR results: 97% and 78% of ears had residual hearing based on behavioral testing at 0.25 and 0.5 kHz, respectively, despite no repeatable ABR at maximum levels. This proportion decreased to 68%, 43%, and 27% at 1, 2, and 4 kHz, respectively. In contrast, the proportion of ears with residual hearing and absent ASSRs was smaller: 87%, 12%, 21%, 18%, and 8% at 0.25, 0.5, 1, 2, and 4 kHz, respectively. Therefore, ASSR testing had better specificity than ABR, particularly at 0.5 kHz through 4 kHz. Even though many ears were found to have residual hearing despite absent ABRs, the amount of residual hearing was typically limited. Between 85% and 90% of the ears with residual hearing had losses of 90 dBHL or greater (profound hearing loss) between 1 and 4 kHz. Still, this means that when using ABR, 10% to 15% of the sample would be classified as having a profound hearing loss when in fact it was moderate to severe in degree. In contrast, in this sample all the ears with absent ASSRs had behavioral thresholds greater than 90 dBHL. Therefore, for our purposes, an absent ASSR resulted in perfect sensitivity in identifying a profound hearing loss in this sample. Further, the ASSR threshold need not even be absent to indicate a profound hearing loss. An ASSR criterion threshold of 100 dBHL or greater at 1, 2, and 4 kHz resulted in no false alarms for profound hearing loss. However, this criterion would result in missing six children with profound losses at 1 kHz and two at 2 kHz.

In a smaller study, Luts et al. (2004) compared click-evoked ABR thresholds, ASSR thresholds, and behavioral thresholds in 10 infants aged 3 to 14 mos at the time of the first objective test. Both ears were not tested in every condition for every infant primarily because of time constraints. Despite this, ABR threshold estimates would have falsely identified 5 of 11 ears with both ABR and behavioral thresholds as having profound hearing loss at 2 kHz when in fact the losses ranged from mild (30 dBHL) to severe (85 dBHL). In those five ears,

only one ASSR threshold would have resulted in the same error. Therefore, ASSR responses resulted in fewer false alarms than ABR responses.

These data suggest that ASSR threshold estimates are more reflective of true auditory sensitivity in the severe to profound hearing loss range than ABR threshold estimates, at least at 1, 2, and 4 kHz. However, at least one study demonstrated that ASSR recordings at high stimulus levels (100 dBHL and above) might be artifact. Gorga et al. (2004) found reliable ASSR recordings in 10 adults with profound hearing loss who had no detection of the ASSR stimuli. These data suggest that ASSR recordings at high stimulus levels might not be accurate predictors of auditory status. However, for our purposes of identifying infants with profound hearing loss, the ASSR seems to be sufficiently sensitive and specific.

Taken together, these data on objective measures of auditory function suggest that combining results from evoked OAEs, ASSR, and ABR, will provide the most accurate picture of an infant's auditory status, even though each measure is capable of both failing to identify hearing loss in individuals with hearing impairment and identifying hearing loss in individuals who have no such loss. New data from John et al. (2004) suggest that ASSR recordings in infants older than 1 mo are more accurate than those recorded in neonates. Therefore, it might be wise to postpone ASSR testing until the infant is older than 1 mo. Unlike ABR, ASSR recordings can be obtained from infants and children with auditory neuropathy/dysynchrony. However, the thresholds obtained have not been shown to correlate well with hearing sensitivity in infants and children suspected of having auditory neuropathy/dysynchrony (Luts, et al., 2004; Rance & Briggs, 2002; Rance, et al., 1999). Therefore, although ASSR seems to be somewhat more sensitive and specific to profound degrees of hearing loss than ABR, ABR should still be a part of the testing protocol with infants to identify infants with auditory neuropathy/dysynchrony. In a similar vein, OAE testing should still be part of the test battery to identify children with auditory neuropathy/dysynchrony.

We do not have perfect measures for evaluating auditory status in infants. There is always a small risk of labeling a child with profound bilateral hearing loss who in fact has more hearing than the test measures suggest. This leaves open the possibility for implanting an infant without profound bilateral hearing loss, particularly one who cannot complete behavioral testing. Considering this small risk, it is especially important to evaluate whether there are great speech and language advantages for infants implanted below the age of 1 yr when many clinicians heavily rely on these physiological measures. If there were no advantage or the advantages were slight, it might be prudent to postpone cochlear implantation until the infant is old enough to reliably complete VRA testing.

Anesthetic Risk in Infancy

Potential reasons for alarm—Although the incidence of anesthesia-related complications in children and infants has steadily declined in the last 50 yr (Morray, 2002), the incidence of mortality and morbidity in infants younger than 1 yr is still significantly higher than for children older than 1 yr and adults (Cohen, et al., 1990; Keenan, et al., 1994; Morray, et al., 2000; Olsson & Hallen, 1988; Rackow, et al., 1961; Tay, et al., 2001; Tiret, et al., 1988). Most investigators have estimated the rate of life-threatening adverse events to be

three to four times higher in infants younger than 1 yr than in children older than 1 yr (Olsson & Hallen, 1988; Rackow, et al., 1961; Tay, et al., 2001), with some estimates as high as an eightfold increase in risk (Keenan et al., 1994; Tiret et al., 1988). Keenan et al. found that the incidence of bradycardia during noncardiac surgery was higher in infants younger than 1 yr (127 per 10,000) than in children in the second, third, or fourth years of life (98, 65, and 16 per 10,000, respectively). Also of concern is that in one study, only 4.1% of anesthesia-related intraoperative incidents recorded electronically by an automated anesthesia records and information management system were voluntarily reported by the administering an-esthesiologist (Sanborn, et al., 1996). This suggests that the vast majority of incidents go unreported and thus are not typically included in studies examining the incidence of anesthesia-associated critical events. As far as we know, there is no known difference in the rate of reporting based on the age of a patient. Therefore, even with significant underreporting of intraoperative anesthesia-related incidents, there seems to be significantly greater anesthetic risk to infants undergoing surgery in the first year of life than children in the second and later years of life. If age were the only factor related to anesthetic risk, these findings would be cause for great alarm. However, recent investigations have pointed to a number of other factors that influence anesthesia-related morbidity and mortality.

Potential reasons for tempered alarm: Why the outlook may not be so bleak for infant cochlear implant candidates—A number of other surgery-and patient-related factors (beyond patient age) influence the risk of anesthesia, including American Society of Anesthesiology (ASA) physical status; whether the surgery is emergency or scheduled; and whether a pediatric anesthesiologist is present. The ASA physical status indexes a given patient's level of risk based on her/his medical condition before surgery, with larger numbers indicating increased risk: level 1 refers to healthy patients; level 2 refers to patients with a mild systemic disease (e.g., mild diabetes); level 3 refers to patients with severe systemic disease (e.g., frequent angina); level 4 refers to patients with severe systemic disease with acute, unstable symptoms (e.g., congestive heart failure); and level 5 refers to patients who are approaching death and who are not expected to survive without the operation. Most children undergoing CI surgery are typically considered ASA physical status 1 or 2. The anesthetic risk for individuals in either of these categories is significantly lower than for individuals classified as ASA 3, 4, or 5 (Keenan, et al., 1994; Olsson & Hallen, 1988; Tay, et al., 2001; Tiret, et al., 1988). In fact, Morray et al. (2000) reported that when ASA physical status was controlled for, patient's age was no longer the sole predictor of anesthesia-related mortality.

Keenan and Boyan (1985), Morray (2002), and Tiret et al. (1988) reported that anesthetic risk was higher in patients undergoing emergency surgery than scheduled surgery. However, at least one study failed to find this (Tay, et al., 2001). Cochlear implantation is considered a scheduled surgery. Therefore, the anesthetic risk is at least equal to or lower than that for surgeries that are performed on an emergency basis.

The proficiency of the individual anesthesiologist has been shown to significantly influence the risk of anesthesia in infants (Keenan, et al., 1991, 1994). Keenan et al. (1991) reported that the incidence of anesthesia-induced cardiac arrest in infants younger than 1 yr over a 7-

yr period was 19.7 per 10,000 anesthetic procedures when a nonpediatric anesthesiologist was present, whereas no incidences were reported when a pediatric anesthesiologist was present. Keenan and Boyan (1985) speculated that one explanation for the higher incidence of complications in emergency procedures is that a pediatric anesthesiologist may not be available when an infant requires emergency surgery.

A final reason for tempered concern for this population is that at least one study found that the majority of intraoperative anesthesia-related incidents in infants younger than 1 yr occurred in infants younger than 1 mo. Including serious complications (e.g., death and cardiac arrest) and less serious complications (e.g., vomiting) in their analysis, Cohen et al. (1990) found that infants between 1 and 12 mos of age had the same rate of complications as older children aged 1 to 5 yrs (7%), whereas those aged less than 1 mo had a 15% rate of complications. However, not all studies found this trend in their samples of infants younger than 1 yr (e.g., Keenan, et al., 1994). Our population of interest consists of infants aged 6 mos or older at the time of cochlear implantation. Therefore, if the bulk of the risk for children younger than 1 yr is due to children younger than 1 mo, as Cohen et al. suggest, then the concern for our population decreases.

Despite the observation that “the exact mortality rate due to anesthesia … is unknown, and probably unknowable” (Keenan & Boyan, 1985, p. 2373), the results from these studies suggest that the alarm raised by investigators who found increased anesthetic risk in infants younger than 1 yr relative to older children should be tempered by other risk factors that covary with age; particularly, ASA physical status and whether anesthesia is administered by a pediatric anesthesiologist. Keenan and Boyan (1985) examined 163,240 anesthetic administrations and found that 27 cardiac arrests were judged to be primarily due to anesthesia. The incidence of cardiac arrest in children younger than 12 yrs was three times higher than in adults. However, none of the 27 cardiac arrests occurred in children younger than 1 yr who were classified as ASA physical status 1 or 2. Taken together, these studies have failed to find evidence of a difference in anesthetic risk based on age when the child is relatively healthy (ASA physical status 1 or 2) and anesthesia is administered by a pediatric anesthesiologist. Still, we are not aware of a large-scale study that has examined the population of interest to CI researchers and clinicians: infants classified as ASA physical status 1 or 2 who undergo scheduled surgery. Such a study would allow us to assess better the actual anesthetic risk to infants undergoing cochlear implantation. Without risk data from our population of interest, the administration of anesthesia to a healthy infant by a pediatric anesthesiologist is generally accepted in the field if there is clear evidence that the benefits of cochlear implantation at 6 mos of age are significantly greater than those at 12 mos of age. Currently, the actual level of evidence for benefit in this very young infant population is low. This investigation was undertaken to examine whether there is an advantage for very early cochlear implantation; that is, cochlear implantation in infants younger than 1 yr. If there were no advantage or the advantage were slight, it might be prudent to postpone cochlear implantation until: (1) the child is old enough to reliably complete VRA testing to avoid the slight risk of implanting an infant without profound hearing loss; and (2) the child has outgrown the age at which some investigators have found an increased risk of complications associated with anesthesia.

