Cochlear Implantation: New Frontiers in Children and Adults

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INTRODUCTION

“Cochlear implants (CI) deliver the ability to recognize speech to the profoundly deaf and are arguably the most effective neural prostheses ever developed”\(^1\).

Recent developments include implantation in children less than 1 year of age, hearing preservation techniques in patients with residual auditory function, bilateral implantation, studies on the economic impact of cochlear implantation and improvements of the implant technology.

At the present time, the highly impoverished electrical input provided to the auditory system by implants to interpret speech works very well mainly in subjects who have developed language before their deafness, patients with residual hearing or in children who receive their implant at a very young age. CIs in young children have shown dramatic results in restoring nearly normal levels of auditory function\(^2\)-\(^5\). In early development deaf children fall behind normally hearing children of the same age on auditory skills and language development, but show a normal rate of development once implanted. The potential negative consequences of later implantation are becoming clear\(^3\),\(^5\)-\(^10\). Theoretically, earlier sensory experience should
provide benefit in sensory development as well as in cross-modal and cognitive development. Sensory input must be provided early to take advantage of the developmental period of neural plasticity\textsuperscript{11,12}. Sininger et al.\textsuperscript{13}, showed that the age at fitting of amplification in children between 1 to 72 months has the largest influence on speech perception, speech production, and language outcomes. The present paper addresses issues of auditory, language and cognitive development as a function of age at implantation: specifically if implantation below 12 months of age is indeed beneficial. Sensory perception and environment exploration contribute to the development of cognition in children. Infants demonstrate an extraordinary ability in processing and integrating sensory experiences to form cross-modal associations between various forms of sensory stimuli\textsuperscript{14}. Since auditory experience begins before full term birth\textsuperscript{15-18}, auditory deprivation has already begun in congenitally deaf infants even before birth. Early CI intervention produces significant improvements in both audition and cognition\textsuperscript{19-24}. Only few authors have reported little difference in outcomes between a small sample of children implanted before 12 months of age and others implanted at later ages\textsuperscript{25}. Evidence supporting improved speech perception and speech production in children implanted under 12 months of age has grown
dramatically over the last 10 years$^{5,23,24,26-34}$. Similarly, absence of significant anesthetic and immediate surgical or postoperative major complications in this very young population is supported by several reports in the CI literature$^{23,26,27}$. The broad consensus that perioperative risks are reduced if anesthesia is administered by a pediatric anesthesiologist$^{38}$ has encouraged several centers worldwide to implant infants younger than 6 months$^{33}$. Up to date researches need to focus attention on the long-term safety and efficacy of children implanted below 6 months of age.

The development and diffusion of CIs have been limited mainly for economic reasons$^{39}$. At the present time, highly specialized hospitals performing CIs in Italy need to adapt their activity according to defined quotas of prostheses. The economic impact of CIs in children has been assessed in many countries, including the United Kingdom$^{40,41}$, United States$^{42}$, Germany$^{43}$ and France$^{39}$. All these studies demonstrated that CIs in profoundly deaf children have a positive effect on quality of life at reasonable direct costs and result in a net saving to society. However, healthcare financing conditions and settings are specific to each country, leading to significant differences in cost analysis. Furthermore, factors related to country demographics and social cohesion may also affect the impact of CI costs on the family. The social cost of CIs in infants has never been investigated.
and to date the economic impact of CIs in children in Italy has not been precisely assessed. Since CI in children below 12 months allows them to achieve age-appropriate expected spoken language skills, it may be also responsible for changes in the cost to society compared to implantation in children at later ages. The payers’ perspective it is the most relevant perspective in cost discussions. Medical, educational, and family costs are supposed to increase with age at implantation.

Since CI indications are expanding to patients with residual hearing another major concern about CI is the preservation of the residual cochlear function when performing surgery and after. The pathophysiology of hearing loss during and immediately after CI activation is largely unknown. Human temporal bone studies have helped to elucidate traumatic mechanisms of intracochlear electrode placement and optimize surgical cochleostomy placement. In recent years, the possibility of preserving residual hearing after CI has been documented by several authors. To minimize trauma to cochlear structures during CI, all manufacturers have focused their engineering efforts on designing and developing special flexible electrodes with reduced cross-sectional dimensions. It has also been suggested to perform ‘Soft CI surgery’ regardless of the amount of pre-operative residual hearing, to reduce cochlear trauma and improve
spiral ganglion cell survival, and, consequently, improve the long-term outcomes.

Pre-operative versus post-operative auditory threshold studies\textsuperscript{52-55} have clearly demonstrated the possible deleterious consequences of CI on residual hearing but have not provided clear evidence of the specific steps that correlate with the corresponding amount of loss. To this end, information on the trauma induced by the type of cochleostomy and of electrode insertion modalities should be gathered in real time, while surgery is ongoing, so that the surgeon can understand the causative manoeuvres and decide whether to modify the surgical procedure to minimize trauma to the cochlea accordingly. Today this can be pursued by utilizing a neurophysiological auditory intraoperative monitoring (NIM) technique that continuously records the ongoing cochlear activity elicited by acoustic stimuli.

Among the different NIM techniques, i.e. electrocochleography (ECoG), auditory brainstem response (ABR) and auditory steady-state response (ASSR), utilized during hearing preservation, ECoG can satisfy these needs properly, furnishing large amplitude potentials and allowing adequate representation of evoked potentials after a few sweeps.
ECoG monitoring for hearing preservation in CI has been demonstrated to be reliable in the animal model\textsuperscript{56} while ASSR has also been adopted in humans\textsuperscript{57}. Intraoperative ECoG during CI may be the best technique to provide useful online feedback to the surgeon to immediately modify surgical procedure, reduce damage to the cochlea and increases the prevalence of preservation of residual hearing.

The present thesis addresses the issue of cochlear implantation in very young children describing the long-term audiological, language and cognitive outcomes of the largest and youngest population of infants ever described in Literature. This population of children who had received a CI at a very young age has also been critically investigated in term of cost-effectiveness with a follow-up of 10 years. The results of these infants who underwent CI are compared with children implanted at later ages.

Along with the effort in decreasing the age of CI in children an intraoperative monitoring technique (ECoG) was adopted to determine in a group of adults the least traumatic electrode array insertion modality for preservation of residual hearing during CI. This real-time electrophysiological monitoring of auditory function could help the surgeon in appreciating potential damaging manoeuvres so as to minimize trauma to the cochlea and increase the understanding of
how subtle technical improvements can increase hearing preservation beyond their current levels.
Infants versus older children fitted with cochlear implants: Performance over 10 years

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ABSTRACT

Objectives: To investigate the efficacy of cochlear implants (CIs) in infants versus children operated at later age in term of spoken language skills and cognitive performances.

Method: The present prospective cohort study focuses on 19 children fitted with CIs between 2 and 11 months (X = 6.4 months, SD = 2.8 months). The results were compared with two groups of children implanted at 12–23 and 24–35 months. Auditory abilities were evaluated up to 10 years of CI use with: Category of Auditory Performance (CAP), Infant-Toddler Meaningful Auditory Integration Scale (IT-MAS), Peabody Picture Vocabulary Test (PPVT-R), Test of Reception of Grammar (TROG) and Speech Intelligibility Rating (SIR). Cognitive evaluation was performed using selected subclasses from the Griffiths Mental Development Scale (GMDS, 0–8 years of age) and Leiter International Performance Scale-Revised (UPS-R, 8–13 years of age).

Results: The infant group showed significantly better results at the CAP than the older children from 12 months to 36 months after surgery (p < .05). Infants' PPVT-R outcomes did not differ significantly from normal hearing children, whereas the older age groups never reached the values of normal hearing peers even after 10 years of CI use. TROG outcomes showed that infants developed significantly better grammar skills at 5 and 10 years of follow up (p < .001). Scores for the more complex subtests of the GMDS and UPS-R were significantly higher in youngest age group (p < .05).

Conclusion: This study demonstrates improved auditory, speech language and cognitive performances in children implanted below 12 months of age compared to children implanted later.

1. Introduction

Cochlear implant (CI) in young children have shown dramatic results in restoring nearly normal levels of auditory function [1–4]. In early development deaf children fall behind normally hearing children of the same age on auditory skills and language development, but show a normal rate of development once implanted. The potential negative consequences of later implantation are becoming clear [2,4–9]. Theoretically, earlier sensory experience should provide benefit in sensory development as well as in cross-modal and cognitive development. Sensory input must be provided early to take advantage of the developmental period of neural plasticity [10,11]. Simmer et al. [12] showed that the age at fitting of amplification in children between 1 and 72 months has the largest influence on speech perception, speech production, and language outcomes. The present paper addresses issues of auditory, language and cognitive development as a function of age at implantation: specifically if implantation below 12 months of age is indeed beneficial. Sensory perception and environment exploration contribute to the development of cognition in children. Infants demonstrate an extraordinary ability in processing and integrating sensory experiences to form cross-modal associations between various forms of sensory stimuli [13]. Since auditory experience begins before full term birth [14–17], auditory deprivation has already begun in congenitally deaf infants even before birth. Early CI intervention produces significant improvements in both audition and cognition [18–21], but it remains to be clarified whether additional improvements result from implantation below one year of age.

The present study expands previous investigations [22,23] in term of number of children below 12 months (19 infants), range of language skills measured, cognitive development and duration of the follow-up (10 years of CI use). The population of infants in the present study has the lowest mean age (6.4 months) with the longest follow-up described to date.
2. Methods

From 1998 to 2008, 243 children were implanted by the present surgeon (VC) in Verona and elsewhere. The present study compares outcome measures from the 73 children who met the following inclusion criteria: (1) implanted under 3 years old, (2) congenital deafness, (3) no prior hearing experience (including hearing aid use), (4) etiology not Measles, (5) no other nonauditory disabilities, (6) normal inner ears and cochleovesicular nerves, (7) nucleus implant, and (8) no device failures. These children had follow up times of three months to 10 years.

Pre-implantation audiological assessments were performed in all children and included neonatal auditory screening using otoacoustic emissions, which led to a suspicion of profound hearing loss. Subsequently, auditory brainstem recording (ABR), round window electrocochleography, electrically evoked round window ABR and behavioral (visual reinforcement) or conditioned play audiometry confirmed bilateral deafness [24]. All children received pre-operative radiological investigations. Computed tomography scans and magnetic resonance imaging showed normal inner ears and cochleovesicular nerves in all subjects. Pediatric, neuropsychiatric, and genetic evaluations were also performed. Children with additional nonauditory disabilities diagnosed by the pediatrician and/or the neuropsychiatrist or deafened from meningitis were excluded.

CI was suggested to all children as soon as a proper diagnosis was achieved. Children came to our Department at different ages and were submitted with parental consent to CI as soon as protocols for surgery had been completed.

For the purpose of comparison all children included had the same implant device (Nucleus CI 24MX) and were congenitally deaf with no prior hearing experience (including no experience with prior use of hearing aids).

All children were operated on using a posterior tympanotomy approach by the same surgeon (VC). The mean duration of surgery was approximately 45 min. As described before [23], impedance measurements of electrodes, neural response telemetry (NRT), and electrically evoked ABR (EAABR) recordings were performed intraoperatively in all patients to test the stimulating activity of each electrode. All CIs were activated after a period of around 30 days post-surgery. The threshold level and maximum comfortable level of each electrode were first assessed, based on intraoperative NRT and EAABR measures, to select the optimal electrode configuration.

