Acta Oto-Laryngologica, 2009; 129: 361–366

ORIGINAL ARTICLE

Long-term follow-up of infants (4–11 months) fitted with cochlear implants

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Abstract

Conclusion: In this study the outcomes from several indices (Category of Auditory Performance, CAP; Peabody Picture Vocabulary Test (Revised), PPVT-R; Test of Reception of Grammar, TROG; and Speech Intelligibility Rating, SIR) in three groups of children with different ages at implantation (from 4 to 36 months) with a follow-up time from 4 to 9 years demonstrate that very early cochlear implantation (<11 months) provides normalization of audio-phonologic parameters with no complications. *Objectives:* The aim of the present study was to investigate the efficacy of cochlear implants (CIs) in infants who were implanted at <11 months of age versus children operated at later age (i.e. 12–36 months) and to document whether children who receive a CI below 11 months of age are able to achieve age-appropriate expected spoken language skills, at a follow-up time from 4 to 9 years. *Subjects and methods:* From November 1998 to November 2007, 185 children received CIs and 34 received auditory brainstem implants in our department. The present study focuses on 13 children implanted at ages younger than 12 months (4–11 months; mean, 8.2; SD = 2.4) and fitted with CIs between November 1998 and March 2004. To avoid bias these children were selected from a larger longitudinal cohort of pediatric CI recipients fitted with CIs because they all were implanted with the same cochlear device (Nucleus CI 24 M) during the same period. Postoperatively auditory abilities were evaluated at the latest follow-up, from 4 to 9 years after surgery, with CAP, PPVT-R, TROG, and SIR. The results obtained in this group of 13 children were compared with those obtained in two groups of children implanted at later ages (12–23 and 24–36 months, respectively). *Results:* No complication has been observed so far. The highest score of CAP function was achieved in all the three groups but at different intervals from CI activation as function of age at CI implantation. The rate of receptive language growth (PPVT-R) provides distinctive evidence that only the scores of the first group overlap the line of normal-hearing children, whereas the second and third group never reached the values of normal peers even after 9 years of CI use. TROG outcomes clearly indicate that only children from the first group (77%) are in the 76–100 percentile at 5 years follow-up. At 9 years follow-up, 100% of children in the first group, 38% in the second group, and 20% in the third group are in the 76–100 percentile. The SIR outcomes at the 5 years follow-up indicate that none of children was identified within the first two categories, only children from the third group (18%) were identified in category 3, all infants of the first group, 80% of group 2, and 63% of the third group were identified in category 5. At the 9 years follow-up, the number of children from the third group identified in category 3 was reduced to 10%, the second and third groups displayed a slightly higher percentage of children in category 5, but the difference from the values observed at the 5-year follow-up is not significant.

Keywords: *Cochlear implant, infants, children, auditory performances, cognitive effects*

Introduction

Several issues have a bearing on decisions about the beneficial effect of very early cochlear implantation for the development of age-appropriate spoken language in infants and children. It is well known that neural organization and/or structure related to speech perception/production are affected by the length of auditory deprivation [1–3] but the extent and potential reversibility of changes in the neural architecture are presently not completely known.

Likewise it is known that special opportunities do exist for preservation/restoration of the auditory system of deaf infants and preschool-age children and vanish for children who are just a few years older at time of implant surgery [4–12]. These studies provide evidence that by decreasing the age at implantation surgery better outcomes can be

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Presented at the CORLAS Meeting, Berlin, 24–27 August 2008.

(Received 12 September 2008; accepted 18 September 2008)

ISSN 0001-6489 print/ISSN 1651-2251 online
DOI: 10.1080/00016480802495453

obtained in terms of spoken language. Thus implantation should ideally occur not only early enough for normal language progress to be achieved but also before delays could be established. The implicit corollary from all these observations is that cochlear implants (CIs) should be fitted in infants/children as soon as a correct diagnosis of severe to profound hearing loss has been obtained. Indeed the significant advantage of early intervention in children with significant hearing loss was demonstrated by Apuzzo and Yoshinaga-Itano [13] in children identified and aided in the first 2 months of life who developed significantly better language than children identified and aided between 3 and 12 months of age.

Now the main questions are as follows. Is it realistic to expect that children who receive a CI early in life will be able to achieve age-appropriate spoken language abilities by the end of the preschool period? How early is early in terms of months or years for the deaf child to receive an implant for avoiding differences in language performance with his or her hearing age-matched peers?

Several studies [14–16] indicate that only some school-age children who received a CI before age 5 years are ‘closing the gap’ with their hearing peers in terms of spoken language production, verbal IQ, and academic achievement, while most of these children continue to exhibit delays for many years. Geers [9] detailed that only 43% of a nationwide sample of 8–9-year-old deaf children who received a CI between 24 and 35 months of age achieved combined speech and language skills within the average range for hearing children of that chronological age.