The present study examines longitudinal data from a small group of children who received CIs in the first year of life and who have no additional disabilities. Combining what we have learned from congenitally deaf children who were implanted at later ages with these new data will result in at least two potentially important outcomes. First, from a clinical perspective we will have preliminary outcomes evidence from which to base decisions regarding very early implantation while considering the accompanying risks. Second, from a scientific perspective we will be able to evaluate converging evidence that is relevant for the investigation of sensitive periods in language development. The goals of this research are to (1) explore whether significant gains are made by children implanted before 1 yr of age relative to those implanted at later ages, while controlling for potential covariates such as better-ear pure-tone average (PTA), estimated family income, gender, and communication mode (CM); and (2) investigate whether there is behavioral evidence for sensitive periods in spoken language development. It is anticipated that this investigation will contribute to the emerging data on this new population of implant recipients on whom little is known, while also controlling for potential covariates (at least one of which is a relatively new area of investigation in this population of children-estimated family income) that might contribute to the enormous variability in outcomes in children with CIs.

We expect that at a given postimplantation age, children implanted before age 1 yr will have more advanced spoken language skills than children implanted at later ages. Further, we predict that there will be a negative relationship between age at implantation and rate of speech and language development and that this will allow us to observe effects of sensitive periods in spoken language development. Finally, we expect these trends will remain despite accounting for participant characteristics and experiences that might influence spoken language outcomes.

Materials and Methods

Participants

The children included in this investigation constituted a subset of the population of pediatric CI recipients followed longitudinally at Indiana University Medical Center. Inclusion criteria included congenital profound sensorineural hearing loss bilaterally, no additional identified disability, implanted with a current device before age 4 yrs, use of SPEAK or Continuous Interleaved Sampling stimulation strategies since initial fitting, tested on at least one of the three outcome measures described below (note that all the children except six—three from each of the two middle age-at-implant groups—completed all three outcome measures), and participants for whom we could estimate the family's income. This resulted in a total of 96 children. The children were stratified into four groups according to age at implantation. Group 1 (N = 6) was implanted between 6 and 12 mo of age (M = 10.2 mos), group 2 (N =32) between 13 and 24 mo of age (M = 18.6 mos), group 3 (N = 37) between 25 and 36 mo of age (M = 29.9 mos), and group 4 (N = 21) between 37 and 48 mo of age (M = 40.8 mos). Figure 1 displays a scatterplot of age at implantation for all 96 participants. Age at implantation was relatively evenly distributed within each age group. Table 1 displays demographic information for each group, including the number of participants, mean age at initial stimulation, mean unaided best-ear PTA (at 0.5, 1, and 2 kHz), proportion of oral

communicators, proportion female, and mean estimated family income. Oral communication (OC) relies on oral speech only and does not use signing, whereas total communication (TC) combines oral speech with signing in English word order. To address the fact that children occasionally switched communication methods during the course of the investigation, we first calculated the proportion of time a given child used OC before determining the proportion of OC users. For example, if a child were tested at five intervals and used OC at three of those intervals, she/he would be considered 0.6 OC in the analyses. In any case, less than 5% of the subjects changed their CM during the course of this study. PTA and proportion female varied little across groups. There was a trend for children implanted at younger ages to have a higher proportion of OC users than children implanted at older ages. Finally, mean estimated family income varied over approximately an $8000 range, although there was no systematic variation related to age at cochlear implantation.

Family Income Estimation

Estimates of family income were included in the analysis because income is one measure of a family unit's socioeconomic status (SES) and there is evidence that SES influences children's health and their cognitive and socioemotional development (see review by Bradley & Corwyn, 2002), including language development (Hart & Risley, 1995). Admittedly, there are factors that mediate the relationship between SES and a child's development; further, there are a variety of methods available to capture a family's resources, such as obtaining information on income, occupational status, maternal and/or paternal educational achievement, some combination of these, and collective SES. This final method is a community-level SES measurement that addresses the neighborhood of residence's influence on a child's development. Because the "choice of how to measure SES remains open" (Bradley & Corwyn, 2002, p. 373), we chose to use median family income at the time the child was implanted to estimate each family's SES. However, when families enroll in our research, we do not inquire about their income. Therefore, we used an indirect measure of each child's family income. This was obtained using data from the 2000 US Census Bureau's TIGER (Topologically Integrated Geographic Encoding and Referencing) database (www.census.gov/main/www/cen2000.html). First, the census tract and block group were identified for each participant's street address at the time of cochlear implantation. The census tract and block group are successively smaller geographical areas around a particular street address. Block group size varies, but is approximately 2 to 4 blocks in size. In the second step, we identified the median household income in 1999 associated with each participant's block group. This method has a number of advantages over traditional measures of SES. First, it avoids the potential bias posed by some questions that patients or their parents may be reluctant to answer, a problem that limits the validity of some SES indices. Additionally, the block group census-based measure is easy to obtain and greatly reduces nonresponse.

Materials

Mr. Potato Head—The Mr. Potato Head Task (Robbins, 1994) is a live-voice, auditory-only modified open-set word recognition test. Mr. Potato Head is a children's toy that consists of a "potato" body and approximately 20 body parts and accessories, which can be attached to the "potato" body. The experimenter reviews the names of the body parts and lets

the child play with the pieces briefly. Then, the child is told to do exactly what the experimenter says. Children are given auditory-only, sentence-length instructions related to the toy, and its various parts. Based on their response to the instructions, two scores are derived: sentence and key word correct scores. Only children's key word correct scores were used. The key word score is based on the number of key words (of 20) identified correctly. For example, one test item is, "He wants *green shoes*." In this example, "green" and "shoes" are the key words. The child would get one word correct if she/he picked up or pointed to any pair of shoes or if she/he picked up or pointed to a green accessory. The child would get both words correct if she/he picked up or pointed to the pair of green shoes. Although the task is technically a closed-set task, because there are so many response options (20), the key word task is sometimes called a modified open-set with estimated chance performance equal to 5% correct.

Reynell developmental language scales—The outcome measure used to assess language development was based on the Reynell Developmental Language Scales (RDLS-III; Edwards, et al., 1997). Scores observed using the RDLS, expressed as age-equivalent scores, were used whenever a child performed above the test's floor. When the child's skills were more rudimentary, predicted RDLS scores were obtained based on data from the MacArthur Communicative Development Inventories (MCDI; Fenson, et al., 1993). The predictive functions were developed in an earlier study (Stallings, et al., 2000) of 91 pediatric CI users who were administered both the RDLS and one of the MCDI forms within the same testing session. In the current investigation, approximately 16% of the language data were predicted from the Words and Gestures version of the MCDI and approximately 15% were predicted from the Words and Sentences version of the MCDI. The RDLS assesses expressive and receptive language separately. The RDLS and MCDI were chosen to assess language development because they have been extensively normed on children with normal hearing and can be applied to users of either OC or TC. The option of conducting tests in these two modalities is important for measuring the children's underlying language abilities, as far as possible, independently of their ability to understand spoken language or to produce intelligible speech. The RDLS has been used extensively with deaf children (including CI users, see e.g., Bollard, et al., 1999; Svirsky, 2000; Svirsky, et al., 2000) and is appropriate for a broad age range (1–8 yr). Normative data are also available for more than a 1000 hearing children (Edwards, et al., 1997). The Kuder-Richardson reliability coefficients are 0.97 for the receptive language test and 0.96 for the expressive language test. Finally, the test format involves object manipulation and description based on questions varying in length and grammatical complexity, reflecting real-world communication, and assessing linguistic competence more accurately than single-word vocabulary tests. The MCDI offer a valid and efficient means of assessing early language development, using a parent report format. Two levels of complexity are available for the MCDI and are administered according to the age of the child. The MCDI/Words and Gestures is designed for 8- to 16-mo olds, and the MCDI/Words and Sentences is designed for 16-to 30-mo olds. Consistent with Stallings et al., parents were instructed to indicate which words a child comprehends (either orally or signed) or produces (either orally or signed).

Procedures—Children were tested before cochlear implantation and at approximately regular 6-mo intervals after initial stimulation. Not all children were tested at every interval because of time constraints, lack of ability to maintain attention for all tests, and missed appointments. The test materials were administered and scored by licensed speech-language pathologists with training in working with deaf children with CIs. Testing was conducted in a quiet room. The stimuli were presented live-voice at approximately 70 dB SPL. The Mr. Potato Head test was administered auditory-only, whereas the RDLS was administered with auditory and visual speechreading cues (for those who use OC) or with auditory, visual speechreading, and sign cues (for those who use TC). Test instruction was performed in the child's primary mode of communication. Signed and spoken responses were accepted for both test measures. Parents filled out the MCDI/Words and Gesture and/or the MCDI/Words and Sentences questionnaires while attending their child's testing session.