Children were subdivided in 3 groups according to age at implantation: the first group comprised 19 infants aged 2 to 11 months (mean 6.4 months; SD = 2.8 months), the second group included 21 children aged 12–23 months (mean 19.3; SD = 3.8) and the third group incorporated 33 children aged 24–35 months (mean 30.1; SD = 5.9). The numbers of children in each group at each follow-up test interval are presented in Table 1. The causes of deafness were genetic in 27, infective from cytomegalovirus in 12, from perinatal anoxia in 6, and unknown in 28 patients. Informed consent was obtained from the parents before surgery.

All children's families used spoken Italian as their primary communication method, and all the participants attended an identical post-implantation rehabilitation program, with individualized intensive auditory training, conversation and speech stimulation.

Postoperatively, all children were evaluated at the latest follow-up, from three months to 10 years from activation, with the following tests: Category of Auditory Performance (CAP) [25] and the Infant-Toddler Meaningful Auditory Integration Scale (ITMAIS) [26,27] to examine auditory abilities; Peabody Picture Vocabulary Test (Revised 3rd Edition) (PPVT-R) [28] to test receptive language level; the Test of Reception of Grammar (TROG) [29] to examine understanding of grammatical contrast in Italian; Speech Intelligibility Rating (SIR) [30] to measure the speech intelligibility of the implanted children.

The Griffiths Mental Developmental Scale (GMDS) is a test instrument administered to measure motor maturity and development, ability to cope with routine situations, auditory and speech functions, hand and finger motor mobility and eye and hand coordination, body consciousness, physical activity, and memory. In order to provide measures of non-verbal-cognitive function in children from 0 to 8 years three separate subscales of the GMDS were administered: locomotor, eye and hand coordination and performance. The GMDS revisions of 1987 [31] and 1996 [32] were chosen to longitudinally evaluate these children with the same version of the GMDS since the study began in 1998.

The Leiter International Performance Scale-Revised (LIPS-R) [33] test battery has been used to evaluate the non-verbal cognitive effects of CIs on children from 8 years of age. Since children are not expected to complete all 20 subtests, in this study, the analysis of the LIPS-R was based on the following subscales: figure ground and form completion for visual/spatial attention, sequential order and repeated patterns for fluid reasoning. The two scales (GMDS and LIPS-R, adopted in this study, were chosen because the administration of their subtests can be achieved through non-verbal instructions in a very accessible and enjoyable way. All children attempted the same subtests.

Statistical analysis was performed using the Wilcoxon Mann–Whitney test, Pearson’s Chi square test and Kruskal–Wallis test, as appropriate.

Ethics approval was obtained from the University of Verona Ethics Committees.

3. Results

No statistically differences between groups emerged in terms of sex distribution (p > .05). The rate of minor peri-operative complications was extremely low since we could only identify one case of wound seroma in the 12–23 month group and a case of wound infection in the 24–35 month group that were both treated conservatively. No anaesthesiological or major surgical complications such as flag breakdown were observed.

No significant differences emerged among the three groups in terms of CAP median scores within the first six-months of follow-up (Fig. 1). According to the Kruskal–Wallis test, the infant group
showed significantly better results than the older children at the 12 month and 16 month post-op test times \((p < .05)\). All groups of children achieved a median CAP score of 7 by 42 months after CI. The IT-MAIS is a structured parental report on the auditory listening behavior of their children. Normative data on the IT-MAIS from normal-hearing infants has been published [27] and it has been used to evaluate auditory progress in infants with CIs [2]. Fig. 2 presents the comparison of the IT-MAIS results at 1, 2, 3, 4 and 5 years post-activations for the three groups. The dashed line presents the normative data from the MAIS on normal hearing children from Kishon-Rabin et al. [27]. The unfilled square and circle present results at one year follow up from Robbins et al. [2].

The SIR test measures normal-hearing listener’s ability to recognize the speech of the child. Fig. 3 shows the results of the SIR test as a function of time since activation for the three implant groups. Five years after initial activation, all the children of the 2–11 month group (100%), 67% of the children of the 12–23 month group and 61% of the children of the 24–35 month group developed speech intelligible to the average listener (Category 5 of the SIR scale). The Chi Square Test showed significant differences between the 2–11 and the 12–23 month groups and between the 2–11 and the 24–35 month group of children, with \(p = .020\) and \(p = .009\), respectively. At 10 years of follow-up, the differences between the 2–11 and the 12–23 month group and between the 2–11 and the 24–35 month group of children were statistically significant, with \(p = .049\) and \(p = .038\), respectively.

On vocabulary development (PPVT-R) the 2–11 months group exhibited progress in receptive language very close to normal hearing children whose development is represented in Fig. 4 by the dashed line. Children in the 2–11 month group scored significantly better than those in the other age groups (\(p = .00001\) and \(p < .0001\), respectively) according to the Wilkons-Mann Whitney test, at the 10 year follow-up.

Grammar development scores on the TROG demonstrated that at five years from activation no child of the 12–23 and 24–35 month group was above the 75th percentile, whereas 77% of children of the 2–11 month group were above the 75th percentile of their normal-hearing peers (Fig. 5). The difference between the 2–11 month group and the others was highly significant \((p < .0001)\). At the 10 year follow-up the percentages increased to 100% for the children of the 2–11 month group who were above the 75th percentile, to 38% of the children of the 12–23 month group and to 19% of the children of the 24–35 month group, respectively. The difference between the 2–11 month and the 12–23 month group \((p = .0001)\) and between the 2–11 and the 24–35 month group \((p < .0001)\) were statistically significant.
The baseline results of all subscales of the GMDS showed no statistical differences between age groups. Scores of two of the three subtests (eye and hand coordination, performance) of the GMDS increased significantly at the 5 year point compared to baseline in all age groups \((p < .05)\). When comparing the performance mean scores of the infants (101–12) with the 12–23 (91–13) and 24–35 month group (88–8) children, the differences were statistically significant with \(p = .046\) and \(p = .0065\) respectively (Fig. 6). No statistically significant differences were observed for the other two subtests at 5 years among different age groups.

Statistically significant improvements in non-verbal cognitive function with the LIPS-R were found at the 10 year follow-up between the 2–11 and 24–35 month group at the form completion \((p = .0472)\), sequential order \((p = .0325)\) and repeated pattern \((p = .0160)\) subscales (Fig. 7). When comparing the youngest group with the 12–23 months children, statistically significant difference were found for the sequential order \((p = .0469)\) and repeated pattern \((p = .0440)\) subscales. No significant differences emerged between the two older groups of children for all subtests.

4. Discussion

Does early cochlear implantation restore sufficient auditory experience to overcome the negative effects of early deprivation on auditory, language and cognitive performance? Does implantation at ages under 12 months provide additional benefits compared to
implantation at older ages? To date published research on early implantation presents a conflicting message. Holt and Svirsky [34] conclude that there is no additional benefit in performance based on a small number of children implanted under 12 months of age. However, Colletti et al. [22] showed a clear advantage in CAP scores and babbling measures in 10 infants implanted before 12 months. Dettman et al. [4] also showed clear advantages in early implantation based on results from 19 children implanted under 12 months of age. Colletti [23] demonstrated that very early cochlear implantation (below 12 months of age) provides normalization of audio-phonologic development with no complications. A recent meta-analysis concluded that evidence of improved performance on auditory perception/speech production outcomes is limited for children implanted below 12 months [35].

When children are implanted later, the delays in the development of auditory performance could represent significant challenges for the development of working memory and general cognitive development [8,36,37]. Indeed, auditory development begins even before full term birth, as it is known that hearing begins early in intrauterine life. The newborn and even the fetus not only can hear relatively well, but also they are capable of distinguishing their mother's heartbeat and voice from others [14,38] and respond to changes in musical notes [16]. Other sensorimotor and cognitive development also rely on auditory development and can be seriously delayed the longer implantation is delayed. Indeed, some developmental trajectories have a biological window that closes if the necessary elements are not available within the "critical period" of development.

The infant population of the present study is the youngest described in the literature with a mean age: 6.4 months (range: 2–11 months; SD = 2.8 months) and with the longest follow-up (10 years). Waitzman et al. [39] and Valencia et al. [40] presented data from children implanted at a mean age of 9.6 months (range: 7–11) and 9.2 months (range: 6.7–11.7) months, respectively. Holt and Svirsky [5] evaluated six children with a mean age of 10.2 months (range: 6–12) followed for up to 5 years. More recently Roland et al. [41] reported data on 50 infants with a mean age of 9.1 months (range: 5–11) followed for up to 7 years. On all auditory and speech tests the youngest group showed superior performance to results from children implanted later. The children implanted below 12 months of age developed auditory capabilities faster (CAP), produced more intelligible speech earlier (SIR), developed language at normal rates and levels (PPVT) and developed grammar skills earlier than children implanted after 12 months of age. This superior performance persisted out to 10 years of follow-up. These data show strong evidence that earlier implantation results in faster development and these children continue to out-perform children implanted later.

Furthermore, the additional sensory input provided by the CIs clearly supports non-auditory cognitive development. The infant group showed significantly increased results on the GMDS performance subtest scores compared to the older children. This finding might be ascribed to the higher demand in terms of sensory input integration to complete the performance subscale task. Early additional auditory verbal and non-verbal stimuli provided by the CIs may offer the infant the chance of developing a more complex and effective learning strategy in a very "critical period" of their development. The activation of the auditory channel enriches the children's sensory stimulation [21] and brings the level of attention to a more sustained level on a wider range of stimuli. On the other hand, the locomotor subscales showed no significant differences as a function of CI fitting age. This subscale evaluates a "lower order" cognitive function compared to the performance subscale. It confirms the role of early auditory stimulation in building complex cognitive function. In view of the results of the GMDS subscales at 5 years post-implantation, children were tested again at 10 years with the LIPS-R to compare the long-term cognitive outcomes. Data from several subsheets of the LIPS-R showed that the infants were able to achieve higher scores on non-verbal cognitive tests. The sequential order and repeated patterns items on fluid reasoning showed the highest improvement in implanted infants compared to older children. Both tests require the "higher" ability to understand the relationship between stimuli and generate rules governing them. Despite the small number of subjects tested, the outcomes of the GMDS and LIPS-R underline the positive effect of early implantation in complex non-verbal cognitive functions. Similar results were recently described in children fitted with the auditory brainstem implant [37]. These findings support the hypothesis that early auditory stimulation might play a fundamental role in the development of higher cognitive functions where multisensory integration is essential. Thus, delays in the onset of hearing can delay aspects of cognitive development.

In conclusion, the present results clearly show better auditory reception, speech production and language development in children implanted younger than 12 months of age than in children implanted later. While all implanted children made excellent progress on all tasks, those implanted under 12 months of age made the gains faster and achieved higher levels of asymptotic performance.

It is important to note that a highly specialized pediatric team of experts is critical for obtaining the best outcomes in infants with CIs. In addition to experienced pediatric surgeons and anesthesiologists, the team should include an experienced pediatric audiologist and pediatric neuroradiologist to achieve the proper diagnoses, treatment and rehabilitation. The risks of cochlear implantation under 12 months of age are minimal in the hands of experienced pediatric surgeons and anesthesiologists [22,39,42]. Restoration of hearing in infants by cochlear implantation shows beneficial effects: auditory, language and cognitive development and should be undertaken as soon as a diagnosis of profound deafness can be confirmed.

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Ethical approval

All authors declare that they made substantial contributions to the intellectual content of the paper and they finally approved it for submission.

All Authors declare that there is no one else who fulfils the criteria but has not been included as an author.

Conflict of interest statement

All authors declare that they have no conflicts of interest.

