Development of Language and Speech Perception in Congenitally, Profoundly Deaf Children as a Function of Age at Cochlear Implantation

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Abstract
Like any other surgery requiring anesthesia, cochlear implantation in the first few years of life carries potential risks, which makes it important to assess the potential benefits. This study introduces a new method to assess the effect of age at implantation on cochlear implant outcomes: developmental trajectory analysis (DTA). DTA compares curves representing change in an outcome measure over time (i.e. developmental trajectories) for two groups of children that differ along a potentially important independent variable (e.g. age at intervention). This method was used to compare language development and speech perception outcomes in children who received cochlear implants in the second, third or fourth year of life. Within this range of age at implantation, it was found that implantation before the age of 2 resulted in speech perception and language advantages that were significant both from a statistical and a practical point of view. Additionally, the present results are consistent with the existence of a 'sensitive period' for language development, a gradual decline in language acquisition skills as a function of age.

Introduction
Profound deafness represents a major hindrance to speech communication, not only because it is very difficult for a person with profound deafness to understand speech without visual cues, but also because congenital profound deafness has a devastating effect on the development of spoken language and speech in children. Until the advent of cochlear implants (CIs), however, there existed no satisfactory treatment for profound deafness. A CI is an electronic device, part of which is surgically implanted into the cochlea and the remaining part worn externally. The CI functions as a sensory aid, converting mechanical sound energy into a coded electric stimulus that bypasses damaged or missing hair cells of the cochlea and directly stimulates remaining auditory neural elements. Most research on cochlear implantation in children has focused on the perception of speech [Oeberger et al., 1991; Staller et al., 1991; Fryauf-Bertschy et al., 1992; Geers and Brenner, 1994; Miyamoto et al., 1995]. However, CIs also provide children with critical auditory sensory input necessary for the development of speech and language production. Hearing loss at an early age, particularly before the onset of language, can have a deleterious effect on the development of speech and language in children. Research on various measures of articulation, speech intelligibility and expressive language has shown that these abilities improve after deaf children have
received CIs and continue to improve with increasing experience with the device [Dawson et al., 1995; El-Hakim et al., 2001a, b; Spencer et al., 1998, 2003; Svirsky et al., 2000a, b, c; Svirsky, 2000; Szagun, 2000, 2001; Tomblin et al., 2000]. In particular, our studies have shown that profoundly deaf children display a gap in their language development, but once they receive CIs they start developing language at a near-normal rate, and the developmental gap remains about the same size (measured in units of language age). These findings suggest that congenitally deaf children may be more expressive and receptive language skills at a normal pace and with only a negligible delay, if they only received CIs early enough in life. This speculation is consistent with research showing that children who receive CIs between 2 and 5 years of age tend to perceive speech much better than those who receive CIs later [Fryauf-Bertschy et al., 1992].

A prerequisite for receiving a CI early in life is the early identification of hearing loss, which has become possible on a large scale only recently in the USA, with the introduction of mandatory newborn hearing screening. Yoshihaga-Iwano and his group [Yoshihaga-Iwano, 1999; Yoshihaga-Iwano et al., 2000] report that the average age of identification of congenital hearing loss has been reduced from 24–30 months to an average of 2 months. This allows relatively early cochlear implantation, which is expected to be beneficial to language development for several reasons. First, earlier implanted children will have shorter lengths of sound deprivation and, conversely, longer auditory experience with a CI than their later implanted peers. Also, these children may benefit from early exposure to sound before the end of critical/sensitive periods for the development of speech and language [Hurford, 1991; Pickett and Stark, 1987; Ruben, 1986]. Indeed, studies have shown that exposure to a specific language in the first 6 months of life alters infants’ phonetic perception [Kuhl et al., 1992]. Previous work by Jusczyk et al. [Jusczyk and Houston, 1998; Jusczyk and Luce, 2002] has shown that by 9 months of age children can recognize their own names, respond appropriately to ‘mummy’ and ‘daddy’, begin segmenting words, retain information about frequently occurring words, and show language-specific preferences for prosodic cues. The latter skill is particularly important because prosody may be necessary for segmenting the acoustic stream into perceptual units. By the age of 10–12 months, sensitivity to nonnative contrasts begins to decline, and infants also appear to integrate different types of word segmentation cues. By 16 months of age, infants show an ability to segment vowel-initial words, and by 17 months they already show lexical competition effects that affect word learning.