Data Analysis

Developmental trajectory analysis—The developmental trajectories of different age-at-implant groups were compared using developmental trajectory analysis (DTA, first described by Svirsky, et al., 2004, and further refined here). The first important concept in DTA is that the parameter under analysis is derived from a comparison between whole developmental trajectories rather than from measures taken at a single age. Figures 2 and 3 illustrate the concept behind DTA, as well as the specific method that was used. As an example, let groups 1 and 2 be two groups of children implanted at different age ranges (with 3 and 4 members in each group, respectively, in this example). The curves for children in groups 1 and 2 are illustrated in the left and center panels of Figure 2, respectively. The right panel of Figure 2 shows how curves are compared from two individuals, one from group 1 and one from group 2. Each curve is determined by connecting all successive pairs of data points for a given individual. In other words, we are using linear interpolation between each pair of successive measurements. The figure illustrates how we are also linearly interpolating between the origin and the first data point. The two curves are compared within the age range that extends from T_{min} to T_{max}, which are indicated in the x axis of the right panel. T_{min} is the age at which the first postimplant testing session was held, for at least one of the two children. In the right panel of Figure 2, the first postimplant testing session takes place at about 11 mo of age for one child and 23 mo for the other, so in this case T_{min} is 11 mo. T_{max} is the latest age at which there are available data (either measured or interpolated) for both children. In the example, the latest data point for the earlier implanted child is 84 mo. The later implanted child does not have data at 84 mo but he does at 74 and 90 mo of age, allowing us to obtain interpolated data at mo 84. Thus, in this example T_{max} is 84 mo. The comparison range is indicated by the horizontal arrow labeled "T."

So far in this example we have constructed two developmental curves, one for each child, and we have chosen the age range over which we will perform a comparison for this pair of individuals. The right panel of Figure 2 also shows how the actual comparison between two children's developmental curves is done. There are several arrows indicating the advantage for one child over the other one at many different ages between T_{min} and T_{max}. In general these values will be different at different ages but it is possible to obtain an average value of

all these differences, and this average value is depicted by the thicker arrow in the figure next to the label “D.” Let us call this average value the “developmental difference” between the two children. DTA uses sets of developmental differences obtained in an orderly sequence, as explained below. It should be noted that the vertical arrows in Figure 2 are simply used to explain the developmental difference concept, but the actual calculation is done with more precision using Eq. (1), which is also explained below.

Figure 3 illustrates how we can examine differences among groups rather than just two individuals. The left column of panels in Figure 3 shows the calculation of the developmental difference between the first member of group 1 (named S1) and each member of group 2; the middle column of panels shows the same calculation for S2 (the second member of group 1) and each member of group 2; and the right column of panels shows developmental difference calculations for the third and last member of group 1 (named S3) and each member of group 2.

The developmental difference, $D_{\mathrm{S1,SA}}$, between subject S1 from group 1 and subject SA from group 2 (illustrated by the vertical blue arrow in the top left panel of Fig. 3) is calculated as follows:

$$D_{\mathrm{S1,SA}} = \frac{\int_{T_{1,\mathrm{A\,min}}}^{T_{1,\mathrm{A\,max}}} [\mathrm{S1}(t) - \mathrm{SA}(t)]\mathrm{d}t}{T_{1,\mathrm{A\,max}} - T_{1,\mathrm{A\,min}}} \quad (1)$$

As explained above, the upper integration limit $T_{1,\mathrm{A\ max}}$ is the maximum age value for which both developmental curves (S1 and SA) are defined, and $T_{1,\mathrm{A\ min}}$ is the age at which the first postimplant testing session was held for S1 (which is earlier than the corresponding age for SA). Thus, the developmental difference, $D_{\mathrm{S1,SA}}$, is not simply the area between the developmental trajectories S1 and SA. It is that area divided by ($T_{1,\mathrm{A\ max}} - T_{1,\mathrm{A\ min}}$), the length of the integration domain. This is a crucial point and it is the reason why $D_{\mathrm{S1,SA}}$ represents the average size of the difference between S1 and SA, averaged over the whole analysis period, as illustrated in the right panel of Figure 2. In any case, the blue arrow in the top left panel of Figure 3 represents the developmental difference between the two curves shown in that panel, those of subjects S1 and SA. The other panels in the left column of Figure 3 show the calculation of the developmental difference between S1 and each one of the other members of group 2 (SB, SC, and SD). The bottom left panel of Figure 3 shows how all these developmental differences are averaged, resulting in a single number that compares subject S1 to group 2 as a whole:

$$D_{\mathrm{S1,Group\ 2}} = \frac{1}{n}\sum_{j=1}^{n} D_{\mathrm{S1,Sj,}} \quad (2)$$

where n is the number of subjects in group 2.

The panels in the middle and right columns of Figure 3 show how this process is repeated for S2 and S3, the other members of group 1, obtaining $D_{S2,Group\ 2}$ and $D_{S3,Group\ 2}$. The bottom line, then, is that we obtain measures of developmental difference between each individual member of group 1, and group 2 as a whole. Let us name these measures $D_{Si,Group\ 2}$. To test whether the developmental trajectories from group 1 are significantly different from those of group 2, the following null hypothesis can be used:

> H_0: The set of numbers $D_{Si,Group\ 2}$ (i = 1 to n) are a sample taken from a normal distribution with a mean of zero.

Here, n is the number of curves in group 1, which is 3 in the example illustrated in Figure 2. Hypothesis H_0 can be easily tested using the Student distribution or (an alternative preferred by many statisticians, particularly for small-sized samples) the Wilcoxon or exact permutation tests. Finally, DTA can also be used to measure the *average* group difference between two sets of developmental trajectories. This is done simply by averaging the developmental differences $D_{Si,Group\ 2}$ for all members of group 1 with respect to the members of group 2:

Average Developmental Difference

$$\text{Average Developmental Difference} = \frac{1}{m}\sum_{i=1}^{m} D_{\text{Si,Group 2}} \quad (3)$$

Here, m is the total number of members of group 1.

DTA also can be used when the two groups to be compared differ in confounding variables that are presumed to have an effect on the outcome measure (such as residual hearing or CM). This may be done by calculating the “confounding variable differences” in a way that is similar to the calculation of developmental differences. First, we calculate the confounding variable difference between one subject from group 1 and each subject from group 2. For example, the confounding variable difference between subjects S1 and SA is:

$$\text{CV}_{\text{S1,SA}} = \frac{\int_{T_{1,\text{A min}}}^{T_{1,\text{A max}}} [\text{CV}_{\text{S1}}(t) - \text{CV}_{\text{SA}}(t)]\,dt}{T_{1,\text{A max}} - T_{1,\text{A min}}}, \quad (4)$$

where $CV_{S1}(t)$ and $CV_{SA}(t)$ are the values of the confounding variable for each subject. If the confounding variables are constant during the analysis period, then $CV_{S1,SA}$ is equal to $(CV_{S1} - CV_{SA})$. Then, the average value of the confounding variable difference between S1 and group 2 is calculated:

$$\text{CV}_{\text{S1,Group 2}} = \frac{1}{n}\sum_{j=1}^{n} \text{CV}_{\text{S1,Sj,}} \quad (5)$$

where n is the number of subjects in group 2.

Finally, instead of testing the null hypothesis H_0 listed above, a regression is performed (or multiple regression, if there is more than one confounding variable) of $D_{\text{Si,Group 2}}$ as a function of $\text{CV}_{\text{Si, Group 2}}$. In the example of a linear case, the regression equation would be $D_{\text{Si,Group 2}} = a \times \text{CV}_{\text{Si, Group 2}} + b$, and the null hypothesis would be:

> H_0': *The intercept of the regression function is zero* (or, in other words, the developmental differences are due exclusively to the effect of the confounding variable).

The DTA procedure is well suited to answer the following question: Which group of children shows a better outcome, averaged throughout the entire follow-up period? This type of analysis is complementary to that used by Geers et al. (2003), comparing different age-at-implant groups at the same age, several years after implantation. The latter indicates whether late-implanted children catch up with early-implanted children at some point, whereas DTA is used to evaluate whether one group had an advantage over the other, averaged over the analysis period. In the present case, the analysis period falls within the first years of life. All other things being equal, if a certain age-at-implantation results in improved speech intelligibility, speech perception skills, or language development, this age at implantation should be preferred, even if the later-implanted group eventually catches up with the earlier-implanted group.

In the present study, each comparison was conducted in both directions. For example, the developmental differences resulting from comparing each child implanted at 13 to 24 mos to the group of children implanted at 25 to 36 mos were calculated, and a test was applied to determine whether this set of differences was significantly different from zero. Then, the process was repeated for the set of developmental differences resulting from comparing each child implanted at 25 to 36 mos with the group of children implanted at 13 to 24 mos. The most conservative of the two comparisons was selected to express the significance of the difference between the groups. The comparisons themselves were performed by stepwise multiple linear regression of each one of the outcome measures to examine the differences between the age-at-implant groups, after adding the following independent variables to the regression: unaided best-ear PTA, CM, gender, and estimated family income.

The programs to implement the DTA method and to find the average curve in a given age-at-implant group were written in MATLAB and are available from the corresponding author.

Hierarchical linear modeling—HLM (Raudenbush, et al., 2004) is a tool for statistical modeling of two- and three-level data structures. It is particularly useful for behavioral data because they tend to have nested structures. For example, data are nested in repeated observations within participants, within age-at-implant groups, and within the covariates. Each sublevel makes up a portion of the hierarchical linear model and represents the relations between that level's factors and its residual variability. For our purposes, a two-level hierarchical linear model was used to examine the differences in the slopes of the developmental trajectories as a function of age at implantation while evaluating the effects of the covariates (CM, family's income [Inc], PTA, and gender). A two-level HLM analysis

involves a two-step process resulting in a model for each step. The level-1 model captures the relations among the participant-level variables. In this first step, we fit a least-squares line to each developmental trajectory curve to obtain slope and intercept values for each curve. To obtain more accurate estimates of slopes for changes in Potato Head scores as a function of time, we only included data up to the point where a child reached ceiling scores (90% correct or higher). The level-1 model used in the first step was

$$Y = \text{Intercept } 1 + \text{Slope}(\text{Test age}) + \text{Error}$$

The regression parameters obtained in the first step were then used as "data" in the second step, in which we performed linear regression for slope as a function of age at implantation and the covariates. In other words, the coefficient used in the level-1 model (slope) becomes an outcome variable in the level-2 model. The model used in the second step was

$$\text{Slope} = \text{Intercept } 2 + \text{B1}(\text{Age at implant}) + \text{B2}(\text{CM}) + \text{B3}(\text{Inc}) + \text{B4}(\text{PTA}) + \text{B5}(\text{Gender}) + \text{R1}$$

Thus, if coefficient B1 turned out to be significantly different from zero, this would mean that age at implant has an influence on the slope of the developmental trajectory of the outcome measure independently of the influence of the confounding variables (CM, income, PTA, and gender).