References


INTRODUCTION

Cochlear implants (CIs) have provided hearing for many deaf children and are the most effective neural prostheses ever developed. The goal for a congenitally profoundly deaf child is to achieve age-appropriate spoken language in the shortest possible time frame. Studies have shown more rapid auditory and cognitive development in early implanted children, and the results demonstrate the safety of CI fitting in children younger than 12 months of age. Other authors have reported little difference in outcomes between a small sample of children implanted before 12 months of age and others implanted at later ages. The development and diffusion of CIs have been limited mainly for economic reasons.

The economic impact of CIs in children has been assessed in many countries, including the United Kingdom, United States, Germany, and France. All these studies demonstrated that CIs in profoundly deaf children have a positive effect on quality of life at reasonable direct costs and result in a net savings to society. However, healthcare financing conditions and settings are specific to each country, leading to significant differences in cost analyses. Furthermore, factors related to country demographics and social cohesion may also affect the impact of CI costs on the family. The social cost of CIs in infants has never been investigated, and to date the economic impact of CIs in children in Italy has not been precisely assessed.

The aims of the present study were to assess whether very early implantation in congenitally deaf and prelingually deafened infants allows them to achieve age-appropriate expected spoken language skills and to determine whether fitting of a CI in children younger than 12 months of age is responsible for changes in the cost to society compared to implantation in children at later ages. The payers’ perspective was chosen because it is the most relevant perspective in cost discussions. The study was designed to test the hypothesis that medical, educational, and family costs increase with age at implantation. Furthermore, CI costs were calculated based on outcome equivalence, comparing cost per developmental vocabulary year at different ages of implantation.
The results of follow-up to the age of 10 years, both in terms of social costs related to the CI and receptive language level, using the Peabody Picture Vocabulary Test-Revised (PPVT-R), are presented in infants implanted younger than 12 months of age and are compared with results obtained in the three groups of children implanted at later ages (12–23, 24–35, and 72–83 months).

MATERIALS AND METHODS

The present study population consisted of 68 children, ages from 2 to 83 months, fitted with a CI in our department from November 1998 to February 2008. All children were followed up at least to the chronological age of 10 years. For the purposes of comparison, all children recruited had the same implant device (Nuclleo® array, Cochlear Ltd., Sydney, Australia) and were congenitally deaf and prelingually deafened infants. Children were also excluded if they were deafened as a result of meningitis, were not Italian native speakers, or presented additional nonauditory disabilities.

The children were subdivided into four groups according to age at implantation: the 2- to 11-month group comprised 11 infants, the 12- to 33-month group 13 children, the 24- to 35-month group 19 children, and the 72- to 83-month group 25 children.

Informed consent was obtained from the parents before surgery. Preimplantation audiological assessments for all children included aided and unaided audiograms, auditory brain stem responses, round window electrocochleography and round window electrical auditory brain stem responses, 17 indi-cated profound bilateral hearing loss in all cases. Computed tomography scans and magnetic resonance imaging showed normal inner ears and cochleovestibular nerves. Pediatric, neuropsychiatric, and genetic evaluations were performed. The causes of deafness were genetic in 27, due to cytomegalovirus infection in 10, due to perinatal anoxia in five, and unknown in 26 patients.

CI was suggested for all children as soon as a proper diagnosis was achieved. Children came to our department at different ages and were submitted to CI with parental consent as soon as protocols for surgery were completed.

All infants were operated on using a transmastoid transfa-novascular approach by the same surgeon (V.C.). Full insertion of the electrode array was obtained in all subjects. CIs were activated after a period of time ranging from 25 to 40 days following surgery. All electrodes were active in all subjects. The threshold level and maximum comfort level of each electrode were first assessed based on neural response telemetry, and electrically evoked auditory brainstem response outcomes were obtained intraoperatively to select the optimal electrode configuration.

Postoperatively, all children were evaluated with follow-up to 10 years of age using the PPVT-R 17 to test their receptive language level.

The study of social costs of CIs in children was conducted retrospectively. Comprehensive data for direct and indirect costs of CI were obtained from parent questionnaires; existing national healthcare and educational system; and Verona Ear, Nose, and Throat Department databases and retail prices for materials used (hearing aids and CI batteries). The healthcare system databases of the Verona Hospital contained information about costs of preoperative assessment, hospitalization, surgery plus implantation (Italian Diagnostic Related Groups (DRGs)), implant failure, and public speech therapy rehabilitation up to the 10th year of age in each group of children.

<p>| TABLE I. Demographic Data From the Four Populations of Prelingually Deaf Infants Fitted With Cochlear Implants |
|---|---|---|---|</p>
<table>
<thead>
<tr>
<th>Groups</th>
<th>No. of Subjects</th>
<th>Sex</th>
<th>Age at Implantation, Median (Interquartile Range)</th>
</tr>
</thead>
<tbody>
<tr>
<td>2-11 mo</td>
<td>11</td>
<td>6 M, 6 F</td>
<td>6 (4–9)</td>
</tr>
<tr>
<td>12-23 mo</td>
<td>13</td>
<td>8 M, 7 F</td>
<td>14 (12.5–15.5)</td>
</tr>
<tr>
<td>24-35 mo</td>
<td>19</td>
<td>11 M, 8 F</td>
<td>24 (24–26)</td>
</tr>
<tr>
<td>72-83 mo</td>
<td>25</td>
<td>11 M, 14 F</td>
<td>74 (72–78)</td>
</tr>
</tbody>
</table>

M = male; F = female.

Parent questionnaires were one of the most important data sources and were developed ad hoc to investigate mainly the educational-rehabilitative costs and the expenses directly sustained by the family before and after implantation. Parent questionnaires covered details of costs for initial assessment and audiometric follow-up before implantation, hearing aids and their maintenance, private speech therapy and educational support, CI usage, checkups, travel, and parents’ days off work. Regarding time off work, parents were asked to estimate the number of days off work per year due to the hearing loss of their children. The loss of income estimation was based on the gross annual salary of each parent.

Regarding the cost of public educational support at school, parents were asked to record on the questionnaire the exact number of hours their children had with a support teacher per week and the number of children the teacher was shared with. This additional public educational cost was estimated on the basis of the number of hours divided by the number of support teachers per student and the mean national cost per hour of a support teacher. Intangible costs (e.g., pain and suffering) and changes in future earnings for children implanted at different ages were not estimated in the present investigation.

The baseline year for all costs was 1998. A discount rate of 3% was applied. Costs limited to the first year of care were not discounted. Costs were expressed in Euros (1E = 0.5725$) and 1E = $1.386 on January 31, 2010). Statistical analysis was performed using the Kruskal-Wallis test followed by Dunn’s post hoc test.

Ethical approval was obtained from the University of Verona Ethics Committee. Informed consent was obtained from all parents.

RESULTS

Demographic data and mean age at implantation for all children included in the present investigation are reported in Table I. All subjects in the three younger implantation groups were full-time users of the CIs, whereas one subject in the oldest implantation group was a nonuser after 4 years. No device failures were observed in any of the children, and no revision surgery was performed in any of them. The rate of minor perioperative complications was extremely low. One case of wound seroma in the 12- to 23-month group and one case of wound infection in the 24- to 35-month group were identified. Both were treated conservatively. No anesthesiological or major surgical complications such as flap breakdown were observed.

All parents completed the ad hoc questionnaire on CI-related costs. The mean costs to society for a prelin-gually deaf child up to 10 years of age implanted at

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different ages are presented in Table II. The costs related to the deafness and CIs were divided into three subcategories: healthcare system, educational, and family costs. The 2- to 11-month group demonstrated the lowest total cost over the first 10 years of life. In particular, family costs played an important role in the increased expense when CI fitting was delayed.

The total net savings to society in the four groups of children ranged from around 21,000 € in the two younger classes to more than 35,000 € when comparing the youngest infants against children implanted after 6 years of age. Decreasing the age of implantation from 6 years to 24 to 35 months achieves a reduction of around 3% in the total cost to society. With a further lowering of the age of implantation, the percentage of savings increases to 9% and 22%, respectively, in the groups of children implanted between 12 to 23 months and those implanted younger than 12 months of age. When comparing the total cost to society of CI among groups, statistically significant differences were observed (P = .0013; Kruskal-Wallis test). Dunn’s post hoc test indicated that significantly higher costs emerged between the youngest group and the two older groups (P < .01).

The Italian healthcare system registers costs of preoperative assessment, hospitalization, surgery plus implantation (Italian DRGs) with a fixed amount of money independent of implantation age. On analyzing the medical costs in detail (Fig. 1), the increased cost of CI maintenance in infants due to earlier implantation is partially offset by the absence of expenses for audiometric follow-up, hearing aids, and speech therapy performed prior to implantation. No statistically significant differences in healthcare system costs were observed between the four groups of children (P = .0692; Kruskal-Wallis test). The Italian educational system offers every deaf child the same amount of rehabilitation benefit independently of how he or she performs at school (Fig. 2). The major factor in determining differences between groups is the number of hours of support teachers offer. The Italian educational system generally provides a minimum of 6 hours weekly for any deaf child, and the youngest children showed a tendency to share a support teacher with other disabled students, leading to a savings in educator costs. Educational costs showed a highly significant difference between groups (P < .0001; Kruskal-Wallis test). Statistically significant differences between the 2- to 11-month group and older children emerged compared to the groups implanted at 24 to 35 months (P < .05; Dunn’s post hoc test) and 72 to 83 months (P < .001; Dunn’s post hoc test).

In contrast to medical and educational costs, family costs show a significant increase with age of implantation (Fig. 3), and this is mainly due to days off work for parents, travel expenses before implantation, and the additional cost of private speech therapy and hearing aids. The cost of CI batteries increased with the time of CI usage. Statistically significant differences in family costs were observed (P = .0002; Kruskal-Wallis test) when comparing the 2- to 11-month group against all other groups.

**TABLE II.** Mean Costs (in Euros) to Society for Prelingually Deaf Children Up to 10 Years of Age Implanted at Different Ages.

<table>
<thead>
<tr>
<th>Age Group</th>
<th>Health System</th>
<th>Educational</th>
<th>Family</th>
<th>Total Cost to Society</th>
<th>Net Savings to Society</th>
</tr>
</thead>
<tbody>
<tr>
<td>2–11 Months</td>
<td>79,587 (±6,410)</td>
<td>22,874 (±2,303)</td>
<td>23,773 (±10,374)</td>
<td>126,034 (±11,945)</td>
<td>—</td>
</tr>
<tr>
<td>12–23 Months</td>
<td>76,563 (±11,178)</td>
<td>24,047 (±2,590)</td>
<td>46,461 (±18,519)</td>
<td>147,070 (±21,669)</td>
<td>21,036 (±11,945)</td>
</tr>
<tr>
<td>24–35 Months</td>
<td>77,615 (±8,845)</td>
<td>26,185 (±1,963)</td>
<td>53,226 (±19,464)</td>
<td>157,026 (±22,610)</td>
<td>30,992 (±18,519)</td>
</tr>
<tr>
<td>72–83 Months</td>
<td>71,448 (±8,022)</td>
<td>24,047 (±2,717)</td>
<td>61,547 (±21,870)</td>
<td>161,242 (±24,621)</td>
<td>35,208 (±22,610)</td>
</tr>
</tbody>
</table>

*Fig. 1. Details of health system costs in prelingually deaf children implanted at different ages. *Kruskal-Wallis test. CI = cochlear implant.*

*Fig. 2. Details of educational costs in prelingually deaf children implanted at different ages. *Dunn’s post hoc test. *Kruskal-Wallis test.*
three groups of older children ($P < .05$, 12–23 months; $P < .01$, 24–35 months and $P < .001$, 72–83 months; Dunn’s post hoc test).