However, there is some evidence that normal language learning and development occur only with early exposure to language. Conversely, when language exposure begins later in life, asymptomatic performance in the language declines [Newport, 1990]. This phenomenon has been named ‘sensitive period’ for language development. Bortfeld and Whitehurst [2001] provide a careful review listing 4 types of evidence that support the concept of biologically determined sensitive periods: ‘wild’ children who have been deprived of normal linguistic interaction during their first years of life; natural variation in the timing of exposure of deaf children to sign language; loss of perceptual or language learning capacity with age, and differences in cerebral localization of language processing for individuals exposed to languages at different ages. Although there is converging evidence coming from all these sources, Bortfeld and Whitehurst [2001] call this evidence ‘less than definitive’. Difficulties encountered by ‘wild’ children in learning language late in life may be due to deprivation of experiences that are unrelated to language, rather than to a sensitive period for language learning. The sign language data are compelling, but it is possible that sign languages are acquired in different ways than oral languages. Additionally, deaf children who were exposed to sign language late in life may have suffered social or cognitive consequences due to the lack of early input. Changes in perceptual or learning capacities may be due to changes in motivation or opportunity, rather than to a biological window that closes with age. Finally, differences in cerebral localization only demonstrate a neural rather than a behavioral sensitive period, unless they are accompanied by related differences on language tasks. Within this context, the examination of speech and language skills of congenitally deaf children who receive CIs at different ages provides an additional and independent type of evidence that, although imperfect, may be relevant for the investigation of sensitive periods in language development. Thus, study of the early implanted population is important clinically because these children may show substantial benefit in speech and spoken language outcomes, and it is important scientifically because it may provide information about sensitive periods in language development.

An additional reason to study the benefit of cochlear implantation in the first few years of life is that such early implantation may carry significant additional risks related to anesthetic complications [Young, 2002]. For example, a study found a higher incidence of bradycardia in
infants younger than 1 year who underwent noncardiac surgery (1.3%) than in children in the second, third or fourth years of life (0.98, 0.65 and 0.16%, respectively) [Keenan et al., 1994]. Bradycardia was associated with significant morbidity, including hypotension in 30%, asystole or ventricular fibrillation in 10% and death in 8% of the cases. If these numbers applied to cochlear implantation, there would be approximately 4 additional deaths for every 10000 children who are implanted at the age of 3 instead of at the age of 4, and about 2.6 additional deaths for every 10000 children who are implanted in the first year of life instead of the second, or in their second year instead of the third. On the other hand, numbers from the study cited above may overestimate the actual anesthesia risk for children undergoing cochlear implantation, because the numbers represent an average obtained from very diverse cases. Most surgeries in the study of Keenan et al. [1994] were elective, but some were emergency surgeries; most operations took less than 4 h, but some took more; a pediatric anesthesiologist was in charge only in about two thirds of the cases, and about 44% of the children in the study were in a high-risk class according to the American Society of Anesthesiology classification (ASA class 3, 4 or 5). In contrast, CI surgeries are elective, usually last less than 4 h and are almost always performed on healthy patients (ASA class 1). If a pediatric anesthesiologist is in charge, the risks may be further reduced for all age groups. A document from the American Academy of Pediatrics [Kass et al., 1996], after reviewing the literature up to 1990, suggests that 'after the first 4–5 months of life, age alone is not the major risk factor', but acknowledges that 'most studies of anesthetic risk are not stratified by age and ASA class, and therefore it is difficult to determine the precise anesthetic mortality rate for ASA class patients between 6 and 12 months of age'. In summary, even though anesthesia risks may be low, there is at least a possibility that early pediatric implantation may carry additional potential risks. Therefore, it becomes even more important to assess the potential benefits of early implantation.