It is important to note that, unlike the other analyses used in this study, the HLM analysis was performed using age at implant as a continuous variable. In other words, the analysis was performed for all 96 subjects without breaking them down into groups.

Results

The purpose of this study was twofold: (1) to examine performance of a small group of children implanted before age 1 yr and to compare their communicative performance with that of children implanted at later ages, while controlling for potential covariates; and (2) to evaluate whether there is behavioral evidence for sensitive periods in spoken language development.

Word Recognition

Word recognition performance on the Mr. Potato Head task is displayed in Figure 4. Panel A displays the average performance of each age-at-implantation group up until the point at which there were data for at least four participants. The longer a given group of participants is followed, the fewer children remain in the group, mostly because some children have not had as long a follow-up period as others. The requirement of having at least four participants in each average curve displayed in Figure 4 helps make that graphical display more meaningful. For comparison purposes, the average scores of normal-hearing children obtained by Kirk et al. (1997) and Robbins and Kirk (1996) are shown by the thick black line. The arrows along the x axis in Panel A indicate the average age at implantation for each group. Panels B to E display both individual and group data for each age-at-implantation

group separately, along with the normative data from normal-hearing children. There is a trend for the average developmental trajectory to change upon implantation: there is an inflection point in the curves of the three older age-at-implant groups within a few months of cochlear implantation after which the rate of development increases. Further, there is a trend for children implanted before age 2 yrs to have higher word recognition scores at a given chronological age than children implanted later than 2 yr. Although the average implanted child, regardless of the age at which she/he received the device, performs more poorly than her/his normal-hearing peers, there are some children in each group who perform similar to normal-hearing children on this word recognition task.

The word recognition data were entered into the DTA. Recall that the output of the DTA is an estimate of the average difference in a specific outcome variable over a specific period of time between each participant's developmental trajectory and that of each participant in every other age at implant group. Recall that this value is called "Average Developmental Difference." Table 2 displays results from the multiple linear regression analyses. The top portion of the table shows the results for the word recognition measure. The Average Developmental Difference values for the children implanted before age 1 yr and those implanted between ages 1 and 2 yr (group 1 versus 2) were not significantly different, but they were significantly different from children implanted between 2 and 3 yr of age (group 1 versus 3). Further, there were significant differences in Average Developmental Difference values between children implanted between ages 1 and 2 yr and those implanted between 2 and 3 yr (group 2 versus 3), and between children implanted between 2 and 3 yr, and those implanted between ages 3 and 4 yr (group 3 versus 4). The significant mean Average Developmental Difference values varied between 15 to 18 percentage points with children in the younger age-at-implant groups achieving higher scores than those in the older age-at-implant groups. There are two ways in which the data can be analyzed: (1) individuals in one group (e.g., group A) could be compared with the average of the second group (e.g., group B); or (2) individuals in group B could be compared with the average of group A. The mean Average Developmental Difference values were identical and reached the same level of significance regardless of which method was used. Therefore, only one mean Average Developmental Difference data column is displayed in Table 2.

The covariates (CM, Inc, PTA, and gender) were entered into the stepwise multiple linear regression to determine if they could account for any additional variability in Average Developmental Difference beyond age-at-implantation. In this case it did matter to some extent which direction the comparisons were made. Therefore, two data columns appear in Table 2 under "stepwise." For word recognition performance, PTA accounted for a significant additional amount of variability in Average Developmental Difference ($p < 0.05$) when comparing groups 2 and 3 (children implanted between 1 and 2 yrs and those implanted between 2 and 3 yrs) and groups 3 and 4 (children implanted between 2 and 3 yrs and those implanted between 3 and 4 yrs) when comparing individual data from the younger age at implant group with average data from the older age-at-implant group. CM accounted for a significant additional amount of variability in Average Developmental Difference when comparing groups 3 and 4 (children implanted between 2 and 3 yrs and those implanted between 3 and 4 yrs) when comparing individual data from the older age-at-implant group with average data from the younger age-at-implant group. These results suggest that some of

the group differences in word recognition ability are partially due to differences in unaided PTA and the CM the child uses.

Language

Figures 5 and 6 are set up similar to Figure 4, but display data for the Receptive and Expressive Scales of the RDLS, respectively. The thick diagonal black line in each panel displays age-appropriate performance, whereas the alternating dotted/dashed lines show 1 and 2 SDs below the mean for typically developing, normal-hearing children on the RDLS. The vast majority of the children had delayed language skills, regardless of the age at which they received their CIs; however, there was a trend for more children in the younger age-at-implantation groups to perform within 2 SDs of the mean of normal-hearing children than for children in the older age-at-implantation groups.

As with the word recognition results, the results from the multiple linear regression analyses for the language measure are displayed in Table 2. The middle and bottom portions of the table show the results for receptive and expressive language, respectively. The Average Developmental Difference values for all group comparisons on receptive language were significant ($p < 0.05$). Stepwise multiple linear regressions for receptive language performance revealed that estimated family income accounted for a significant additional amount of variability in Average Developmental Difference ($p < 0.05$) when comparing groups 1 and 2 (children implanted between 6 and 12 mos and those implanted between 1 and 2 yrs) and groups 1 and 3 (children implanted between 6 and 12 mos and those implanted between 2 and 3 yrs) and when individual data from the younger age-at-implant group are compared with average data from the older age-at-implant group. PTA accounted for a significant additional amount of variance in Average Developmental Difference when comparing groups 3 and 4 (children implanted between 2 and 3 yrs of age and those implanted between 3 and 4 yrs of age) and when individual data from the younger age-at-implant group are compared with average data from the older age-at-implant group. Further, PTA accounted for a significant additional amount of variance in Average Developmental Difference when comparing groups 2 and 3 (children implanted between 1 and 2 yrs of age and those implanted between 2 and 3 yrs of age) and when individual data from the older age-at-implant group are compared with average data from the younger age-at-implant groups. These results suggest that group differences in receptive language partially are due to differences in PTA and estimated family income.

Similar to the word recognition results, the Average Developmental Difference values for expressive language performance for the children implanted before age 1 yr and those implanted between age 1 and 2 yrs (group 1 versus 2) were not significantly different, but they were significantly different from children implanted between ages 2 and 3 yrs (group 1 versus 3). Further, there were significant differences in Average Developmental Difference values between children implanted between ages 1 and 2 yrs and those implanted between 2 and 3 yrs (group 2 versus 3) and between children implanted between 2 and 3 yrs and those implanted between ages 3 and 4 yrs (group 3 versus 4). No additional variance in expressive language Average Developmental Difference could be accounted for with the covariates in the stepwise multiple linear regression analyses.

DTA Statistics Summary

Overall the results of the multiple linear regression analyses on the DTA output suggest that there are significant differences in the developmental trajectories of children implanted earlier rather than later in the early childhood years. However, the advantage of implanting children before 12 mos of age versus waiting until the child is between ages 1 and 2 yrs only is evident in receptive language skill development, not in expressive language and word recognition skill development in our group of children. For the most part, PTA and income played a limited role in accounting for an additional amount of variance in Average Developmental Difference in both word recognition and receptive language ability. CM accounted for additional variance in Average Developmental Difference in one instance and gender never accounted for additional variance. Further, none of the selected covariates accounted for differences in the developmental trajectories of expressive language development among the groups.

Hierarchical Linear Modeling Analysis

Although the DTA analyses examined performance differences among children implanted at various ages, HLM (Raudenbush, et al., 2004) was used to examine differences in the slopes of the developmental trajectories as a function of age at implantation while evaluating the effects of covariates. The slopes of the developmental trajectories reflect the rate at which children are acquiring a specific skill. If the slope is equivalent across age-at-implant groups, this would suggest there is no sensitive period for developing that specific skill once auditory input is provided some time during the first 4 yrs of life. However, if the slopes get increasingly shallow as the age at implantation gets greater, it would suggest that there is a sensitive period for developing that specific skill during the first 4 yrs of life. The two-step process began with fitting a least-squares line to each curve thereby obtaining a slope and an intercept for each. The second step involved performing a linear regression for slope as a function of age at implantation and the covariates.

Table 3 displays a summary of the HLM analysis. The results indicate that CM was the only covariate that had a significant influence on the rate of word recognition acquisition: children using OC had faster rates of word recognition acquisition than children raised in TC environments. Further, age at implantation did not significantly influence the slopes of the word recognition developmental trajectories. In contrast, age at implantation did significantly influence the rate of both receptive and expressive language acquisition: children implanted earlier in life had faster rates of spoken language acquisition than children implanted later in life. Estimated family income had a significant effect on the rate of receptive and expressive language development. However, the effect of income was different for receptive and expressive language development: children from families with higher estimated incomes had faster rates of receptive language development, but slower rates of expressive language development, relative to children from families with lower estimated incomes.

Discussion

The first goal of this research was to explore whether significant gains in spoken language skills are made by children implanted before 1 yr of age relative to those implanted at later ages, while controlling for potential covariates. Up through about 3 yrs of age, our small group of children implanted between ages 6 and 12 mos failed to show significant performance differences on two of the three outcome measures from those implanted in the second year of life, but did show significantly higher scores than children implanted at 2 yrs of age or later. Further, children implanted before age 2 yrs had higher performance than children implanted in the third or fourth years of life and children implanted in the fourth year of life had significantly poorer performance than children implanted earlier. These findings support those of previous investigators (Fryauf-Bertschy, et al., 1997; Nikolopoulos, et al., 1999; Svirsky et al., 2004; Tyler, et al., 1997), while extending the results to a new population of CI recipients: infants implanted younger than 1 yr. The results differ to some extent from those of Dettman et al. (2007) who reported significantly different rates of receptive and expressive language development in infants implanted in the first year of life relative to those implanted in the second year of life. Both our and Dettman et al.'s participants had significantly better receptive language development when implanted at the age of 1 yr, despite using different language measures. On the other hand, we did not find significant differences in rates of expressive language development. Dettman et al. did not evaluate spoken word recognition, so we cannot compare the development of that skill across the two studies. Dettman et al. had five more participants in the youngest age-at-implant group than was used in the current investigation. We also used different language measures. Both of these factors could have influenced the somewhat discrepant results.