All children completed the PPVT-R test at 10 years of age (Fig. 4). Four patients in the 24- to 35-month group and 15 in the oldest group of children dropped out after completing the PPVT-R test at 2 years. Regarding vocabulary development (PPVT-R), the 2- to 11-month group exhibited slightly lower development of receptive language (vocabulary age of 9.5 years at 10 years of age) versus normal hearing children. Children in the 12- to 23-month and 24- to 35-month groups improved substantially in vocabulary, but at 10 years of age still scored significantly lower than normal hearing children of the same age. Children implanted after 6 years of age showed a substantial delay in vocabulary development, with a vocabulary age of 5.8 years at 10 years of age. Outcomes of the PPVT-R were significantly different in the various age groups ($P < .0001$; Kruskal-Wallis test). Children in the 2- to 11-month group scored significantly better than older age groups ($P < .05$, $P < .01$, and $P < .001$, respectively) according to Dunn’s post hoc test.

DISCUSSION

It is now clear that CIs in adults and children, but the possible additional economic benefit of very early implantation in infants has not been reported and is not known. A few decades ago, the first year of life was believed to be of little interest as far as the acquisition of language is concerned. Today, early speech development and language acquisition are seen as a continuous process starting in intrauterine life, continuing in the brainstem in very early childhood, and finally well into late childhood, in the cerebral cortex. The development of the auditory system, and in particular the early development of speech perception, is therefore strictly dependent on acoustic stimulation and on access to relevant acoustic and linguistic information very early in life.11

The total cost of CI fitting at 10 years of age was divided by the real age of the children (10 years) (Fig. 5) and the vocabulary age from the PPVT-R (Fig. 6) to obtain the cost per year of age and the cost per performance per year. The second calculation represents the costs per effective vocabulary age year. When comparing these data, a larger increase could be observed in the cost of gaining one vocabulary year between the youngest group and the other children (Table III and Fig. 6), whereas lower differences between groups emerged when comparing the cost per year of age.

![Fig. 3. Details of family costs in prelingually deaf children implanted at different ages. CI = cochlear implant. Dunn’s post hoc test. *Kruskal-Wallis test.](image)

![Fig. 4. Mean chronological age at the Peabody Picture Vocabulary Test-Revised (PPVT-R). *Kruskal-Wallis test. Dunn’s post hoc test.](image)

![Fig. 5. Costs of prelingually deaf children implanted at different ages per year of chronological age.](image)

![Fig. 6. Costs of prelingually deaf children implanted at different ages per year of vocabulary age at the Peabody Picture Vocabulary Test-Revised (PPVT-R).](image)

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It is well recognized that profoundly hearing-impaired infants must be identified and treated with CIs very early in their lives to improve their chances of joining hearing children in mainstream education and social life. If appropriate surgical modifications are adopted, infants as young as 6 months or even younger can be safely implanted.1,5,13 Very early implantation minimizes language delays, allowing age-equivalent language development. Access to sound facilitates the acquisition of rapid word-learning skills, and the development of these skills correlates with later vocabulary levels as represented by the PPVT-R.

The present study indicates that a significant net savings to society is achieved by decreasing the age of implantation to younger than 12 months of age. In the light of these outcomes, many costs and services provided prior to implantation (hearing aids and their maintenance, speech therapy, educational costs, days off work, and travel expenses for parents) emerge as substantially cost-ineffective. Although medical costs undergo a slight cost reduction by delaying the age of implantation, the costs for education, and in particular for the family, increase dramatically for children implanted at older ages. The net savings to society over a 10-year time period for an infant implanted at an age younger than 12 months is approximately €21,000, as against children implanted between 12 to 23 months, and rises to more than €35,000 when the age of implantation is over 6 years. Thus, implanting children in the first years of life minimizes not only language delays but also the overall costs to families. Furthermore, all subjects implanted before 36 months were full-time users of the implant, whereas one subject implanted after 6 years became a nonuser. This finding supports the view that especially age of implantation, educational considerations, and family support may play an important role in becoming a nonuser.17

Highly specialized teams of pediatric experts, including experienced pediatric audiologists, neuroradiologists, surgeons, and anesthesiologists are needed to achieve proper diagnoses, treatment, and rehabilitation in infants fitted with CIs. The risks of cochlear implantation in children younger than 12 months of age are minimal in the hands of experienced surgeons and anesthesiologists.1,5

Nevertheless, when studying a pediatric population retrospectively, a recall bias could frequently cause overestimation of utility gains by parents. In view of this important limitation, the PPVT-R was prospectively administered in the present study because it gives an objective and comprehensive evaluation of children’s language development. At the age of 10 years infants implanted younger than 12 months of age may reach a vocabulary age of 9.5 years, whereas comparable children implanted at 6 years of age reach a vocabulary age of only 5.8 years. The substantial difference observed in the cost for gaining 1 year of vocabulary age on the PPVT-R between the youngest group and older children supports the efficacy of early implantation in terms both of outcomes and the net savings to society.

Comparison with similar studies from different countries is not easy due to specific healthcare financing conditions, educational settings, type of costs (direct and indirect), and period of time evaluated. Cost analyses performed in France and Germany apparently showed approximately similar costs, whereas studies conducted in the United States and United Kingdom demonstrated higher costs.

The small number of subjects under 12 months of age and the cost-outcome analysis performed only up to the 10th year of age are the major limitations of the present study. However, performing CI surgery in children younger than 12 months of age was not a universally accepted procedure in 1998 at the beginning of the present study. At the time of writing the number of infants fitted with CIs younger than 12 months is 32.

An on-going study with extended data on a larger number of infants, including those fitted with CIs younger than 6 months of age, seems to corroborate the present study. However, performing CI surgery in children younger than 12 months of age was a nonuser.17

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**CONCLUSION**

The present study provides two important indications, namely that the improvement in receptive language levels over time and the overall costs are strictly related to the age of implantation. In particular, the cost for gaining 1 year of vocabulary age in children is inversely related to the age at implantation.

**BIBLIOGRAPHY**


Cochlear Implants in Children Younger Than 6 Months

Liliana Colletti, PhD1, Marco Mandala, MD1, and Vittorio Colletti, MD1

Evidence supporting improved speech perception and speech production in children implanted younger than 12 months has grown dramatically over the past 10 years. Similarly, absence of significant anesthetic and immediate surgical or postoperative major complications in this very young population is supported by several reports in the cochlear implant (CI) literature. The broad consensus that perioperative risks are reduced if anesthesia is administered by a pediatric anesthesiologist has encouraged several centers worldwide to implant infants younger than 6 months.

The aim of the present study was to supplement previous investigations focusing attention on the long-term safety and efficacy of children implanted younger than 6 months and expanding the range of auditory-based performance.

Materials and Methods

Patients

Between November 1998 and June 2011, 386 children were implanted at the Ear, Nose, and Throat (ENT) Department of Verona and elsewhere by the senior author (VC). The present study is focused on a group of 12 infants aged 2 to 6 months (group 1), 9 infants aged 7 to 12 months (group 2), 11 children aged 13 to 18 months (group 3), and 13 children aged 19 to 24 months (group 4) all identified with profound bilateral hearing loss and fitted with a unilateral CI (Cochlear Nucleus Series, Cochlear Ltd, Sydney, Australia). None of the children in the present series had any hearing trials before surgery, and none were using sign language either pre- or postoperatively. All children in groups older than 12 months at implantation had their thresholds confirmed by behavioral audiometry.

Keywords

cochlear implant, younger than 6 months, infants, safety, complication, outcome, speech perception, speech production, language outcomes

Received September 3, 2011; revised February 16, 2012; accepted February 17, 2012.

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All children were followed longitudinally and completed the 48-month follow-up period. A control group of 20 chil-
dren with normal hearing and matched with the CI recipi-
ents for chronological age was also investigated. All children’s families used spoken Italian as their first lan-
guage, and all participants attended an identical postimplant auditory rehabilitation program.

**Preimplant Measures**

Preimplant audiologic and radiologic assessments were per-
formed in all children following a personal protocol used to determine CI and hearing aid candidacy in children
who failed neonatal screening. This included otomicro-
scopy, tympanometry and acoustic reflex thresholds, and
click auditory brainstem response (ABR) threshold assess-
ment. If there were no measurable ABR thresholds, round
window electrocochleography (RW ECoG) was performed
using click stimuli. If the threshold was lower than 75 dB hearing level (HL), then we referred the infants to an
audiologist for hearing aid fitting and ABR follow-up at 6
to 9 months. If the threshold deteriorated, then the infant
returned to us for further evaluation. If the child had no
cochlear microphonics and no compound action potentials
with logon/tone-burst RW ECoG at 500, 750, and 1000 Hz,
he or she was evaluated radiologically with computed tomo-
graphy and magnetic resonance imaging and revaluated by
a pediatric neurologist. If the imaging studies excluded a
severe malformation of the cochlea and cochlear nerve defi-
ciency, then the severely to profoundly deaf child may have
been a candidate for CI. We did not perform a hearing aid
trial in these cases because, in our experience, the use of
hearing aids delays the provision of auditory stimulation in
infants without any acoustically induced electrical cochlear
activity. In the present study, pediatric and neuropsychiatric
evaluations excluded children with additional disabilities and
subjects deafened by meningitis. This protocol was vali-
dated on 45 children in whom it was possible to perform
behavioral pure-tone audiometry 1 to 3 years after the elec-
trophysiological testing. The use of logon/tone-burst RW
ECoG reduces the percentage of potential CI candidates by
approximately 25%, based on ABR findings.

Children came to our department at different ages and
received CIs with parental informed consent as soon as an
accurate diagnosis was obtained and preoperative surgical
protocols had been completed. In 18 children younger than
6 months, CI was delayed by approximately 9 to 15 months
because of parental concern.

**Surgical Technique**

The surgical technique has been detailed in a previous
study. A well-trained pediatric anesthesiologist adminis-
tered intraoperative anesthesia and followed the infants
to 2 and 12 months at the intensive care unit for
approximately 6 hours after surgery. To avoid protrusion
and prevent device migration, the receiver-stimulator was
completely placed in a large bony seat in all children and
tightened down with 3-O PDS tie-down sutures.

**Intraoperative Measures and Device Fitting**

Intraoperative measures and device fitting have been
detailed in previous studies. All programming was per-
formed by an audiologist in the presence of a rehabilitation
therapist and using a combination of electrophysiological
information and behavioral responses.

**Auditory-Based Communication Measures**

The effect of age at CI fitting on auditory-based perfor-
mance was assessed at regular intervals in children starting
at 2, 3, and 4 years of device use. This battery of tests does
not rely on parental or caregiver questionnaires or reports;
rather, it is based on our team’s direct observations of behav-
ioral performance using established methods and test
materials.

Outcome measures included speech perception (Category 
of Auditory Performance [CAP]), receptive language
development (Test di Valutazione del Linguaggio, livello pre-scolare [TVL]), receptive vocabulary (Peabody Picture
Vocabulary Test–Revised), and speech production (Fanzago
Vocabulary Test, PFLI [commonly known as the Bortolini test], video
recording analysis, and International Phonetic Alphabet
transcription).

Because we previously reported a ceiling effect using the
CAP at 42 months’ follow-up in children fitted between 2
and 24 months, at the last follow-up (48 months of CI
experience), we used the CAP II (NEAP [Nottingham Early
Assessment Package]; The Ear Foundation, Nottingham,
UK), which introduces 2 new categories: CAP 8 (follows
group conversation in a reverberant room or where there is
some interfering noise, such as a classroom or restaurant)
and CAP 9 (use of telephone with an unknown speaker in
unpredictable context).