The main goal of the present study was to compare the speech perception and language skills of congenitally deaf children who received CIs in the second, third or fourth year of life. This comparison was performed using a new method named developmental trajectory analysis (DTA). DTA examines the curves representing change in an outcome measure over time (i.e. developmental trajectories) for groups of children that differ along a potentially important independent variable such as age at intervention. Rather than comparing outcomes at a single point in time, or comparing only the slopes of developmental trajectories, DTA assesses the area under each developmental trajectory and provides an estimate of the average difference in outcome throughout the comparison period. A secondary goal of the study was to compare the language skills shown by profoundly deaf children with CIs to those of age-matched children. In addition to the clinical interest of these comparisons, the study of children implanted at different ages may provide important information about the existence of 'sensitive' periods for language development.

**Methods**

**Subjects**

The CI subjects were recruited from the clinical population at the Indiana University Medical Center and from the St. Joseph Institute for the Deaf in St. Louis, Mo., USA. Subjects were tested between 2 and 8 times using the tests listed below (see Outcome Measures). The first session always took place just prior to initial activation of the CI (between 1 and 3 months). When a subject was tested more than once, the testing sessions were at least 6 months apart. Participation in the study was offered to all monolingual English-speaking children implanted before the age of 5 years who used the SPEAK/ACE or CI2S strategies since initial device fitting, and who had no other handicapping conditions such as mental retardation or speech motor problems. More than 90% of the children who qualified for the research protocol actually participated in the study. All subjects were congenitally, profoundly deaf. They were divided into three groups according to age at implantation as shown in table 1, which also includes information concerning the amount of residual hearing and communication mode used by each group. Twelve children were implanted in the second year of life, 34 in the third year and 29 in the fourth year. Residual hearing was calculated by averaging hearing thresholds at 500, 1000 and 2000 Hz, measured in decibels. All children were in school settings that promoted the development of auditory/oral skills, with or without the use of signs. American sign language was not the primary mode of communication for any of the children. Some used oral communication (OC), while others used total communication (TC), which is the simultaneous use of oral English and signs. Note that TC involves the use of signs that reproduce English grammar and therefore all responses were monolingual, regardless of whether they were expressed manually or orally. A few children changed their communication mode in the course of the study. In those cases, the percentage of sessions that took place while the child used each method of communication was calculated. For example, a child who used TC in the first 2 testing sessions, was switched to OC and then tested 3 more times, would be considered a '60% OC user' for the purpose of calculating the proportion of OC and TC users within each age-at-implantation group. Although members of our research team do not provide aural rehabilitation and speech therapy, they do make recommendations for each child and remain in contact with schools to make sure that all children receive appropriate rehabilitation. Children in our study typically see a speech-language pathologist 2–3 times per week.

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Table 1. Demographic information of participants in the study

<table>
<thead>
<tr>
<th>Range of age at implantation months</th>
<th>Number of subjects</th>
<th>Mean age at implantation and SD, months</th>
<th>Unaided PTA (best ear mean and SD) dB</th>
<th>Range of unaided PTA (best ear) dB</th>
<th>Use of oral communication, %</th>
</tr>
</thead>
<tbody>
<tr>
<td>16-24</td>
<td>12</td>
<td>19.7 (1.9)</td>
<td>112 (5)</td>
<td>105-118</td>
<td>54</td>
</tr>
<tr>
<td>25-36</td>
<td>34</td>
<td>29.8 (3.4)</td>
<td>110 (9)</td>
<td>90-120</td>
<td>54</td>
</tr>
<tr>
<td>37-48</td>
<td>29</td>
<td>40.6 (2.5)</td>
<td>108 (7)</td>
<td>97-120</td>
<td>58</td>
</tr>
</tbody>
</table>

PTA = Pure-tone average. All were congenitally deaf, monolingual English-speaking children implanted before the age of 5 years, who had used the SPEAK/ACE or CIS strategies since initial device fitting and who had no other handicapping conditions.