In a subsequent investigation, Nicholas and Geers [17] indicated that only children receiving a CI before age 24 months (i.e. between 12 and 16 months) can be expected to exhibit levels of spoken language competence that are on a par with hearing age-matched peers before they enter kindergarten, whereas those children implanted after 24 months of age do not catch up with hearing peers. Thus, the likelihood of achieving normal language in the preschool period decreases as age at implantation increases, and children fitted with CIs after 3 years of age may experience great difficulty in catching up with hearing age-mates. Thus the levels of spoken language competence, the breadth of vocabulary utilized, and the complexity of sentences expressed seem to be directly affected by the age at which a child is fitted with the CI.

Taken together these results suggest that further research is needed to document whether further decreasing age at implantation to below 12 months allows children to achieve age-appropriate expected spoken language skills and to provide evidence that the advantages of very early auditory stimulation persist into elementary and high school.

In a recent paper Colletti et al. [18] presented preliminary data on 10 children aged 4–11 months fitted with cochlear implants. The outcomes from that exploratory investigation indicated that surgery was uneventful, with no immediate or delayed complications and the indices used (CAP and babbling) suggested that early cochlear implantation tended to yield normalization of audio-phonologic parameters, with performance of children implanted very early being similar to that of their normally hearing peers. The positive impact of early implantation on babbling was clearly shown by the fact that the earlier the activation of the CI, the closer the results were to the outcomes of normally hearing children. The preliminary findings of that study were distinctly encouraging, but the small number of infants and the short follow-up were obvious limitations of that investigation.

In the present paper we report the follow-ups from 4 to 9 years up of three groups of children with different age at implantation (4–11, 12–23, and 24–36 months) fitted from November 1998 to March 2004 at the University of Verona, Italy and evaluated with a complete series of age-appropriate tests.

Subjects and methods

From November 1998 to November 2007, 185 children received CIs and 34 received auditory brainstem implants in our department. The present study includes 55 children, aged from 4 months to 3 years, fitted with a CI from November 1998 to March 2004, with a follow-up investigation from 4 to 9 years. Children were subdivided into three groups according to age at implantation: the first group comprised 13 infants aged 4–11 months (mean 8.2 months; SD = 2.4 months); the second group included 18 children aged 12–23 months (mean 15.8; SD = 1.8), and the third group comprised 22 children aged 24–36 months (mean 26.9; SD = 2.1).

To avoid bias these children were selected from a larger longitudinal cohort of pediatric CI recipients fitted with CIs, because they were all implanted with the same cochlear device (Nucleus CI 24 M) during the same period. To obtain a more homogeneous evaluation of the three groups, children with preliminary experience with hearing aids, with meningitis, and with associated disabilities were excluded from the present series.

Informed consent was obtained from the parents before surgery. Preimplantation, audiological assessments for these children included aided and unaided audiograms, auditory brainstem response (ABR), round window electrocochleography (RW ECoG), and round window electrical auditory brainstem response (RW EABR) [19] and indicated profound bilateral hearing loss in all cases.

Computed tomography (CT) scans and magnetic resonance imaging (MRI) showed normal inner ears and cochleovestibular nerves. Pediatric, neuropsychiatric, and genetic evaluations were performed.

The causes of deafness were genetic in 20, infective from cytomegalovirus in 9, from perinatal anoxia in 5, and unknown in 21 patients.

All infants were operated on using a transmastoid approach. The mean duration of surgery was approximately 45 min. Impedance measurements of electrodes, neural response telemetry (NRT), and electrically evoked auditory brainstem responses (EABR) recordings were performed intraoperatively in all patients to test the stimulating activity of each electrode. An electrophysiologic investigation was carried out at the end of surgery during suturing and continued immediately after surgery during awakening to reduce the length of the anesthesia. The average overall length of anesthesia was 75 min.

CIs were activated after a period of time ranging from 25 to 40 days post-surgery. The threshold level and maximum comfortable level of each electrode were first assessed, based on NRT and EABR outcomes obtained intraoperatively, to select the optimal electrode configuration.

Postoperatively all children were evaluated at the latest follow-up, from 4 to 9 years after surgery, with the following tests: Category of Auditory Performance (CAP) to examine auditory abilities; Peabody Picture Vocabulary Test (Revised) (PPVT-R) to test receptive language level; Reception of Grammar (TROG) to examine understanding of grammatical contrast in Italian; and SIR to measure the speech intelligibility of the implanted children. Statistical analyses were performed using the Wilcoxon Mann-Whitney test and the Pearson's chi-square tests.