The TVL is a test of receptive and expressive language
that is appropriate for ages 30 months to 6 years. The TVL
scales have been widely used for children with normal hear-
ing, children with specific language impairment, late talkers,
children with cognitive deficits, and children with hearing
impairments. The test is organized into several sections:
word comprehension, sentence comprehension, sentence
repetition, naming test, and elicited speech production on
specific subjects. The TVL tools are toys and pictures, and
the tasks include object manipulation and description based
on structured questions.

Outcome measures of auditory-based performance (ie, word
comprehension, sentence comprehension, and sentence repeti-
tion) were taken for each child from the 4 different groups, as
close as possible to 30 months of age and then at 6-month inter-
vals, until 48 months of device experience was reached.

The Peabody Picture Vocabulary Test–Revised (PPVT-
R) was administered to all children to test receptive lan-
guage level.

The Fanzago test is based on 22 tables representing 114
pictures that include all consonants and vowels of standard
Italian in every possible position (initial, median, and conso-
nant clusters). Words were presented to the child using a
live voice with no visual cues, and the child was asked to repeat what he or she heard. Each child’s speech production was transcribed and evaluated according to the normal speech developmental pattern of a native Italian speaker. The total number of incorrectly repeated phonemes and clusters was expressed in terms of a phonetic difficulties percentage. 21

The Bortolini test consists of 90 pictures representing objects and events, which the child describes/names, and 3 stories (2 stories with 6 pictures each and 1 story with 4 pictures). The picture-stimuli can elicit words that include most of the occurrences of all the standard Italian language phonemes (in initial and median position, as well as consonant clusters). The first 32 pictures are commonly used to obtain a quick result, as performed in the present study. Using a PC, the pictures were presented to the child one at a time, and the child was asked to describe what he or she saw. The child’s speech production sample must include at least 100 words for children aged 30 months and 250 to 300 words for older children. The child’s speech production sample was videotaped and transcribed according to the International Phonetic Alphabet. The sample was then evaluated in terms of phonetic inventory: each phoneme produced in the initial and median positions, in at least 2 different words, was included in the phonetic inventory (expressed in percent phonemes correctly produced). 22

Safety

For the safety issues, the following parameters were investigated in each child over a 4-year longitudinal follow-up: duration of surgery, heart rate, cardiac arrest, bradycardia, asystole/ventricular fibrillation, hypotension, body temperature variation, blood pressure variation, blood loss, bronchospasm, pulmonary insufficiency, bronchoscopy, laryngospasm, duration of hospitalization, and peri- and postoperative complications (flap necrosis, delay in wound healing, fever, facial nerve injury, otitis media).

Statistical Analysis

The Kolmogorov-Smirnov test was used to check the data distribution. The analysis of variance (ANOVA) test with the Tukey post hoc test or the χ² test was used to assess the differences among groups as appropriate. Statistical significance was set at P < .05.

Approval was obtained by the University of Verona Institutional Review Board and in all hospitals where CI surgery was performed.

Results

Demographic data are presented in Table 1. No statistically significant differences between groups emerged in terms of sex distribution (P > .05), and all etiologies were equally present in the 4 groups. Subjects developed normally with no additional disabilities during the study period. The mean duration of surgery was approximately 60 minutes, and it was statistically significantly lower in the 2- to 6-month cohort (52 ± 8 minutes, P < .01) and the 7- to 12-month group (58 ± 9 minutes, P < .05) compared with the older children (69 ± 7 and 71 ± 11 minutes, respectively, in the 13- to 18-month and 19- to 24-month groups). Correct implantation, with complete insertion of the cochlear electrodes, was achieved in all patients, and the tests confirmed correct functioning of the electrodes. All 45 patients used their CIs all day long on a daily basis.

Safety

No major anesthesiological or surgical complications such as cardiac arrest, facial palsy, or flap breakdown were observed. Among minor anesthesiological complications, 2 children aged 13 and 24 months presented transitory bronchospasm and hypotension, both of which resolved with medical treatment. No laryngospasm was observed in any child, and no intensive care was necessary. Mean heart rate was 132 ± 9, 124 ± 12, 111 ± 14, and 105 ± 12 beats/min in children in the 2- to 6-month, 7- to 12-month, 13- to 18-month, and 19- to 23-month groups, respectively. The difference was statistically significant, with the youngest group experiencing the highest heart rate (P < .05), reflecting the age-appropriate heart rate of infants. No sudden rise or fall in body temperature was observed during surgery in any child. Blood pressure range during surgery was 60 to 95 mm Hg, 60 to 100

<table>
<thead>
<tr>
<th>Table 1. Demographic and Clinical Data of the Study Populations</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Group</strong></td>
</tr>
<tr>
<td>Number of patients</td>
</tr>
<tr>
<td>Age at implantation, mo, mean ± SD</td>
</tr>
<tr>
<td>Sex, M/F, No.</td>
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<tr>
<td>Etiology, No.</td>
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Abbreviation: NA, not applicable.
mm Hg, and 65 to 100 mm Hg for children in groups 1, 2, and 3 to 4, respectively. The difference among groups was not statistically significant ($P > .05$).

No perioperative surgical complications were encountered in the present series of children. Blood loss was recorded as less than 30 mL in all patients.

There were 3 minor postoperative complications (0.7%) in the total population: 2 cases of wound seroma (1 in the 7- to 12-month group and 1 in the 13- to 18-month group) and 1 case of wound infection in the 19- to 24-month group; all were treated conservatively.

Children in the youngest group were discharged within 2.4 ± 1.3 days, 7- to 12-month-old children within 1.9 ± 1.1 days, and children in the 2 older groups after 1.6 ± 0.8 and 1.3 ± 0.7 days. Statistically significant differences only emerged when comparing the 2- to 6-month group with the 19- to 24-month cohort ($P < .05$). Two children, 16 and 19 months old, were readmitted to the hospital because of vomiting and fever 2 days after discharge. They were treated with intravenous infusion of antibiotics and discharged after 3 days. Delayed wound healing (>10 days after surgery) was observed in 3 children in the 13- to 18-month group and in 2 subjects in the 19- to 24-month cohort.

Within 2 years of implantation, postoperative otitis media was observed in the same ear as the CI in 3 children in the 7- to 12-month group; all were treated medically with no further complications. No complications related to CI activation or long-term use were evident in any subject; none of the children suffered facial nerve stimulation.

**Auditory-Based Performance**

The CAP II test showed statistically significant differences between groups, with the 2- to 6-month cohort showing higher scores than all other implanted children (Figure 1; $P < .01$). In addition, the performance of the youngest group did not differ significantly from the normal-hearing group.

Word and sentence comprehension (TVL) at the 42- to 47-month follow-up (Figures 2 and 3) showed that the 2- to 6-month group scores were not statistically significantly different from the 7- to 12-month ones, and no statistically significant difference was evident between the first group and the normal-hearing children. On the other hand, the difference between the 7- to 12-month group and the normal-hearing children was statistically significant ($P < .01$).

In the TVL, sentence repetition task at the 42- to 47-month interval, the differences were statistically significant between the first 2 groups (Figure 4; $P < .05$).

Receptive vocabulary (PPVT-R) revealed significant differences, with the youngest group achieving consistently better results than the other CI groups (Figure 5) and performance close to the normal-hearing group.

At the 48-month interval, the Fanzago speech production test results revealed better articulation proficiency in the youngest group compared with the second youngest group ($P < .001$), showing that what was initially (24 months) only slightly noticeable became more salient after a few years of auditory experience (Figure 6).

In PFIL speech production tests, the differences among the 4 groups were statistically significant both at 24 and 48 months, demonstrating that age at fitting was a significant factor in these findings (Figure 7).

**Discussion**

Since the earliest reports, severely deaf children fitted with CIs have shown dramatic speech perception and production improvements, so that they may now enjoy a similar quality of life as their normal-hearing peers. This progress may be credited to several mutually supportive factors, some attributable to technologic advances and some to a number of daring otologists who decided to implant individuals with considerably more residual hearing and at progressively younger and younger ages.

Currently, in many centers, children aged 6 to 8 months are being implanted when insufficient benefit from hearing aids can be identified, reporting significantly improved auditory and linguistic performance. Several converging lines of research support very early CI in children, suggesting that this procedure might also be desirable for infants younger than 6 months. Critical periods for the development of hearing may extend from the sixth month of fetal life to the early postnatal period with regard to phonology and, later, in other spoken language elements. Auditory development begins well before birth, and fetal auditory sensory abilities are observed from about 26 to 28 weeks' gestational age. At birth, the auditory sensory mechanism of the human neonate is fully functional and ready to establish neural connections based on auditory experience.
Early language exposure, through social interaction, shapes the developing nervous system. Without this, linguistic abilities diminish quickly, and only early access to language...

Figure 2. Average results for the word comprehension subscale of the TVL (Test di Valutazione del Linguaggio, livello pre-scolare) over time for the 4 implant groups. #Tukey post hoc test. ##Analysis of variance test.

Figure 3. Average results for the sentence comprehension subscale of the TVL (Test di Valutazione del Linguaggio, livello pre-scolare) over time for the 4 implant groups. #Tukey post hoc test. ##Analysis of variance test.

Figure 4. Average results for the sentence repetition subscale of the TVL (Test di Valutazione del Linguaggio, livello pre-scolare) over time for the 4 implant groups. #Tukey post hoc test. ##Analysis of variance test.

Figure 5. Receptive language growth (Peabody Picture Vocabulary Test–Revised) score over time (months) in the 4 groups of children. #Tukey post hoc test. ##Analysis of variance test.

Early language exposure, through social interaction, shapes the developing nervous system. Without this, linguistic abilities diminish quickly, and only early access to language...
provides a profoundly deaf child an opportunity to develop within the normal continuum. Deaf children identified and treated younger than 6 months have better and more rapid language development than children identified and treated later.11

In the present study, the population of infants younger than 6 months has the lowest mean age (3.9 months) described in the literature. These infants demonstrated systematically better auditory-based performance compared with all the other infants and children. Both receptive vocabulary and speech production in the youngest group were comparable with normal-hearing children and significantly better than growth rates achieved by children implanted after 6 months. The results show that these children were provided with the possibility of achieving their full potential, offsetting the need to learn at a faster than normal rate to attain age-appropriate norms.

Indeed, with a longer follow-up, differences outlined in the present study may disappear for some essential functions such as language comprehension but probably not for more complex abilities, related to specific and sophisticated phonetic, semantic, and morphosyntactic skills. It is believed that these can only be acquired during the early critical and sensitive developmental period, when sensory inputs lead to specialization of specific areas of the brain for language.12-14

A specialist pediatric team of experts, including a pediatric audiologist, neuroradiologist, surgeons, and anesthesiologists, is critical to achieve proper diagnosis, safe treatment, and correct rehabilitation in these children. Most of the outcomes of safety measures indicate equivalent values for younger and older children. As CI for children younger than 6 months is not as yet an established routine, the youngest group was observed postoperatively for a longer period, with longer hospitalization. Interestingly, the younger children had a shorter surgical time compared with the older ones because of their very thin skulls and because of the degree of pneumatization of the mastoid bone. In fact, the time for all the surgical steps and the amount of bone work were greatly reduced, from preparing the island of bone to accommodate the entire speech processor to reaching the cochlea, via an antrectomy and a posterior tympanotomy.