Outcome Measures

The outcome measure used to assess language development was based on the Reynell Developmental Language Scales (RDLS-III) [Edwards et al., 1997] and the MacArthur Communicative Development Inventories [MCDI] [Fenson et al., 1993]. The RDLS assess expressive and receptive language separately. Expressive language scores were used in this study, in part because these scores are less likely than receptive language scores to be inflated by the use of iconic information when the test is administered using TC and also because the overall results were quite similar using both types of scores. 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 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-month-olds, and the MCDI/Words and Sentences is designed for 16- to 30-month-olds. The RDLS have been used extensively with deaf children (including CI users; see for example Bollard et al. [1999], Richter et al. [2002], Svinicki et al. [2000b], Vermeulen et al. [1999]) and are appropriate for a broad age range (from 1 to 8 years). Normative data are also available for more than 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.

Scores observed using the RDLS and 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 MCDI data. 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. The function that predicts RDLS expressive language scores as a function of subscores in the MCDI/Words and Gestures is the following:

\[
\text{REXP age equivalent } = 1.3386 + 2.0917 \times \text{WGlabeled} + 1.9359 \times \text{WName} + 0.0276 \times \text{WSwp} \text{rs} + 0.2793 \times \text{ca_mos},
\]

where WGlabeled indicates a child's ability to label items, WName a child's ability to respond to his name, WSwp.rs the number of words produced, and ca_mos is the child's chronological age in months. The predictive function using subscores from the MCDI/Words and Sentences is:

\[
\text{REXP age equivalent } = 13.7111 + 0.2942 \times \text{WSirw} \text{rs} + 0.0254 \times \text{WSwp} \text{rs} + 0.6518 \times \text{WScep} \text{rs},
\]

where WSirw.rs is the number of irregular words produced, WSwp.rs is the total number of words produced and WScpl.rs is the MCDI's measure of syntactic complexity. Adjusted R-squared values obtained in that study indicated that the predictive functions explained between 71% and 81% of the variance in RDLS scores.

Children were also assessed by the Mr. Potato Head task [Robbins, 1994], a modified open-set test of spoken word recognition. Mr. Potato Head is a toy with a plastic body with approximately 20 body parts (such as ears or nose) and accessories (such as hats or glasses). Children were asked to manipulate the toys in response to commands given in the auditory-only mode, e.g., "Put a hat on Mr. Potato Head". The children's responses were scored as the percentage of key words correctly identified. This test assesses the recognition (perception and understanding) of words denoting body parts, accessories and actions. Because the required response is an action instead of the spoken repetition of a sentence, results are not confounded by a subject's ability to speak intelligibly.

Data Analysis

Many methods are available to assess the effects of age at implantation. One possibility is to wait a number of years until all children reach a predetermined age and then assess differences. In the case of pediatric CI users, the most extensive and well-controlled study of this type has been carried out by Geers and her colleagues at CID, who studied a large group of deaf children with CIs aged 8-9 years [Geers and Brenner, 2003; Strube, 2003]. One problem with this approach is that it does not evaluate the impact of early implantation throughout the child's developmental trajectory (i.e., change over time). It is important to investigate whether late implanted children catch up with early implanted children at some point, but it is also important to evaluate whether one group had an advantage over the other during 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 ends up catching up with the earlier implanted group eventually. To use language age as an example, figure 1 shows 2 hypothetical develop-
Fig. 1. DTA compares developmental trajectories by calculating the average size of the difference between two curves (vertical arrow D). This is calculated as the integral of the difference between the two curves (the shaded area) divided by the comparison interval T.