These new findings suggest that in this small sample of six children implanted between 6 and 12 mos of age, no significant gains in expressive language development and spoken word recognition are observed by implanting children before their first birthday than doing so before age 2 yrs. There was, however, an advantage for receptive language development. The majority of the small group implanted between 6 and 12 mos of age was followed for 2 to 2.5 yrs after cochlear implantation and thus, was only 3 yrs old at the most recent testing interval. Therefore, it is possible that performance differences might become evident as the children develop and gain more experience with their devices, or in studies with a greater N. Although these measures of spoken language development have been used extensively with the pediatric CI population, it is also possible that other measures might be more sensitive to early performance differences between children implanted before 1 yr of age and those implanted between 1 and 2 yrs of age. Perhaps the greatest limitation to this investigation is the small number of participants in the youngest age-at-implant group. Few infants younger than 12 mos have been implanted at most CI centers making it difficult to achieve the power we have become accustomed to in CI research. Our not finding significant differences in performance on spoken word recognition and expressive language development between the children implanted in the first and second years of life should be interpreted with this power limitation in mind.

Significant performance differences remained among groups despite accounting for four potential confounds: better-ear PTA, family's estimated income, CM, and gender. PTA,

estimated family income, and in one case, CM accounted for an additional significant amount of variance (beyond age at implantation) in performance among age-at-implant groups. In this group of children, gender accounted for no additional variance in outcome. Further, no covariates accounted for any additional variance in expressive language development among the groups of children.

Our second goal was to investigate whether there is evidence for sensitive periods in spoken language development. Prelingually deaf children with CIs provide a rare opportunity to examine sensitive periods in a natural environment. Because of their early onset of deafness, the children lacked auditory input for various lengths of time. Then through intervention with a CI, children were given access to auditory input at various ages. These circumstances provide a unique opportunity to study the effects of spoken language deprivation and subsequent intervention. Children implanted at later ages (up to 4 yrs), had slower rates of oral language development than children implanted at earlier ages, suggesting that there is a sensitive period for spoken language development within the first 4 yrs of life. In contrast to the language scores, the rate of increase in open-set speech recognition was similar regardless of the age at implantation (up to age 4 yrs), suggesting that any sensitive period for relatively simple spoken word recognition development may extend beyond the first 4 yrs of life.

The behavioral evidence for sensitive periods reported here parallels the evidence for sensitive periods in the electrophysiological literature. Development of the auditory cortex typically extends through the teenage years (Moore & Guan, 2001). A review of auditory cortex maturation by Eggermont and Ponton (2003) shows that maturation begins structurally with axons in the superficial layer I of the auditory cortex through 4.5 mos of age and then moves to layers IV, V, and VI through 5 yrs of age. Between 5 and 12 yrs of age, layers II and III axons begin to mature and cortical axons mature only after 12 yrs. Changes in electrophysiological recordings are believed to reflect the maturation of these structures: the ABR, P_2, and N_2 are believed to reflect maturation of the superficial layer I axons; the middle latency response and mismatch negativity are believed to reflect maturation of layers IV, V, and VI; the maturation of P_1 and emergence of N_1 are believed to reflect layers II and III axon maturation; and finally, the maturation of N_1 is believed to reflect the maturation of cortical axons. Eggermont and Ponton examined the development of these electrophysiological markers in children who received CIs. They found that the P_1 latency was delayed in children with CIs relative to their normal-hearing age-matched peers, but that the P_1 latencies were commensurate with hearing-age-matched peers. The authors suggested that the middle layers of cortical tissue are in a state of arrested development without auditory input, but that once cochlear implantation takes place, the development of the structures generating the P_1 begin to develop at an appropriate rate (although it is still delayed by approximately the length of deafness). In two case studies of pediatric CI recipients who were implanted at age 6 yrs and were either deaf from birth or from age 3.5 yrs, Ponton and Eggermont (2001) and Ponton, et al. (1999) reported that initial maturation of the P_1 latency began maturing at the same rate as normally hearing children, but that N_1 never emerged, suggesting an “arrest or alteration in the maturation of the layer II axon neurofilaments” (Ponton & Eggermont, 2003, p. 251) without auditory input during the sensitive period of development, believed to be between 3 and 6 yrs of age.

Consistent with Ponton et al.s' findings, Sharma et al. (2002) found delayed P_1 latencies just after implantation in cochlear implanted children. Further, they found that children implanted after 11 yrs of age, showed little or no change in the P_1 latency over 1 to 2 yrs of CI experience, but that children implanted by age 7 yrs showed maturation of (decreases in) the latency of P_1. More important to the sensitive periods argument was the finding that children implanted before age 3.5 yrs showed large decreases in the latency of P_1 over the early months of CI use.

Taken together, these electrophysiological and our new behavioral findings suggest that the auditory system is maximally plastic during the first 2 to 3.5 yrs of life. The electrophysiological data suggest a longer window (~3.5 yrs) than the behavioral data (~2 yrs). Further, there seems to be some limited plasticity until 7 yrs of age, after which much slower and more limited gains are made. Although our behavioral data can only address effects of auditory deprivation until age 4 yrs, they do suggest that the window of opportunity for language acquisition is not yet closed at that age, but is starting to close.

Of the covariates examined, CM significantly influenced the rate of word recognition but not language development and estimated family income had a significant influence on the rate of language but not word recognition development. One possible explanation for why CM only influenced word recognition development is that because we were interested in children's auditory word recognition abilities, this task was the only one administered exclusively in the auditory modality. Perhaps the OC children were at an advantage in the word recognition task because they were tested in their everyday mode of communication. The findings on family income parallel data on normal-hearing, typically developing children, suggest that even in this population of children who receive impoverished spoken language input (at least early in life), the correlation between SES and rate of language development remains. The relationship between SES and language development might not be a direct one, though. Data from Hoff (2003) suggesting that the relationship between SES and rate of language development might primarily be mediated by factors related to maternal speech.

Our exploratory results suggest that the benefits of cochlear implantation in the first year of life instead of the second are relatively small. However, this is based on a small sample of six children followed for 2 to 2.5 yrs postimplantation. It is possible that other measures of performance might yield greater performance differences and/or that following a larger number of children for a longer period of device use might reveal some additional performance gains. On the other hand, our review of the literature suggests that the risk of anesthesia and the risk of misidentifying children as profoundly hearing impaired are also relatively low, provided appropriate measures are taken. These measures include the administration of several different tests to confirm the level of hearing impairment (OAE, ABR, ASSR, and VRA, if possible) and the presence of a pediatric anesthesiologist during surgery.

Even though children implanted before 1 yr of age had limited advantages in spoken language skills with respect to those implanted in the second year of life, they did show better performance than those implanted after age 2 yrs. Further, the rate of language acquisition was significantly faster for children implanted at earlier than at later ages,

thereby providing evidence of a sensitive period for language development through the first 4 yrs of life. This finding has both theoretical and practical implications. From a theoretical perspective, it provides rare behavioral evidence that corroborates the physiological evidence suggesting sensitive periods for language development. From a practical perspective, it suggests that language acquisition will approach age-appropriate levels faster the earlier a child is implanted. It does not suggest that language development will not occur if children are implanted in the fourth year of life, but it does suggest that, on average, their rate of acquisition will be slower and they will continue to lag behind their normal-hearing peers for a longer time. Language acquisition is critical for the development of literacy skills and success in academic settings. Presumably, the earlier a child attains age-appropriate or nearly age-appropriate language skills, the more prepared she/he will be to successfully enter school.

Finally, these data suggest that certain patient variables, particularly better-ear PTA, family's estimated income, and CM, account for some variability in cochlear implantation outcomes. The search for causes of the large intersubject variability in co-chlear implantation outcomes has often met with limited success. Some factors that have been reported to account for some of the variability in outcomes include CM, family size, performance IQ (Geers, et al., 2003), working memory capacity (Pisoni & Cleary, 2003; Pisoni & Geers, 2000), and articulation rate and verbal rehearsal speed (Pisoni & Cleary, 2003). A family's estimated income is a new factor that has not been widely explored in the CI literature. Our data suggest that income primarily has an influence on children's rates of language development rather than on the development of relatively simple spoken word recognition skills. Geers et al. (2003) has suggested that the relationship between spoken language outcomes in pediatric CI users and family SES is mediated by performance IQ. We did not examine performance IQ or maternal input to the children in this study, so we cannot speculate on whether the relationship between estimated family income and language development rate is mediated by performance IQ as Geers et al. have suggested, by maternal input as Hoff (2003) has suggested, or if the relationship is more direct. In any case, our data do suggest a relationship between a family's resources and children's language development rates. However, the relationship is not straightforward. Children from families with a higher estimated income had faster rates of receptive language development, but slower rates of expressive language development, relative to children from families with lower estimated incomes. It is possible that children from families with higher estimated incomes have mothers who provide more language input (as Hoff suggested) and that this transfers to receptive language skills first and the transfer to expressive skills is not yet apparent. Another explanation could be that children of families with lower estimated incomes have mothers who provide less language input (as Hoff suggested) thereby opening up more opportunities for children to use their expressive language skills.

Although this investigation longitudinally followed a large cohort of children with CIs (N = 96), the primary group of interest—those implanted in the first year of life—included a small number of children (N = 6). As discussed before, both the risks and the benefits of implanting children at 6 to 12 mos as opposed to 13 to 24 mos seem relatively low. It is important to note that these considerations change in the case of children who had meningitis because the cochlear ossification that can result from this disease makes surgery

more difficult. A diagnosis of meningitis, then, tips the scales in favor of earlier implantation. Our results do suggest that there is a clear benefit to implanting before a child's second birthday. Not only do children achieve higher spoken language abilities at earlier ages, they also show a faster rate of spoken language acquisition, thereby providing more evidence for early sensitive periods in spoken language development. As more families push to have their infants implanted before the age of 1 yr and as those children have more experience with their devices, further examination of the issue of the "best" age at which to implant children might reveal some additional important findings.