Because of the small sample size of infants younger than 6 months fitted with CIs, a generalization of the reduced complication rate results observed in the present study is certainly limited. The apparently high rate of delayed wound healing (11%) may be related to the low cutoff (>10 days) adopted, whereas other centers may not consider wound healing of 13 to 15 days to be delayed. Indeed, wound seroma formation is higher than that reported in most pediatric CI series, but the total wound complication rate in our series (6.7%) is close to or even lower than literature reports of 5.6%5 and 10%.10 Furthermore, this article reports an uncontrolled observational study on a small group, with personal audiological criteria and with limited support from the literature due to the innovative nature of the study. The data from the present study must be considered explorative, necessitating a more extensive study in terms of numbers of patients followed up.

In summary, given the current electrophysiological diagnostic procedures, allowing for very early detection of profound hearing loss, associated with advanced anesthesiological and rehabilitation techniques, we believe that the advantages and disadvantages of CI surgery in infants younger than 6 months should be more closely considered and investigated.
Author Contributions

Vittorio Colletti, conception, design, patient care, data acquisition and interpretation, drafting, revision, final approval; Marco Mandala, conception, design, patient care, data acquisition, analysis and interpretation, drafting, chart review, revision, final approval; Vittorio Colletti, conception, design, patient care, data acquisition and interpretation, drafting, revision, final approval.

Disclosures

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Electrocochleography during Cochlear Implantation for Hearing Preservation

Marco Mandala, MD1, Liliana Colletti, PhD1, Giovanni Tonoli, MD1, and Vittorio Colletti, MD1

No sponsorships or competing interests have been disclosed for this article.

Abstract

Objective. To determine whether intraoperative electrocochleography during cochlear implant surgery provides online feedback to modify surgical procedure, reduce trauma, and increase preservation of residual hearing.

Study Design. Prospective cohort study.

Setting. Tertiary referral center, Otolaryngology Department, University of Verona.

Subjects and Methods. Twenty-seven adult patients undergoing cochlear implant surgery who had low- to mid-frequency (0.25-2 kHz) auditory thresholds measured preoperatively were enrolled. Fifteen subjects had compound action potentials measured to assess cochlear function during surgery. In those patients, surgery was modified according to electrocochleographic feedback. Twelve control subjects underwent cochlear implant surgery with blinded electrocochleographic monitoring.

Results. The average preoperative pure-tone audiometry thresholds (0.25-2 kHz) were 74.3 ± 10.2 and 81.5 ± 12.7 dB hearing level (HL) in the electrocochleographic feedback and control cohorts, respectively (P < .05). Compound action potential recordings showed a mean maximum latency shift of 0.63 ± 0.36 ms and normalized amplitude deterioration of 59% ± 19% during surgery. All of these changes reverted to normal after electrode insertion in all but 1 subject in the electrocochleographic feedback group. The average shifts in postoperative pure-tone average thresholds (0.25-2 kHz), evaluated before activation, were 8.7 ± 4.3 and 19.2 ± 11.4 dB HL in the electrocochleographic feedback and control cohorts, respectively (P = .0051). Complete hearing preservation (loss of ≤ 10 dB) at 1 month before activation was achieved in 85% (11/13) of electrocochleographic feedback subjects and in 33% (4/12) of control patients (P = .0154).

Conclusion. Monitoring cochlear function with electrocochleography gives real-time feedback during surgery, providing objective data that might help in modifying the surgical technique in ways that can improve the rate of hearing preservation.

Keywords

intraoperative, monitoring, electrocochleography, cochlear implant, hearing preservation

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The pathophysiology of hearing loss during and immediately after cochlear implant (CI) activation is largely unknown. Human temporal bone studies have helped to elucidate traumatic mechanisms of intracochlear electrode placement and optimize surgical cochleostomy placement.1-4 In recent years, the possibility of preserving residual hearing after CI has been documented by several authors.5-8 To minimize trauma to cochlear structures during CI, all manufacturers have focused their engineering efforts on designing and developing special flexible electrodes with reduced cross-sectional dimensions. It has also been suggested to perform “soft CI surgery” regardless of the amount of preoperative residual hearing, reduce cochlear trauma and improve spiral ganglion cell survival, and, consequently, improve the long-term outcomes.

Preoperative vs postoperative auditory threshold studies9-12 have clearly demonstrated the possible deleterious consequences of CI on residual hearing but have not provided clear evidence of the specific steps that correlate with the corresponding amount of loss. To this end, information on the trauma induced by the type of cochleostomy and of electrode insertion modalities should be gathered in real time, while surgery is ongoing, so that the surgeon can understand the causative maneuvers and decide whether to modify the surgical procedure to minimize trauma to the cochlea accordingly. Today this can be pursued by using a neurological auditory intraoperative monitoring

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Among the different NIM techniques (ie, electrocochleography [ECoG], auditory brainstem response [ABR], and audiometry steady-state response [ASSR]) used during hearing preservation, ECoG can satisfy these needs properly, furnishing large-amplitude potentials and allowing adequate representation of evoked potentials after a few sweeps. Electrocochleography monitoring for hearing preservation in CI has been demonstrated to be reliable in the animal model, whereas ASSR has also been adopted in humans.

In the present study, we verified whether intraoperative ECoG during CI provides useful online feedback to the surgeon to immediately modify surgical procedure, reduce damage to the cochlea, and increase the prevalence of short-term preservation of residual hearing.

Methods

Twenty-seven patients participated in the study between January 2008 and June 2011. Eligibility criteria included the presence of bilateral severe-to-profound sensorineural hearing loss in the mid- to high-frequency range with residual hearing thresholds mainly at low frequencies. All patients were fitted with a full-length (31.5-mm) FLEX\textsuperscript{TM} electrode (MED-EL, Innsbruck, Austria) specifically designed for atraumatic insertion. Patients were alternatively assigned to the ECoG feedback group (ECoG FG) or the ECoG non-feedback group (ECoG NFG) where monitoring was blinded to the surgeon. Thirteen adults with measurable auditory thresholds in the low to mid-frequencies preoperatively had CIs fitted, and intraoperative compound action potentials (CAPs) were measured at multiple times during surgery, with surgery modified according to ECoG feedback (ECoG FG). Two subjects who underwent ECoG monitoring were excluded from the ECoG FG and analyzed separately because they experienced a persistent perilymphatic outflow at the time of cochleostomy. Twelve subjects (ECoG NFG) had CIs fitted without ECoG feedback (blinded monitoring). In this group, in fact, the ECoG recordings were not visualized on the screen.

In this study, threshold, amplitude, and latency of CAPs were sequentially measured at several points during surgery. Every patient had auditory thresholds (0.25, 0.5, 1, and 2 kHz) measured and compared pre- and postoperatively. Postoperative evaluation was performed before CI activation. This was done to avoid any possible interfering effect related to CI activation that could alter the interpretation of the potentially causative surgical factors, and these thresholds were compared with the preoperative thresholds.

The ECoG CAP parameters were obtained using the Medelec Synergy N-EP (CareFusion, San Diego, California). Electrocochleography was recorded using a custom-made cotton wick electrode (+) placed close to the round window (RW) (Figure 1) and 2 subdermal electrodes placed, respectively, over the ipsilateral tragus (+) and the sternum (“ground”). Alternating clicks (11 pps) and 0.25-, 0.5-, 1-, and 2-kHz tone bursts were initially presented from 100 dB hearing level (HL) to the threshold level after electrode placement and at the end of surgery. Then, ECoG latency and amplitude variations at 100 dB HL were analyzed during surgery. The ECoG potentials were filtered through a 100- to 3000-Hz bandpass filter and averaged over several responses. The acoustic stimuli were calibrated and delivered from a Walkman-type earphone connected directly to the evoked potential system. The earphone was coupled to the ear canal, and a bandage was placed over the meatus to hold it in place and to prevent fluid from entering the ear canal. The pinna was then reflected anteriorly. The postauricular area was prepped and draped in a sterile fashion. A classic mastoidectomy was performed with a facial recess approach to the RW. At this point, the first ECoG measurements were performed to obtain baseline data. Four sets of data were collected to test the reliability of the procedure. Each CAP recording took 3 to 5 seconds to measure so that the complete set of frequencies could be obtained in around 1 minute. In 5 patients, the tone bursts could not clearly evoke CAP recording, and the testing was continued with clicks only. Subsequently, both CI surgery and ECoG evoked potentials were continuously recorded and simultaneously displayed on the screen only in the ECoG FG. This allowed the surgeon of the ECoG FG to immediately observe any change in morphology of the potentials and, if necessary, modify the procedure accordingly. The video-recorded surgeries, with the superimposed ECoG recordings, were later submitted to a detailed analysis (Figure 2). In addition, both the ECoG FG and the ECoG NFG patients were alternatively submitted to electrode insertion via a cochleostomy at the anterior-inferior edge of the RW niche or via a RW membrane opening. Data before, during, and after the several surgical steps are detailed in the Results section.
The Kolmogorov-Smirnov test was used to check the data distribution. The Student $t$ test and the Fisher exact test were used to compare measurements between the 2 cohorts. The analysis of variance (ANOVA) test with the Tukey post hoc test was used to assess the differences between multiple measurements (CAP latency shift and normalized amplitude variation from baseline). Statistical significance was set at $P < .05$.

Approval was obtained by the University of Verona Institutional Review Board.

Results

Demographic data from the 2 populations are reported in Table 1. No statistically significant differences in terms of age and sex could be observed between the 2 cohorts ($P > .05$). All patients had complete insertions of the electrodes, and no postoperative complications were encountered in any subject.

The preoperative average pure-tone audiometry (PTA) threshold (dB HL) was not statistically different between ECoG FG and ECoG NFG patients ($P > .05$; Table 1). Postoperative audiological evaluation showed a statistically lower PTA average threshold in the ECoG FG patients ($83 \pm 9.5$ vs $100.7 \pm 16.9$ dB HL in ECoG NFG patients; $P = .0034$), who consequently showed a significantly lower threshold shift when compared with the ECoG NFG patients ($8.7 \pm 4.3$ vs $19.2 \pm 11.4$ dB HL; Figure 3, $P = .0051$).

Complete hearing preservation (loss of $\leq 10$ dB at the 0.25-, 0.5-, 1-, and 2-kHz pure-tone average) at 1 month postoperatively and before CI activation was achieved in 85% (11/13) of ECoG FG subjects and in 33% (4/12) of ECoG NFG subjects ($P = .0154$). Intraoperative ECoG average threshold values, both for clicks and tone burst-evoked CAPs, showed in the ECoG FG patients a mean shift of $7.6 \pm 3.9$ dB HL with no statistically significant differences before (baseline) and at the end of surgery ($P = .2$; $t$ test). In the ECoG NFG patients, the mean threshold shift between baseline and completion of surgery was $25.9 \pm 21.4$ dB HL ($P = .0007$; $t$ test). The difference in ECoG threshold shift at the end of surgery between the 2 groups showed statistically and significantly better outcomes ($P = .0015$; Table 1) in the ECoG FG patients.

Data plotted in Figure 4 show mean CAP latency shifts and normalized amplitude variations at 100 dB HL for both click and tone bursts (assembled values to facilitate comparison) at different stages of surgery from baseline to the end of the procedure in the ECoG FG patients. No significant differences in latency results were identified before and after drilling the cochleostomy or opening the RW membrane ($P > .05$; Tukey post hoc test). A statistically significant increase in latency was observed after the first stage of electrode insertion into the scala tympani ($P < .0001$; Tukey post hoc test). This observation led to performing the subsequent array insertion in a slow and stepwise modality ($P > .05$). All patients had complete insertions of the electrodes, and no postoperative complications were encountered in any subject.