mental trajectories, one for a child implanted shortly after 15 months and another implanted at 50 months. The regression lines for each curve are shown with thick dashed lines. Although the later implanted child almost achieves parity with the earlier implanted child around the age of 84 months, the earlier implanted child has had a functional communicative advantage for several years. Thus, all other things being equal, earlier implantation is better in this example. Speech perception can also be used as an example. Imagine two groups of children, one implanted at the age of 2 and the other implanted at the age of 4, both scoring 0% correct in tests of word recognition prior to implantation. Let us assume that the group of children implanted at 2 reach average word identification scores of 80% correct by the age of 3 and stay at about that level until the age of 8. Let us further assume that children implanted at 4 reach scores of 80% at the age of 5 and also stay at that level until the age of 8. Again, all other things being equal, implantation at 2 years of age is superior to implantation at 4 years in this hypothetical example, because the group of children implanted at 2 are able to understand 80% of the words they hear at the ages of 3 and 4, while the other group of children cannot understand any words at those ages. Even if the later implanted children do catch up later, the quality of life of the earlier implanted children is superior during an important part of their preschool years (ages 3 and 4). This reasoning is based on the premise that, all other things being equal, it is better to understand speech than being unable to understand it.

Other methods examine whether the rate of change in an outcome measure is affected by age at implantation. This is a type of analysis that has been used by Kirk et al. [2002] and other authors [Oserberger et al., 2002]. An interesting alternative method to analyze rates of growth in outcome measures after an intervention is binary partitioning analysis [El-Hakim et al., 2002]. However, it is possible for a late implanted child (or group) to have a higher rate of change after implantation than an earlier implanted child (or group) and yet be clearly worse off than that child, as in the example of figure 1. The gray dashed regression line corresponds to postimplantation data for the child implanted at 50 months and is steeper (has a greater rate of change) than the black dashed regression line corresponding to the child implanted at 15 months. Nevertheless, the earlier implanted child shows higher language scores at any given age up to 84 months than the late implanted child. Thus, although analyzing the rate of change after implantation can reveal important scientific and clinical information, it may not be the best way to determine the effect of age at implantation on the child's developmental trajectory.

In addition, the analyses described above are not optimal to determine the preferred age at implantation for the congenitally deaf population. Instead, a new methodology termed DTA is proposed. Let X and Y be two groups of children implanted at different age ranges (with n and m members in each group, respectively), and y(i) and x(i) the developmental trajectories for the individual children in each group, for the outcome measure of interest. For example, x(i) (j = 1 - n) may represent all the curves of children implanted between 12 and 24 months of age, and y(i) (i = 1 - m) may represent all the curves of children implanted between 25 and 36 months. Let Y(t) be the average curve in group Y. To express this relation in mathematical terms,

\[ Y(t) = \frac{1}{m} \sum_{i=1}^{m} y(i) \]

Then, the 'mean developmental difference' D between each member of group X (for j = 1 - n) and the average of group Y is calculated as follows:

\[ D_{XY} = \int [X(t) - Y(t)] dt / T \]

where the upper integration limit T is the maximum value for which both x(t) and Y(t) are defined. Thus, the developmental difference D_{XY} is the area between the developmental trajectory corresponding to member j of group X, and the average of group Y, which is then divided by the length of the integration domain. One intuitive interpretation for D is that it represents the average size of the difference between the two curves mentioned above, averaged over the whole analysis period. The developmental difference, D, at a particular age is depicted with a vertical arrow in the example shown in figure 1. Averaged over the whole analysis period, D becomes about 18 months of language age, although the difference between the two curves varies as a function of chronological age: it is as small as 0 (at ages of 0–12 months) or as large as 50 months of language age (at the chronological age of 54 months).