Results

The median CAP scores were identical for the three groups over the first 6 months of follow-up. After the initial 6 months of CI use, children in the first group showed the most rapid increase in CAP, scoring 7 at the 24-month follow-up. Children in the second and third groups scored 7 at 36, and 42 months after surgery, with a delay of 12 and 18 months, respectively. With the Wilcoxon Mann-Whitney test the difference in the value of the delay in achieving CAP 7 between the first and second group and between the first and third group was statistically significant, with $p = 0.02$ and $p = 0.001$, respectively.

With the SIR test, 5 years after the activation, all the children in the first group (100%), 80% of the children in the second group, and 63% of the children in the third group had developed speech intelligible to the average listener (category 5 of the SIR scale). With the chi-square test the differences between the first and the second and between the first and the third group of children were statistically significant, with $p = 0.0081$ and $p = 0.003$, respectively. At 9 years of follow-up the percentage of children that reached category 5 in the second and third group was 83% and 69%, respectively. The difference in the values observed at the 5-year follow-up was not significant. The differences between the first and the second and between the first and the third group of children were statistically significant, with $p = 0.009$ and $p = 0.004$, respectively.

The first group exhibited normal development of receptive language with an overlap of the line of normal-hearing children, as assessed by PPVT-R, whereas children in the second and third groups showed slower progress, with an average lag of approximately 6 months and 1 year, respectively, when compared with the children of the first group and the normal-hearing population rate, at any chronological age. The second and third group never reached the values of normal peers even after 9 years of CI use. Children in the first group scored significantly better than those in the other age groups ($p = 0.008$ and $p = 0.0021$, respectively) according to the Wilcoxon-Mann Whitney test, at the various observed times.

TROG evaluations demonstrated that at 5 years from activation no child in the second and third group was above the 75th percentile, whereas 77% of children in the first group were among the 76th and 100th percentile of their normal-hearing peers. At 9-year follow-up the percentages increased to 100%, all the children in the first group, to 38% of the children in the second group, and to 20% of the children in the third group, respectively. The differences between the first and the second group ($p = 0.0019$) and the first and the third group ($p = 0.0039$) were statistically significant.

Discussion

There is a general agreement that early cochlear implantation leads to increased rates of language acquisition, as the children are still in the critical period for their development [1,3,12].

The likelihood of achieving normal language in the preschool period decreases as age at implantation increases, and children fitted with CIs at 3 years of age may experience great difficulty in catching up with hearing age-mates [17]. The levels of spoken language competence, the breadth of vocabulary utilized, and the complexity of sentences expressed seem to be directly affected by the age at which a child is fitted with the CI, so that when these children at 4–5 years of age approach kindergarten the expected language quotient scores of the very young CI recipients are well within the range documented for hearing age-mates [17]. To date, however, few published studies have indicated how early is early for normal development of communication in infants and children who receive implants.

The purpose of this study was to investigate the efficacy of CIs in infants who were implanted at <11 months of age versus children operated at later age (i.e. 12–36 months) and had a follow-up time from 4 to 9 years.

In a previous paper we presented preliminary data [18] on 10 children aged 4–11 months fitted with CIs. The outcomes from that exploratory investigation indicated that surgery was uneventful, with no immediate or delayed complications, and the indices used in that study (i.e. CAP and babbling) suggested that early cochlear implantation tended to yield normalization of audio-phonologic parameters. The outcomes of that study were distinctly encouraging but the small number of infants and the short follow-up were obvious limitations of that investigation.

In the present paper we extended the investigation on early cochlear implantation to a larger group of children, with a longer follow-up time, and with an extensive number of tests to evaluate audition, language, and speech skills (CAP, PPVT-R, TROG, SIR).

A review of the literature on early cochlear implantation in infants indicates that this is probably the first investigation on the long-term outcome of a large group of infants operated on at a very young age (from 4 to 11 months, with an average age of 8.2 months) and compared with two groups of children operated at later ages. To obtain a more homogeneous evaluation of the three groups, children with preliminary experience with hearing aids, with meningitis, and with associated disabilities were excluded from the present series.

All the children in the present series were implanted soon after the diagnosis of profound hearing loss had been obtained with electrophysiological measurements such as ECoG with RW gold ball electrode, RW ECoG [19]. This is a reliable technique to establish the auditory level in infants, furnishing useful indications for candidacy for CI. None of the children in each group had any hearing aid trial before implant, as it is well known that in an intervention program with hearing aids, children did not perform any better with their implants than children who underwent implantation at the same age but did not receive hearing before CI [7].

The first important observation relates to the safety of fitting a CI in infants as young as 4 months of age. The surgical technique is similar to that used in older children and factors requiring caution are the thickness of the skull with exposure of the dura. The main sources of concern have to do with the surgical and anesthesiologic risk, which may be augmented by factors such as preterm birth, cardiorespiratory disorders or low weight. The child's weight is more a concern than the age at birth. To date, we have not encountered any anesthesiologic or operative complications. A skilled team of surgeons and anesthetists is of paramount importance for reduction of operative times.