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Table 1. Demographic and Clinical Data of the Study Populations

<table>
<thead>
<tr>
<th></th>
<th>ECoG FG</th>
<th>ECoG NFG</th>
<th>$P$ Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Number of subjects</td>
<td>13</td>
<td>12</td>
<td></td>
</tr>
<tr>
<td>Age, y</td>
<td>58.6 ± 12.3</td>
<td>61.4 ± 15.1</td>
<td>$6^a$</td>
</tr>
<tr>
<td>Sex, male/female</td>
<td>7/6</td>
<td>5/7</td>
<td></td>
</tr>
<tr>
<td>Preoperative PTA (0.25, 0.5, 1, 2 kHz, dB HL)</td>
<td>74.3 ± 10.2</td>
<td>81.5 ± 12.7</td>
<td>$3^b$</td>
</tr>
<tr>
<td>Postoperative PTA (0.25, 0.5, 1, 2 kHz, dB HL)</td>
<td>83 ± 9.5</td>
<td>100.7 ± 16.9</td>
<td>.0034^a</td>
</tr>
<tr>
<td>Baseline ECoG threshold</td>
<td>44.3 ± 12.1</td>
<td>47.6 ± 16.3</td>
<td>$6^a$</td>
</tr>
<tr>
<td>End-of-surgery ECoG threshold</td>
<td>51.9 ± 16.4</td>
<td>87.8 ± 31.6</td>
<td>.0015^a</td>
</tr>
</tbody>
</table>

Abbreviations: ECoG FG, electrocochleography feedback group; ECoG NFG, electrocochleography non-feedback group (blinded monitoring); HL, hearing level; PTA, pure-tone average.

^aFisher exact test.

^bHeterogeneity of variance test.

^cMean threshold among clicks and tone burst stimulations.
in the ECoG FG patients. The subsequent stepwise insertions of the array and the packing of cochleostomy or RW area induced temporary and insignificant \( (P > .05; \text{Tukey post hoc test}) \) latency changes. The Tukey post hoc test of the amplitude data recorded at each surgical step compared with that at baseline indicated that significant changes (reduction) in normalized amplitude variations \( (P < .001) \). Just after the last phase of electrode insertion, CAP amplitude deterioration recovered \( (P > .05) \), but it again declined when packing the cochleostomy or RW area \( (P < .05) \).

Both latency and amplitude changes reverted to normal in all but 1 subject at the end of complete electrode insertion. Postoperative analysis of CAP mean latency shift and normalized amplitude variations in the ECoG NFG showed no significant differences before opening the cochlea or RW membrane \( (P > .05; \text{Tukey post hoc test}) \). A statistically significant increase in latency and amplitude was observed from the cochleostomy step to the complete electrode insertion \( (P < .0001; \text{Tukey post hoc test}) \); Figure 5). Latency and amplitude changes did not recover at the end of surgery in any subjects of the ECoG NFG. Most subjects who achieved complete hearing preservation underwent cochleostomy for CI fitting \( (9/14) \). Statistically significant differences in postoperative outcomes in terms of latency shift at PTA were not observed between patients who underwent cochleostomy \( (14 \text{ subjects}) \) or RW membrane \( (11 \text{ subjects}) \) electrode insertion despite the fact that subjects who underwent cochleostomy showed slightly better hearing preservation outcomes \( (12.1 \pm 6.1 \text{ vs } 15.8 \pm 11.5 \text{ dB HL}; P = .3) \).

When comparing CAP latency and amplitude changes between subjects who underwent cochleostomy or RW membrane electrode insertion, no statistically significant differences could be observed at any stage of surgery apart from the final step of packing with fascia all around the electrode at the site of entry into the cochlea (cochleostomy or RW membrane). After this procedure, the RW-electrode group showed significantly higher latencies and amplitude deterioration CAPs compared with packing of the cochleostomy/electrode group \( (P < .01; \text{Figure 6}) \). The 2 subjects who showed a spontaneous perilymphatic outflow at the time of cochleostomy exhibited, contrary to what was observed in all other monitored patients, a sudden and dramatic improvement in CAP latency and amplitude that rapidly decreased when placing fascia over the cochleostomy \( \text{(Figure 7).} \) Both subjects obtained hearing preservation within 20 dB HL.

Figure 3. Postoperative pure-tone audiometry (PTA) threshold shift of the 2 populations investigated: electrocochleography \( (\text{ECoG}) \) monitored and control groups \( (t \text{ test}) \). FG, feedback group; HL, hearing level; NFG, nonfeedback group.

Figure 4. Latency shift and normalized amplitude variations at 100-dB hearing level \( (\text{HL}) \) stimulation at different stages of the surgery in the electrocochleography \( (\text{ECoG}) \) feedback group. RW, round window; RWm, round window membrane. " Analysis of variance test. " Tukey post hoc test.

Figure 5. Postoperative pure-tone audiometry (PTA) threshold shift of the 2 populations investigated: electrocochleography \( (\text{ECoG}) \) monitored and control groups \( (t \text{ test}) \). FG, feedback group; HL, hearing level; NFG, nonfeedback group.
Discussion

NIM Options for Hearing Preservation in CI Surgery

The aim of NIM\(^{15}\) is to provide indications as to the changes in the neurophysiological status of the auditory pathways during surgery. Early detection of significant damage to these structures can potentially lead to interruption and reversal of the damage process, with the ultimate goal of preventing hearing loss. Auditory steady-state response\(^{16,17}\) and ABR are unaffected by sedation or sleep and allow the detection of good physiologic responses in children and adults.\(^{18}\) However, they share similar limitations in the noncontinuity of testing and in the excessive time required for data acquisition, preventing analysis of each individual surgical maneuver in a specific and detailed manner and limiting correlations with the causes and effects of cochlear damage in real time. These limitations are absent in ECoG, allowing the procedure to be used effectively for intraoperative monitoring of CI surgery.

Experience with ECoG

At the beginning of our investigation, we evaluated all the parameters of the ECoG response: cochlear microphonics (CM), summating potentials, and CAPs. It was soon realized that CAPs were the most sensitive markers of early interaction between the surgical action and cochlear structures. The fact that the CAP proved to be a more sensitive indicator of cochlear injury can be attributed to the assumption that the CMs represent the output of a larger fraction of the cochlea than does the CAP, so that a limited local change would have less effect on the CM than on the CAP.\(^{19}\) It is believed that the value of using the CM as a routine technique for intraoperative monitoring during CI surgery is minimal because this signal cannot be measured in most hearing-impaired patients.\(^{20}\) The speed and accuracy of the measurements of a near field as ECoG are unlikely to be obtained with ABRs or ASSRs. Even with these limitations, Oghalai et al\(^{14}\) were able to obtain a significantly higher...
percentage of hearing preservation with intraoperative ASSR monitoring.

From this study, we learned several points important to the improvement of CI surgery for hearing preservation.

1. Drilling a cochleostomy on the promontory, with either a high- or low-speed drill, does not induce a permanent alteration of CAPs.

2. Similarly, opening the access to the scala tympani via a small or a large cochleostomy or via the RW membrane does not produce significant changes in CAP latency and amplitude, provided that no suction of perilymph from the cochleostomy is performed.

3. Should perilymph be suctioned, prompt administration of a few drops of physiologic solution into the scala tympani might reestablish the previous CAP amplitude if changes in amplitude are simply due to an excessive perilymph evacuation without permanent distortion effects on the basilar membrane and significant alteration in the normal physiological response to acoustic stimuli of the cochlea to the point of hair cell function loss.

4. Direct and abrupt suction of fluid from the opening of the scala tympani to remove bone dust or blood is responsible for significant and often permanent deterioration of CAPs.

5. Following cochleostomy, an excessive outflow of perilymph associated with CAP amplitude increase and a latency decrease might be observed. Interestingly, if the cochleostomy is immediately closed with fascia, the CAP parameters revert immediately to their previous values. In some patients, this event can be observed repeatedly and might indicate a condition of perilymphatic hypertension with distorted cochlear hydrodynamics. Similar changes in CAP amplitude (increasing) and latencies (decreasing) may be observed during soft electrode array insertion, indicating minor alterations in the cochlear micromechanics at the level of the basilar membrane.

6. Electrode insertion modalities (fast and single shot vs a very slow and stepwise series of shot insertions) might be responsible for dramatic and permanent shifts in amplitude up to a loss of all the CAPs with loss at all the residual frequencies. This observation suggests that significant trauma to the basilar membrane due to incorrect electrode insertion determines sudden complete impairment of hair cell function that shows no evidence of recovery even when prolonging the observation period. This was also reported recently with ASSR intraoperative monitoring.\(^{14}\)

7. Electrode insertion via cochleostomy or RW does not induce differences in CAP responses, providing evidence that both approaches are substantially identical.

8. When fascia is placed on the 2 openings, it may repeatedly and systematically be verified that reduced amplitudes and increased latencies of CAPs are significantly more evident in the RW membrane compared with the cochleostomy condition. This suggests that the fascia on the RW might simulate a condition of RW blockage, inducing an increase in inner ear perilymph pressure with modification of the basilar membrane micromechanics. This finding may also explain the increased threshold observed by Oghalai et al\(^{14}\) when plugging the cochleostomy with fascia.

This article describes the technique of intraoperative monitoring that is presently routinely used in our department for CI surgery. We have learned how to make subtle, yet valuable, changes in our surgical technique in an attempt to minimize the factors that may interfere with hearing preservation during the CI surgical procedure.
According to the ECoG monitoring experience so far acquired, several steps have emerged as important in preserving hearing when performing CI surgery.

1. The site of exposure of the scala tympani should be carefully selected with a cochleostomy (anteriorly-inferiorly close to the RW membrane) and with an RW approach in the inferior-anterior margin.
2. Constant and copious irrigation should be performed during drilling of the bony lip of the RW niche and the promontory for cochleostomy.
3. There should be no suction on the cochleostomy or RW membrane opening to remove bone dust. This is extremely traumatic (hydrodynamic shock) to the fine structure of the cochlea, altering CAP latency and amplitude and leading to a lower rate of hearing preservation.
4. Exposing the scala tympani via a cochleostomy or the RW membrane is not (necessarily) associated with hearing loss. In some patients, a slight improvement in CAP parameters may be observed, indicating that release of the intracochlear pressure improves the relationship between the basilar membrane and hair cell stereocilia.
5. The electrode array must be inserted into the scala tympani very slowly and with a minimum of 3 stages.
6. The simple act of placing temporalis fascia at the end of the procedure around the opening of the cochleostomy performed through the RW membrane can produce a significant deterioration of CAPs and, subsequently, residual hearing worsening.

Conclusions

Electrocochleography can be performed effectively during CI surgery with real-time monitoring and short-term improvement in the degree of hearing preservation. The online feed-back provided to the surgeon allows immediate appreciation of potential damaging maneuvers so as to minimize trauma to the cochlea and increase the understanding of how subtle technical improvements can increase hearing preservation beyond their current levels.

The rate of short-term complete hearing preservation of the monitored cohort (85% within 10 dB HL) is among the highest ever described in the literature (below 50%).

However, at this time, whether CI ECoG-assisted surgery for hearing preservation may indeed lead to stable improvements in long-term functional outcomes is unknown, but it certainly provides feedback indicating whether surgery and electrode insertion have induced significant acute trauma to the cochlea.

Author Contributions

Marco Mandala, conception, design, patient care, data acquisition, analysis and interpretation, drafting, chart review, revision, final approval; Liliana Colletti, conception, design, patient care, data acquisition and interpretation, drafting, revision, final approval; Giovanni Tonoli, conception, design, patient care, data acquisition, analysis and interpretation, drafting, chart review, revision, final approval; Vittorio Colletti, conception, design, patient care, data acquisition and interpretation, drafting, revision, final approval.