To test whether the developmental trajectories from X, as a group, are significantly different from the average curve of group Y, the following null hypothesis can be used: H0 = the set of development differences D_{XY} (j = 1 - n) is a sample taken from a normal distribution with a mean of zero.

This hypothesis can be easily tested using the Student distribution or (as an alternative preferred by many statisticians, particularly for small-sized samples) the Wilcoxon or exact permutation tests. DTA can also be used when the two groups to be compared, X and Y, differ in terms of confounding variables which are presumed to have
an effect on the outcome measure (such as residual hearing or communication mode). One way to address this problem is to calculate the ‘confounding variable differences’ $D_{ij} = D_i - D_j$, where $D_i$ is the value of the confounding variable for subject $i$, and $D_j$ is the average value of the confounding variable for group $j$. Then, instead of testing the null hypothesis listed above, a regression is performed (or multiple regression, if there is more than one confounding variable) of $C_j$ as a function of $D_{ij}$. In the example of a linear case, the regression equation would be $C_j = a + bD_{ij} + \mu$, and the null hypothesis would be $H_0: \mu = \text{the intercept of the regression function is zero}$ (or, in other words, the developmental differences are due exclusively to the effect of the confounding variables).

In the present case, table 1 shows that the three age-at-implantation groups did not differ significantly along the two potential confounds thought to be most influential in speech and language outcomes for pediatric CI users: residual hearing and communication mode. In fact, there was a very slight advantage for the two groups of children who were implanted later. Thus, if an advantage in language development and speech perception outcomes was found for the earlier implanted group in this study, the advantage could not possibly be due to the effect of residual hearing or communication mode. Given the small differences in the two confounding variables, differences among groups were assessed with the test approach rather than the multiple regression approach. All pairwise comparisons among the three age-at-implantation groups were carried out. Each comparison was conducted in both directions. For example, the developmental differences resulting from comparing each child implanted at 12–24 months to the average curve for children implanted at 25–36 months 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 25–36 months to the average curve for children implanted at 12–24 months. The most conservative of the two comparisons was selected to express the significance of the difference between the groups. These comparisons were carried out for both outcome measures, language age and speech perception scores.

Potential advantages of the proposed DTA analysis method include the following: no assumptions are made concerning the shape of developmental trajectories, all available data points can be used, missing data are handled easily, the method assesses the whole developmental trajectory rather than individual points and, finally, it has high face validity for the purpose of assessing the effect of age at implantation on outcome measures.

**Results**

The thin dark lines in the top panel of figure 2 show individual language data for children implanted between 12 and 24 months of age. Three normal-hearing reference curves are provided for comparison: the black diagonal line indicates the progress that would be expected of an average child with normal hearing, and the two gray curves represent 1 and 2 standard deviations below the mean for the normal-hearing population. A skill level that is 1 standard deviation below the mean is equivalent to being on the 16th percentile of the normal-hearing population. In other words, it represents language skills that are better than those of one sixth of the normal-hearing population of the same age, which is well within normal limits (although quite rare for children with congenital profound hearing impairment). In contrast, 2 standard deviations below the mean is a level that puts an individual in the second percentile of a normal distribution. Finally, the thick gray curve represents an average of all the individual curves (each individual curve is interpolated from the origin to the first recorded data point, just prior to implantation). Many children in this group had scores that were very close to the average values for children with normal hearing. One way to quantify this observation is to examine how many children implanted before the age of 2 showed near-normal expressive language skills, defining ‘near normal’ as obtaining scores within 1 standard deviation of the normal hearing norms, for at least 2 out of 3 consecutive testing intervals. Although available data are too preliminary to answer the question with any degree of certainty, some trends are emerging. So far, only 3 of the 12 congenitally deaf children implanted before their second birthday have been followed up beyond the age of 4, but 2 of them showed near-normal scores starting at the ages of 4 and 4.5, respectively, and the third was close to obtaining near-normal scores just before his/her fifth birthday. Additionally, 2 of the remaining 9 children have obtained near-normal scores at least once, before the age of 4. These results suggest that language scores for CI users get closer to average values from normal-hearing children as a function of age and, moreover, this happens both in absolute terms (using raw scores or age-equivalent scores) and in relative terms (using Z scores that represent the CI user’s performance compared to the normal-hearing population).