Acknowledgments

The authors thank Noelia Guazzone who assisted in compiling the family income data, Elizabeth Ying, Shirley Henning, and Sonja Solomonson who collected the data, Bruce Tomblin who gave us advice on the use of HLM and how to obtain family income data, Elad Sagi who helped with figure preparation, and the children and their families who participated in the research. The programs to implement the DTA method and to find the average curve in a given age-at-implant group were written in MATLAB and are available from the corresponding author.

This work was conducted while both authors were at the Indiana University School of Medicine.

This work was supported by grant NIH-NIDCD R01 DC00423 (to R. T. M.), T32 DC00012 (to D. B. P.), and R01 DC03937 (to M. A. S.).

References

Bollard PM, Chute PM, Popp A, et al. Specific language growth in young children using the CLARION co-chlear implant. Ann Otol Rhinol Laryngol. 1999; 177(suppl):119–123.

Bradley RH, Corwyn RF. Socioeconomic status and child development. Annu Rev Psychol. 2002; 53:371–399. [PubMed: 11752490]

Clifton, RK., Morrongiello, BA., Kulig, JW., et al. Developmental changes in auditory localization in infancy. In: Aslin, R.Alberts, J., Petersen, MR., editors. Development of perception. New York: Academic Press; 1981. p. 141-160.

Cohen MM, Cameron CB, Duncan PG. Pediatric anesthesia morbidity and mortality in the perioperative period. Anesth Analg. 1990; 70:160–167. [PubMed: 2301747]

Dettman SJ, Pinder D, Briggs RJS, et al. Communication development in children who receive the cochlear implant younger than 12 months: risks versus benefits. Ear Hear. 2007; 28(suppl):11S–18S. [PubMed: 17496638]

Edwards, S., Fletcher, P., Garman, M., et al. The Reynell developmental language scales. III. The University of Reading edition. Los Angeles, CA: Western Psychological Services; 1997.

Eggermont JJ, Ponton CW. Auditory-evoked potential studies of cortical maturation in normal hearing and implanted children: correlations with changes in structure and speech perception. Acta Otolaryngol. 2003; 123:249–252. [PubMed: 12701751]

Eisenberg LS, House WF. Initial experience with the cochlear implant in children. Ann Otol Rhinol Laryngol. 1982; 91(suppl):67–73.

Fenson, L., Dale, PS., Reznick, JS., et al. The MacArthur communicative development inventories. San Diego, CA: Singular Publishing Group; 1993.

Fryauf-Bertschy H, Tyler RS, Kelsay DM, et al. Cochlear implant use by prelingually deafened children: the influences of age at implant and length of device use. J Speech Lang Hear Res. 1997; 40:183–199. [PubMed: 9113869]

Geers A, Brenner C, Davidson L. Factors associated with development of speech perception skills in children implanted by age five. Ear Hear. 2003; 24(suppl):24S–35S. [PubMed: 12612478]

Gorga MP, Neely ST, Dorn PA. Distortion product otoacoustic emission test performance for a priori criteria and for multifrequency audiometric standards. Ear Hear. 1999; 20:345–362. [PubMed: 10466570]

Gorga MP, Neely ST, Hoover BM, et al. Determining the upper limits of stimulation for auditory steady-state response measurements. Ear Hear. 2004; 25:302–307. [PubMed: 15179120]

Gorga MP, Neely ST, Ohlrich B, et al. From laboratory to clinic: a large scale study of distortion product otoacoustic emissions in ears with normal hearing and ears with hearing loss. Ear Hear. 1997; 18:440–455. [PubMed: 9416447]

Hart, B., Risley, TR. Meaningful differences in the everyday experience of young American children. John, MS., Brown, DK., editors. Baltimore, MD: Brookes; 1995.

Hoff E. The specificity of environmental influence: socioeconomic status affects early vocabulary development via maternal speech. Child Dev. 2003; 74:1368–1378. [PubMed: 14552403]

John MS, Brown DK, Muir PJ, et al. Recording auditory steady-state responses in young infants. Ear Hear. 2004; 25:539–553. [PubMed: 15604915]

Keenan RL, Boyan P. Cardiac arrest due to anesthesia. JAMA. 1985; 253:2373–2377. [PubMed: 3981764]

Keenan R, Shapiro J, Dawson K. Frequency of anesthetic cardiac arrests in infants: effect of pediatric anesthesiologists. J Clin Anesth. 1991; 3:433–437. [PubMed: 1760163]

Keenan RL, Shapiro JH, Kane FR, et al. Bradycardia during anesthesia in infants: an epidemiologic study. Anesthesiology. 1994; 80:976–982. [PubMed: 8017662]

Kirk, KI., Diefendorf, AO., Pisoni, DB., et al. Assessing speech perception in children. In: Mendel, LL., Danhauer, LJ., editors. Audiologic evaluation and management and speech perception assessment. San Diego: Singular Publishing Group Inc; 1997. p. 101-132.

Luts H, Desloovere C, Kumar A, et al. Objective assessment of frequency-specific hearing thresholds in babies. Int J Pediatr Otorhinolaryngol. 2004; 68:915–926. [PubMed: 15183583]

Moore JK, Guan YL. Cytoarchitectural and axonal maturation in human auditory cortex. J Assoc Res Otolaryngol. 2001; 2:297–311. [PubMed: 11833605]

Moore JM, Thompson G, Folsom RC. Auditory responsiveness of premature infants utilizing visual reinforcement audiometry (VRA). Ear Hear. 1992; 13:187–194. [PubMed: 1397759]

Moore JM, Wilson WR, Thompson G. Visual reinforcement of head-turn responses in infants under 12 months of age. J Speech Hear Disord. 1977; 42:328–334. [PubMed: 881813]

Morray JP. Anesthesia-related cardiac arrest in children: an update. Anesthesiol Clin North America. 2002; 20:1–28. [PubMed: 11892500]

Morray JP, Geiduscheck JM, Ramamoorthy C, et al. Anesthesia-related cardiac arrest in children: initial findings of the Pediatric Perioperative Cardiac Arrest (POCA) Registry. Anesthesiology. 2000; 93:6–14. [PubMed: 10861140]

Nikolopoulos TP, O'Donoghue GM, Archbold S. Age at implantation: its importance in pediatric cochlear implantation. Laryngoscope. 1999; 109:595–599. [PubMed: 10201747]

Norton SJ, Gorga MP, Widen JE, et al. Identification of neonatal hearing impairment: evaluation of transient evoked otoacoustic emission, distortion product otoacoustic emission, and auditory brain stem response test performance. Ear Hear. 2000; 21:508–528. [PubMed: 11059707]

Olsson GL, Hallen B. Cardiac arrest during anaesthesia. A computer-aided study in 250,543 anaesthetics. Acta Anaesthesiol Scand. 1988; 32:653–664. [PubMed: 3213390]

Pisoni DB, Cleary M. Measures of working memory span and verbal rehearsal speed in deaf children after cochlear implantation. Ear Hear. 2003; 24(suppl):106S–120S. [PubMed: 12612485]

Pisoni DB, Geers AE. Working memory in deaf children with cochlear implants: correlations between digit span and measures of spoken language processing. Ann Otol Rhinol Laryngol. 2000; 185(suppl):92–93.

Ponton CW, Eggermont JJ. Of kittens and kids: altered cortical maturation following profound deafness and cochlear implant use. Audiol Neurootol. 2001; 6:363–380. [PubMed: 11847464]

Ponton CW, Moore JK, Eggermont JJ. Prolonged deafness limits auditory system developmental plasticity: evidence from an evoked potentials study in children with co-chlear implants. Scand Audiol. 1999; 51(suppl):13–22.

Rackow H, Salanitre E, Green LT. Frequency of cardiac arrest associated with anesthesia in infants and children. Pediatrics. 1961; 28:697–704. [PubMed: 14489693]

Rance G, Beer DE, Cone-Wesson B, et al. Clinical findings for a group of infants and young children with auditory neuropathy. Ear Hear. 1999; 20:238–252. [PubMed: 10386850]

Rance G, Briggs RJ. Assessment of hearing in infants with moderate to profound impairment: the Melbourne experience with auditory steady-state evoked potential testing. Ann Otol Rhinol Laryngol. 2002; 189(suppl):22–28.

Rance G, Dowell RC, Rickards FW, et al. Steady-state evoked potential and behavioral hearing thresholds in a group of children with absent click-evoked auditory brain stem response. Ear Hear. 1998; 19:48–61. [PubMed: 9504272]

Raudenbush, SW., Bryk, AS., Congdon, R. HLM6: Hierarchical linear and nonlinear modeling. Chicago: Scientific Software International; 2004.

Robbins, AM. The Mr. Potato Head Task. Indianapolis. Indianapolis, IN: Indiana University School of Medicine; 1994.

Robbins AM, Kirk KI. Speech perception assessment and performance in pediatric cochlear implant users. Semin Hear. 1996; 17:353–369.

Rossetti, LM. The Rossetti Infant-Toddler Language Scale. East Moline, IL: LinguiSystems; 1990.

Sanborn KV, Castro J, Kuroda M, et al. Detection of intraoperative incidents by electronic scanning of computerized anesthesia records. Anesthesiology. 1996; 85:977–987. [PubMed: 8916813]

Sharma A, Dorman M, Spahr A. Rapid development of cortical auditory evoked potentials after early cochlear implantation. Neuroreport. 2002; 13:1365–1368. [PubMed: 12151804]

Staller, SJ., Beiter, AL., Brimacombe, JA. Children and multichannel cochlear implants. In: Cooper, H., editor. Practical aspects of audiology: cochlear implants: a practical guide. San Diego, CA: Singular Publishing Group, Inc; 1991.

Stallings LM, Svirsky MA, Gao SJ. Assessing the language abilities of pediatric cochlear implant users across a broad range of ages and performance abilities. Volta Rev. 2000; 102:215–235.