The second observation relates to the outcomes from several indices (CAP, PPVT-R, TROG, SIR) in three groups of children with different age at implantation (4–11, 12–23, and 24–36 months, respectively), with a follow-up time ranging from 4 to 9 years.

The average CAP function of age for the three groups of children indicates that the highest score (CAP 7) was achieved in all the three groups but at different intervals from CI activation and after 24, 36, and 42 months, respectively, for the first, second, and third groups. Thus if one considers only the highest score of the CAP testing as the target, very early implantation does not seem justified. However, taking into account the statistically significant delays in reaching CAP 7 in the second and third groups, the first group demonstrates a much shorter delay, which results in a longer exposure to acoustic experience of the order of 1 to about 2 years. The relevance of this delay and how this correlates with facilitating working memory should be in any case considered [17].

The rate of receptive language growth as assessed with PPVT-R provides distinctive evidence that only the children in the first group have scores overlapping the line of normal-hearing children, whereas children in the second and third groups never reached the values of normal children and the difference remained statistically significant even after 9 years of CI use. Performance on this measure is highly correlated with both receptive and expressive language skills in children with profound hearing loss [20] and comparisons of communication abilities performed in the present study among age-matched peers who differed in age at implantation revealed that children who were implanted very early (<11 months) had higher receptive and expressive language abilities than children who were implanted after that time.

TROG outcomes clearly indicate that only children from the first group (74%) are in the 76–100 percentile at 5 years follow-up. At the 9 years follow-up, 100% of children in the first group, 45% in the second group, and 20% in the third group are in the 76–100 percentile. Spoken language grammar acquisition in prelingually deaf children using CIs after 11 months of age at implantation was found to be considerably delayed. Improvement in children who received an implant under the age of 11 months was similar to normal-hearing peers, and this finding supports the trend toward device implantation at a younger age, if grammatical competence in spoken language is to be achieved. When deaf children wearing a CI catch up with their normal-hearing peers with regard to spoken language at preset intervals following implantation, it follows that the device enables them to use audition effectively.

The SIR outcomes at the 5-year follow-up indicate that none of the children was identified within the first two categories; only children from the third group (18%) were identified in category 3, all infants in the first group, 80% in the second group, and 60% in the third group were identified in category 5. At the 9-year follow-up the number of children from the third group identified in category 3 was reduced to 10%, the second and third groups displayed a slightly higher percentage of children in category 5, but the difference from the values observed at the 5-year follow-up is not significant. These results suggest that very early CI provides long-term communication benefit to profoundly deaf children.

As a corollary to these outcomes, related to perceptual and communicative parameters, none of the children from the present series is presently attending a school for the deaf and at 5-year follow-up 30% of the children from the second group and 60% of children from the third group, respectively, need itinerant teachers

Conclusion

The goal for a congenitally profoundly deaf child is to achieve age-appropriate spoken language in the shortest possible time-frame. In the present study we tried to answer the following questions. 1) How early is early in terms of months or years for a deaf child to receive an implant and to gain normal language development, without perceptive, linguistic, speech, cognitive or communicative delays compared with his/her hearing age-mates? 2) Is it realistic to expect that children who receive a CI very early in life will be able to achieve age-appropriate spoken language abilities by the end of the preschool period? 3) Are there significant surgical/anesthesiologic risks in fitting infants with a CI and are the risks outweighed by the benefits obtained in terms of audition, language, speech, and educational status?

Infants implanted between 4 and 11 months can be expected to exhibit levels of spoken language competence that are on a par with hearing age-mates much before they enter the (Italian) primary school, whereas children fitted with CIs after 12 months of age may experience some difficulties catching up with hearing age-mates.

The levels of spoken language competence, the breadth of receptive vocabulary, and the comprehension of complex sentences have been shown to be directly affected by the age at which a child is fitted with the CI. Since most of the children with CIs in this study suffered from congenital hearing loss, it is probable that their hearing impairment was already present when the cochlea was completed in utero and since the cochlea is normally completed by 20 weeks in utero the extent of auditory deprivation must be considered to start from that time. Furthermore, to reduce the length of auditory deprivation we believe that when a hearing aid is not indicated (severe to profound hearing loss) a CI should be implanted as soon as possible. This approach intends to promote a more automatic rather than 'therapized' spoken language development.

Taken together these results favor very early cochlear implantation and suggest that further research on documenting whether infants who receive a CI below 6 months of age are able to achieve age-appropriate expected spoken language skills even earlier than 5–6 years of age, and whether the advantages of very early auditory stimulation persist into middle/high school and in professional activities.

Declaration of interest: The author reports no conflicts of interest. The author alone is responsible for the content and writing of the paper.

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