The middle panel of figure 2 shows data from children implanted at 25–36 months. Like in the group of children implanted at 12–24 months, there were some children in this group with near-normal development curves, but there were also several children who fell 2 standard deviations or more below the normal hearing mean. Finally, children implanted between 37 and 48 months (bottom panel of fig. 2) normally stayed below the −2 standard deviation curve, even after many years of CI use. Figure 3 shows the three average curves for the data from each group of children. The arrows indicate the average age at which each group received cochlear implants. It is interesting to note that before implantation the three curves are practically superimposed. After 19–20 months, which is the average age at implantation for children who

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received CIs between 12 and 24 months, the curve for that group starts to separate from the curves corresponding to the other two age-at-implantation groups. Similarly, the curves for the two later implanted groups start to separate around 29 months, which is the average age at implantation for children who received CIs between 25 and 36 months. At the latest data point, children in the three age-at-implantation groups show clearly different levels of language proficiency: those implanted at 12–24 months are, on average, close to 1 standard deviation below the normal hearing means; children implanted at 25–36 months are, on average, close to 2 standard deviations below the normal hearing means, and those implanted at 37–48 months are well below both benchmarks.

DTA results indicated that the average advantage for children implanted between 12 and 24 months (measured in units of language age) was 5.7 months with respect to children implanted at 25–36 months (p < 0.01). In other words, the average estimated language age for children in the first group was 5.7 months higher than for children in the second group, at the same chronological age. Children implanted at 37–48 months lagged behind those implanted at 25–36 months by 5.6 months (p < 0.05), and those implanted at 12–24 months by 10 months (p < 0.001).

Figure 4 shows average speech perception scores for the three age-at-implantation groups as well as comparison data obtained from normal-hearing children [Kirk et al., 1997; Robbins and Kirk, 1996]. The trends are the same as for the language development scores: prior to implantation, the developmental curves for the three groups are practically identical, and after implantation differences among groups start to emerge. Note that the earlier implanted group starts showing an advantage over

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Fig. 2. The thin lines in each panel show individual language development curves as a function of age, and the thick gray line is the average of all individual curves. Language skills were assessed with the expressive section of the RDLs and expressed as age-equivalent scores. If the RDLs could not be administered, language age was estimated based on results from the MCDI. Each panel shows data for a different age-at-implantation group, from top to bottom: 12–24 months, 25–36 months and 37–48 months. Three lines are provided as a reference to compare data from CI users to the normal-hearing population. The thick diagonal line shows the language development that would be expected of an average child with normal hearing, whereas the two thinner gray lines under the diagonal represent performance levels that are 1 and 2 standard deviations below the normal-hearing average, respectively.
Fig. 3. The three thick curves are the average language development curves for each age-at-implantation group (which were shown separately in each panel of Fig. 2). The normal-hearing comparison lines are the same as in figure 2. Note that the average curve for children implanted at 12–24 months reached the –1 standard deviation level (16th percentile for the normal-hearing population) at around 60 months of age, whereas the average curve for children implanted at 37–48 months was well below the –2 standard deviation (2nd percentile). The arrows indicate the average age at implantation for each group.

Fig. 4. Average curves for word scores in the Potato Head sentences test for each age-at-implantation group. Arrows indicate average ages at implantation. Children implanted at 12–24 months reached ceiling levels on this test about 1 year later than normal-hearing children, while children in the other age-at-implantation groups lagged behind even further, and it is still not clear whether they will ever reach ceiling scores as a group.