Svirsky MA. Language development in children with profound and prelingual hearing loss, without cochlear implants. Ann Otol, Rhinol Laryngol. 2000; 185(suppl):99–100.

Svirsky MA, Robbins AM, Kirk KI, et al. Language development in profoundly deaf children with cochlear implants. Psychol Sci. 2000; 11:153–158. [PubMed: 11273423]

Svirsky MA, Teoh SW, Neuburger H. Development of language and speech perception in congenitally, profoundly deaf children as a function of age at cochlear implantation. Audiol Neurootol. 2004; 9:224–233. [PubMed: 15205550]

Tay CLM, Tan GM, Ng SBA. Critical incidents in paediatric anaesthesia: an audit of 10,000 anaesthetics in Singapore. Paediatr Anaesth. 2001; 11:711–718. [PubMed: 11696149]

Tiret L, Nivoche Y, Hatton F, et al. Complications related to anaesthesia in infants and children: a prospective survey of 40,240 anaesthetics. Br J Anaesth. 1988; 61:263–269. [PubMed: 3179147]

Tyler RS, Fryauf-Bertschy H, Kelsay DM, et al. Speech perception by prelingually deaf children using cochlear implants. Otolaryngol Head Neck Surg. 1997; 117:180–187. [PubMed: 9334763]

Waltzman SB, Roland JT. Cochlear implantation in children younger than 12 months. Pediatrics. 2005; 116:e487–e493. [PubMed: 16199675]

Widen JE. Behavioral screening of high-risk infants using visual reinforcement audiometry. Semin Hear. 1990; 11:342–356.

Widen JE, Folsom RC, Cone-Wesson B, et al. Identification of neonatal hearing impairment: hearing status at 8 to 12 months corrected age using a visual reinforcement audiometry protocol. Ear Hear. 2000; 21:471–487. [PubMed: 11059705]

Zimmerman-Phillips, S., Osberger, M. Meaningful Auditory Integration Scale (IT-MAIS) for Infants and Toddlers. Sylmar, CA: Advanced Bionics Corp; 1997.

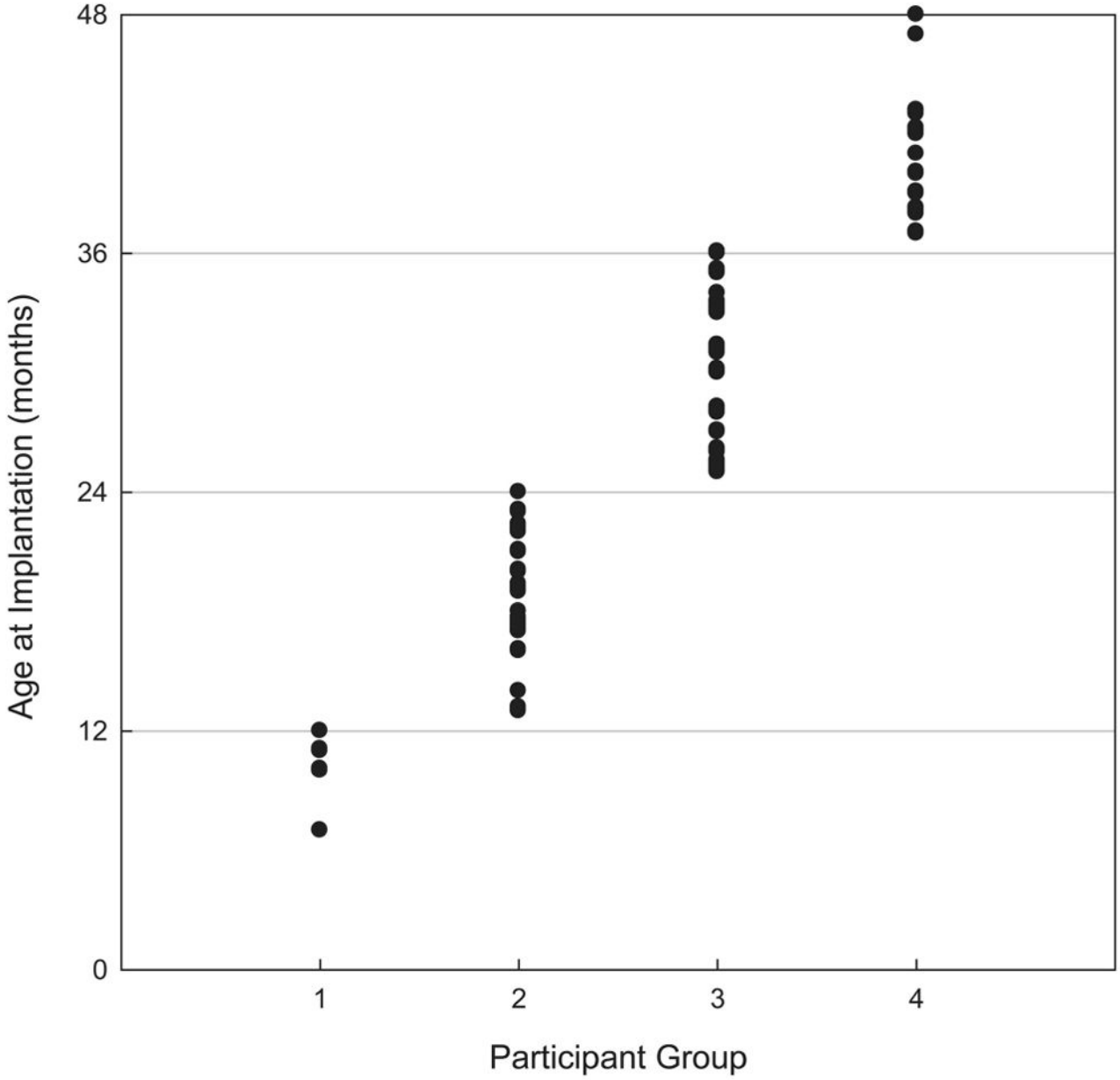

Fig. 1.
Scatterplot displaying age at implantation for each individual participant.

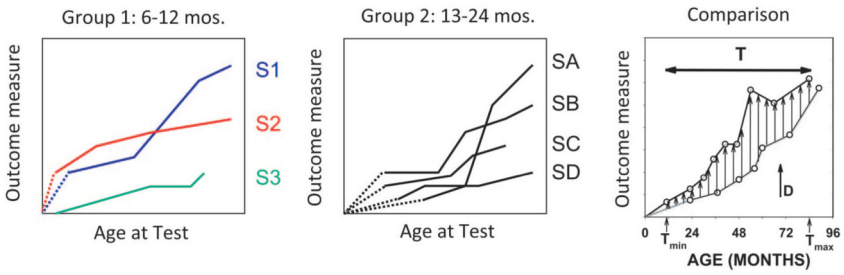

Fig. 2.

Left panel, Hypothetical curves for an outcome measure as a function of time in a group of three children implanted at 6 to 12 mos of age. Center panel, Similar to left panel, for a group of four children implanted at 13 to 24 mos. Right panel, Determination of the "developmental difference" between two children, one implanted at 11 mos and last tested at age 84 mos, and the other one implanted at 23 mos with a last testing age of 89 mos. The age range T, depicted by the horizontal arrow that extends from T_{min} to T_{max} (which are 11 and 84 mos in this example), is the age range over which comparison is done. The vertical arrows between the two curves represent the difference in outcome measure values for the two children at several ages between T_{min} and T_{max}. The average value of these arrows is the developmental difference between the two children and is depicted by the vertical arrow with the D label. With this method, differences between two developmental curves are boiled down to a single number. For a precise description of "developmental difference," see Eq. (1).

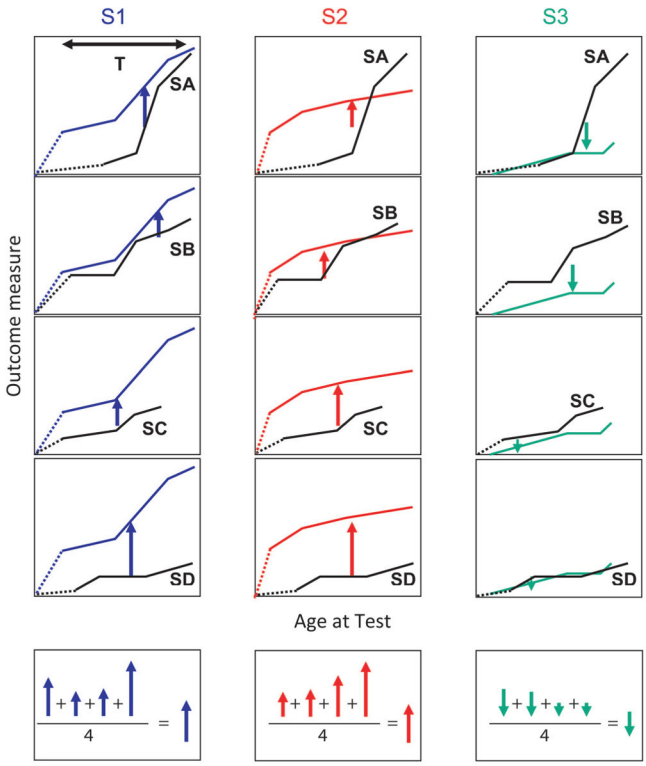

Fig. 3.

Calculation of developmental differences for the example data shown in the left and center panels of Figure 2. The left column of panels in this figure shows the average developmental differences between the first member of group 1 (in blue) and each member of group 2. The center and right columns of panels show the same for the second (in red) and third members (in green) of group 1, respectively. The arrows in the top four rows of panels indicate the developmental difference between the two curves in the corresponding panel, which are calculated with Eq. (1) and explained at a conceptual level in Figure 2. Arrows point up or down depending on whether the member of groups 1 or 2 has higher outcome measure values through the age comparison range. The bottom left panel shows the comparison between subject 1 and group 2, obtained by averaging the developmental differences between subject 1 and each individual member of group 2 [see Eq. (2) for a precise description]. The other bottom panels show the same type of comparison for subjects 2 and 3.

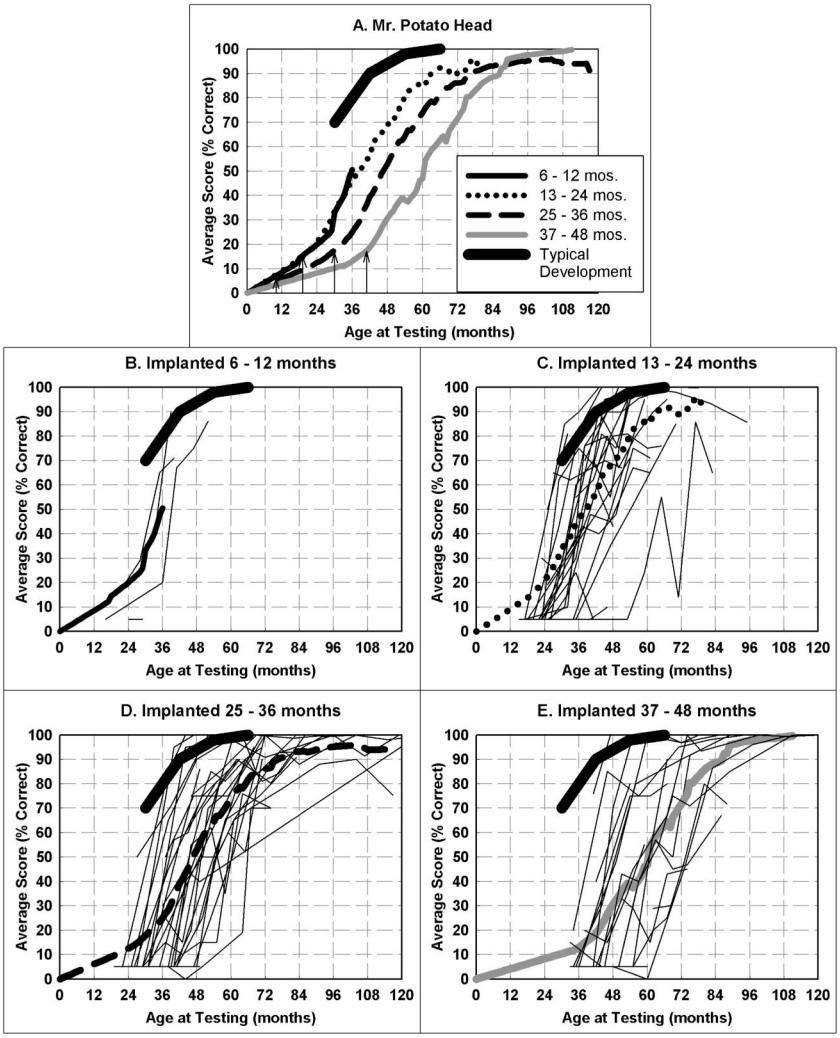

Fig. 4.

Panel A shows average word recognition scores on the Mr. Potato Head task for each age-at-implant group, as well as scores for typically developing normal-hearing children, as a function of age. Arrows indicate mean age at implantation for each group. Panels B to E show individual and group-averaged scores for each age-at-implant group.

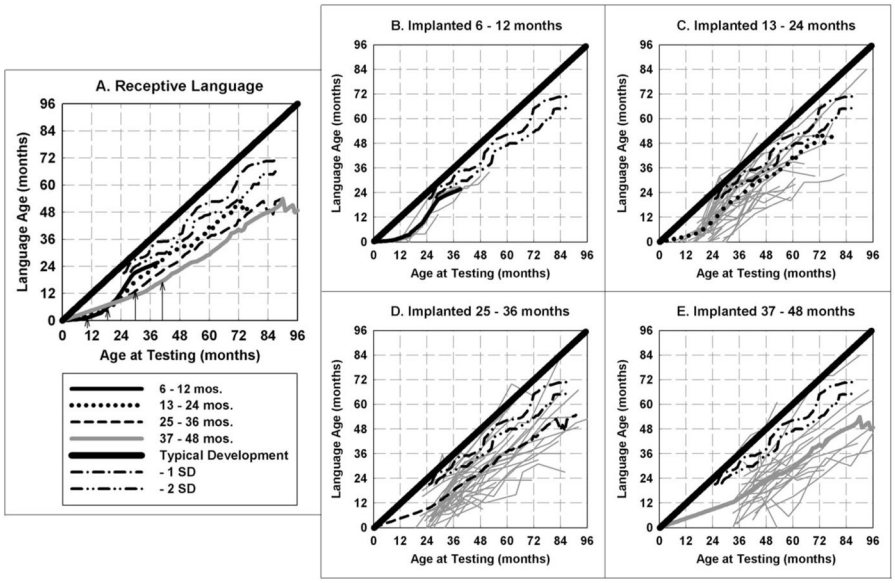

Fig. 5.

Panel A shows average age equivalent scores on the Receptive Language section of the RDLS for each age-at-implant group. Some of these scores are predicted values based on the MCDI parent questionnaire. The thick diagonal line shows average scores for the children used to obtain the test's norm. Two lines underneath the diagonal indicate 1 and 2 SDs below the mean for the same normative sample. Panels B to E show individual and group averaged scores for each age-at-implant group.

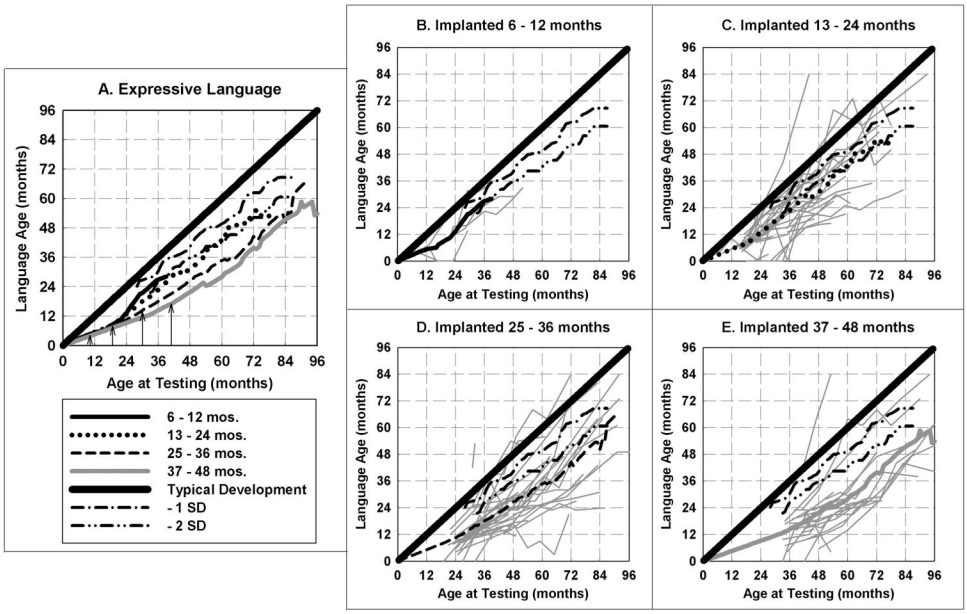

Fig. 6.

Expressive language scores, displayed following the same format as Figure 5.

TABLE 1

Demographic information for the four participant groups

Group, based on age at implant (mo)	n	Mean age at initial stimulation (mo)	Mean unaided best-ear PTA (dB HL)	Proportion oral comm mode (%)	Proportion female (%)	Mean estimated family income
Group 1 (6–12)	6	10.2	116.5 (3.1)	88.2	50.0	$55,639.83
Group 2 (13–24)	32	18.6	111.0 (7.5)	72.6	40.6	$51,287.78
Group 3 (25–36)	37	29.9	110.3 (8.8)	63.3	48.6	$58,122.49
Group 4 (37–48)	21	40.8	107.8 (8.0)	48.7	47.6	$59,814.43

SD values for unaided best-ear PTA are included in parentheses.

TABLE 2

Multiple linear regression analyses on output data from DTA

Outcome measure	Group comparison	Average developmental difference*	Stepwise	
			Individual earlier vs. avg. later implanted	Individual later vs. avg. earlier implanted
Word recognition	Group 1 vs. 2	−10.539	N/A	−1.134 (PTA, p = 0.002)
	Group 1 vs. 3	15.577†	N/A	N/A
	Group 2 vs. 3	17.971†	20.173† (PTA, p = 0.010)	N/A
	Group 3 vs. 4	18.364†	19.983† (PTA, p = 0.001)	14.330† (CM, p = 0.016)
Receptive language	Group 1 vs. 2	2.499†	1.670 (Income, p = 0.034)	N/A
	Group 1 vs. 3	7.204†	7.903† (Income, p = 0.023)	N/A
	Group 2 vs. 3	4.635†	N/A	4.877† (PTA, p = 0.040)
	Group 3 vs. 4	5.196†	6.060† (PTA, p = 0.025)	N/A
Expressive language	Group 1 vs. 2	1.727	N/A	N/A
	Group 1 vs. 3	4.592†	N/A	N/A
	Group 2 vs. 3	3.450†	N/A	N/A
	Group 3 vs. 4	5.239†	N/A	N/A

Covariates that significantly influenced Average Developmental Difference are indicated along with their p values.

*Average Developmental Difference was identical and reached the same level of significance regardless of if individuals in Group A were compared to the average of Group B or if individuals from Group B were compared to the average of Group A. Therefore, only one data column is displayed.

†p < 0.05.

N/A, Linear regression model accounted for no additional variance in Average Developmental Difference; DTA, developmental trajectory analysis.

TABLE 3

Hierarchical linear modeling statistics summary: least-squares estimates of significant and near-significant fixed effects

Outcome measure	Level-1 coefficient	Level-2 predictors	Coefficient	*p*
Word recognition *	Slope	Comm. mode	0.53425	0.001
		Income	0.00001	0.106
		PTA	0.01440	0.111
		Age at Implant	−0.00580	0.557
Receptive language	Slope	Age at Implant	−0.01099	<0.0001
		Income	0.00001	<0.0001
Expressive language	Slope	Age at Implant	−0.01099	0.026
		Income	−0.00001	<0.0001

* Scores above 90% that were achieved after reaching ceiling (90%) were removed from the analysis.