Discussion

These data support the hypothesis that implantation in the second year of life results in better speech perception and language development outcomes than later implantation. In this respect, results are consistent with previous studies that have found advantages in communicative outcomes for children who are implanted earlier in life [Fryauf-Bertschy et al., 1992]. The advantage for the earlier implanted children represents an effect that is both statistically significant and large in size. For example, based on the present data it may be possible to speculate that many children implanted at 12–24 months (perhaps a majority of them) will reach the age of 6 years and enter school with near-normal language skills (at least when those skills are assessed using the RDLS), whereas this is not happening for most children implanted later. Similarly, most children implanted at 12–24 months performed near the ceiling level on the Potato Head test at least a
year before the age of 6, while this is not true of children in the other groups. Instead of performing at ceiling levels at the age of 5, children implanted at 25–36 months only identify an average of 4 out of 5 key words in the relatively simple Potato Head test, and those implanted at 37–48 months only identify 3 words out of 5. These perceptual and language differences during the first few years of a child’s school experience may have a negative effect on learning, even if the perceptual and language differences tended to disappear by the age of 8 or 9, as the studies of Geers and Brenner [1994, 2003] would suggest. However, there is one important caveat that should be taken into account when interpreting the present language data or any other dataset obtained using standard norm-referenced tests: when a CI user achieves scores similar to those of normally developing children it does not mean that the CI user has ‘normal’ language, it only suggests that he or she has age-appropriate skills in the language tasks assessed by the test. Indeed, recent studies suggest that CI users may have more significant difficulties in developing certain aspects of grammar than in developing lexical skills [Svirskey et al., 2002; Szagun, 2000, 2001]. In any case, it seems clear that cochlear implantation in the second year of life rather than later has some advantages in terms of communicative skills, and these advantages probably outweigh any additional surgical risk.

In the present study, DTA helped compare the speech and language outcomes over time for children implanted at different ages. The Methods section listed several potential advantages of DTA, but perhaps the most important one is its ability to provide a reasonable estimate of the average difference between two groups of developmental curves without making any assumptions about the shape of those curves. This method may also be useful to compare effect sizes and significance in response to any clinical intervention (including those outside the fields of speech, language and hearing) when it is important to evaluate that intervention over an extended period of time.

One important aspect of the present results is the large intersubject variability. Even though the three age-at-implantation groups showed important differences, each group had at least a few outstanding performers. Thus, the results suggest that cochlear implantation before the age of 2 may be beneficial, but excellent results can be achieved at later ages as well. On the other hand, many children with CIs show language development curves that remain well below the –2 standard deviation line. This includes some children implanted in their second year of life, most of the children implanted in the third year and the vast majority of those implanted in the fourth year. Although children who are implanted later seem to develop speech perception and language skills at a lower pace than children who are implanted earlier, there are numerous individual exceptions to this trend. In consequence, the present results are consistent with the ‘sensitive period’ view [Johnson and Newport, 1993] that postulates a gradual decline in language acquisition skills as a function of age. These results are also consistent with studies of language development in German-speaking CI users [Szagun, 2001]. However, there is an important caveat when examining CI data for their potential relevance to the existence of sensitive periods: the auditory signal provided by a CI is less than optimal, it provides less information than the auditory signal received by children with normal hearing. Thus, it is at least possible that sensitive periods may exist for speech and language development when listeners are exposed to the impoverished signal provided by a CI, but not necessarily when exposed to a normal acoustic signal.

The present study falls short of a randomized double-blind study, which would provide more definite answers concerning the effect of age at implantation on speech and language development. However, conducting such a study would be at least questionable from an ethical point of view, given the available evidence that earlier cochlear implantation is beneficial. Instead, future studies may attempt to refine the present analyses by considering other potential confounding variables and by studying outcomes in children implanted in the first year of life. Additionally, other outcome measures should be considered in future work, including measures of the child’s ability to speak intelligibly and more detailed measures of language development that examine specific skills such as the use of grammar.

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References


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