Disclosures

Competing interests: None.

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DISCUSSION

The goal for a congenitally profoundly deaf child is to achieve age-appropriate spoken language and cognitive abilities in the shortest possible time-frame.

Since the earliest reports, severely deaf children fitted with CIs have shown dramatic speech perception and production improvements, so that they may now enjoy a similar quality of life as their normal hearing peers.²⁻⁴

Currently, in many centers, children below 12 months of age are being implanted when insufficient benefit from hearing aids can be identified, reporting significantly improved auditory and linguistic performance⁵,²³,²⁴,²⁶⁻³⁴. Several converging lines of research support very early CI in children, suggesting that this procedure might also be desirable for infants even under 6 months of age.

Auditory development begins even before full term birth, as it is known that hearing begins early in intrauterine life. The newborn and even the fetus not only can hear relatively well, but they are capable of distinguishing their mother’s heartbeat and voice from others¹⁵,⁵⁸ and respond to changes in musical notes¹⁷. Critical periods for the development of hearing may extend from the 6th month of fetal life to the early post-natal period with regard to phonology and, later, in
other spoken language elements. At birth, the auditory sensory mechanism of the human neonate is fully functional and ready to establish neural connections based on auditory experience. Early language exposure, through social interaction, shapes the developing nervous system. Without this, linguistic abilities diminish quickly and only early access to language provides a profoundly deaf child an opportunity to develop within the normal continuum. Deaf children identified and treated under 6 months have better and more rapid language development than children identified and treated later.

Other sensorimotor and cognitive development also rely on auditory development and can be seriously delayed the longer implantation is delayed. Indeed, some developmental trajectories have a biological window that closes if the necessary elements are not available within the “critical period” of development. The delays in the development of auditory performance could represent significant challenges for the development of working memory and general cognitive development.

Does early cochlear implantation restore sufficient auditory experience to overcome the negative effects of early deprivation on auditory, language and cognitive performance? Does implantation at ages under 6 months provide additional benefits compared to implantation at older ages? To date published research on early
implantation presents a conflicting message. Holt and Svirsky (2008)\textsuperscript{25} conclude that there is no additional benefit in performance based on a small number of children implanted under 12 months of age. However, Colletti et al. (2005)\textsuperscript{23} showed a clear advantage in babbling measures in 10 infants implanted before 12 months. Dettman et al. (2007)\textsuperscript{5} also showed clear advantages in early implantation based on results from 19 children implanted under 12 months of age. Colletti L. (2009)\textsuperscript{24} demonstrated that very early cochlear implantation (below 12 months of age) provides normalization of audio-phonologic development with no complications. A recent meta-analysis concluded that evidence of improved performance on auditory perception/speech production outcomes is limited for children implanted below 12 months\textsuperscript{34}.

Waltzman et al. in 2005\textsuperscript{29} and Valencia et al. in 2008\textsuperscript{31} presented data from children implanted at a mean age of 9.6 months (range: 7-11) and 9.2 months (range: 6.7-11.7) months, respectively. More recently Roland et al. (2009)\textsuperscript{33} reported data on 50 infants with a mean age of 9.1 months (range: 5-11) followed for up to 7 years. On all auditory and speech tests the youngest group showed superior performance to results from children implanted later.

The present research demonstrated that children implanted below 12 months of age and, even more infants under 6 months, developed
auditory capabilities faster, produced more intelligible speech earlier, developed language at normal rates and levels and developed grammar skills earlier than children implanted after 12 months of age. This superior performance persisted out to 10 years of follow-up. These data show evidence that earlier implantation results in faster development and these children continue to out-perform children implanted later.

Furthermore, the additional sensory input provided by the CIs clearly supports non-auditory cognitive development. The infant group showed significantly increased results on complex non-verbal cognitive tests (Griffiths Mental Development Scales – Leiter Intenational Performance Scale Revised) compared to the older children. This finding might be ascribed to the higher demand in term of sensory input integration to complete the tasks. Early additional auditory verbal and non-verbal stimuli provided by the CIs may offer the infant the chance of developing a more complex and effective learning strategy in a very “critical period” of their development. The activation of the auditory channel enriches the children’s sensory stimulation and brings the level of attention to a more sustained level on a wider range of stimuli. Similar results were recently described in children fitted with the auditory brainstem implant. These findings support the hypothesis that early auditory stimulation
might play a fundamental role in the development of higher cognitive functions where multisensory integration is essential. Thus, delays in the onset of hearing can delay aspects of cognitive development. In particular, the population of infants below 6 months presented herein has the lowest mean age (3.9 months) described in the literature. These infants demonstrated systematically better auditory-based performance compared with all the other infants and children. Both receptive vocabulary and speech production in the youngest group were comparable with the normally hearing children and significantly better than growth rates achieved by children implanted after 6 months. The results show that these children were provided with the possibility to achieve their full potential, offsetting the need to learn at a faster than normal rate to attain age-appropriate norms. With a longer follow-up, differences outlined in the group of infants examined may disappear for some essential functions such as language comprehension, but probably not for more complex abilities, related to specific and sophisticated phonetic, semantic and morphosyntactic skills. It is believed that these can only be acquired during the early critical and sensitive developmental period, when sensory inputs lead to specialization of specific areas of the brain for language.
A specialist pediatric team of experts, including a pediatric audiologist, neuroradiologist, surgeons and anesthesiologists is critical to achieve proper diagnosis, safe treatment and correct rehabilitation in these children. Most of the outcomes of safety measures indicate equivalent values for younger and older children with no higher rate of complications in infants below 6 months. It is now clear that CIs\textsuperscript{39-43} are highly cost-effective in adults and children, but the possible additional economic benefit of very early implantation in infants has not been reported and is not known. It is well recognized that profoundly hearing-impaired infants must be identified and treated with CIs very early in their lives to improve their chances of joining hearing children in mainstream education, social life and working.

The present research indicates that a significant net saving to society is achieved by decreasing the age of implantation below 12 months of age. In the light of these outcomes, many costs and services provided prior to implantation (hearing aids and their maintenance, speech therapy, educational costs, days off work and travel expenses for parents) emerge as substantially cost-ineffective.

While medical costs undergo a slight cost reduction on delaying the age of implantation, the costs for education and in particular for the family increase dramatically for children implanted at older ages.
The net saving to society over a 10 year time period for an infant implanted at an age below 12 months is approximately 21,000 € as against children implanted between 12-23 months and rises to more than 35,000 € when the age of implantation is over 6 years. Thus, implanting children in the first years of life minimizes not only language delays but also the overall costs to families. Furthermore, all subjects implanted before 36 months were full-time users of the implant while one subject implanted after 6 years became a non-user. This finding supports the view that especially age of implantation, educational considerations, and family support may play an important role in becoming a non-user.

At the age of 10 years infants implanted below 12 months may reach a vocabulary age of 9.5 years at the Peabody Picture Vocabulary Test – Revised while comparable children implanted at 6 years of age reach a vocabulary age of only 5.8 years. The substantial difference observed in the cost for gaining one year of vocabulary age between the youngest group and older children supports the efficacy of early implantation in terms both of outcomes and the net saving to society. Nevertheless, when studying a pediatric population retrospectively a recall bias could frequently cause overestimation of utility gains by
parents. Furthermore, the questionnaire adopted to study mainly family costs might have overestimated certain cost categories.

Comparison with similar studies from different countries is not easy due to specific health care financing conditions, educational settings, type of costs (direct and indirect) and period of time evaluated. Cost analyses performed in France\textsuperscript{39} and Germany\textsuperscript{43} apparently showed approximately similar costs while studies conducted in the United States\textsuperscript{42} and United Kingdom\textsuperscript{40,41} demonstrated higher costs.

Preservation of residual hearing during and after CI surgery is a key point since indications for bionic hearing restoration are widely expanding. The aim of NIM\textsuperscript{67} is to provide indications as to the changes in the neurophysiological status of the auditory pathways during surgery. Early detection of significant damage to these structures can potentially lead to interruption and reversal of the damage process, with the ultimate goal of preventing hearing loss. ASSR\textsuperscript{68,69} and ABR are unaffected by sedation or sleep, and allow the detection of good physiologic responses in children and adults\textsuperscript{18}. However, they share similar limitations in the non-continuity of testing and in the excessive time required for data acquisition, preventing analysis of each individual surgical manoeuvre in a specific and detailed manner and limiting correlations with the causes and effects of cochlear damage in real-time. These limitations are
absent in ECoG, allowing the procedure to be utilized effectively for intraoperative monitoring of CI surgery.

The speed and accuracy of the measurements of a near field as ECoG are unlikely to be obtained with ABRs or ASSRs. Even with these limitations, Oghalai et al.\textsuperscript{57} were able to obtain a significantly higher percentage of hearing preservation with intra-operative ASSR monitoring.

According to the ECoG monitoring experience so far acquired, several steps have emerged as important in preserving hearing when performing CI surgery.

1. Careful selection of the site of exposure of the scala tympani: with a cochleostomy (anteriorly-inferiorly close to the round window (RW) membrane) and with a RW approach in the inferior-anterior margin.

2. Constant and copious irrigation during drilling of the bony lip of the RW niche and the promontory for cochleostomy.

3. No suction on the cochleostomy or RW membrane opening to remove bone dust. This is extremely traumatic (hydrodynamic shock) to the fine structure of the cochlea, altering compound action potentials (CAPs) latency and amplitude and leading to a lower rate of hearing preservation.
4. Exposing the scala tympani via a cochleostomy or the RW membrane is not (necessarily) associated with hearing loss. In some patients, a slight improvement in CAP parameters may be observed indicating that release of the intracochlear pressure improves the relationship between the basilar membrane and hair cell stereocilia.

5. The electrode array must be inserted into the scala tympani very slowly and with a minimum three stages.

6. The simple act of placing temporalis fascia at the end of the procedure around the opening of the cochlea performed through the RW membrane can produce a significant deterioration of CAPs and subsequently, residual hearing worsening.

The rate of short-term complete hearing preservation of the monitored cohort (85% within 10 dB HL) is among the highest ever described in the literature (below 50%)\(^{53-55}\). However at this time, whether CI ECoG assisted surgery for hearing preservation may indeed lead to stable improvements in long-term functional outcomes is unknown, but it certainly provides feedback indicating whether surgery and electrode insertion have induced significant acute trauma to the cochlea.

The major limitation of all these researches are that they report mainly uncontrolled observational studies on small groups, with personal audiological criteria and with limited support from the
literature due to the innovative nature of the studies. The data from the present studies must be considered as explorative and necessitating a more extensive study in terms of numbers of patients followed-up.

**CONCLUSIONS**

The progress in restoring auditory function with CIs may be attributed to several mutually supportive factors, some attributable to technologic advances and some to a number of otologists who decided to implant individuals at progressively younger and younger ages and with considerably more residual hearing.

The presented researches demonstrated audiological, language and cognitive outperformance of deaf infants implanted under 6 months of age. Furthermore, costs of CI to society are inversely related to the age at implantation. The risk of CIs under 6 months of age are minimal in the hands of a highly specialized pediatric team of experts.

Intraoperative monitoring for hearing preservation in CI surgery can be performed effectively during CI surgery and lead to improvement in the degree of hearing preservation. The online feedback provided to the surgeon allows immediate appreciation of
potential damaging manoeuvres so as to minimize trauma to the cochlea and increase the understanding of how subtle technical improvements can increase hearing preservation beyond their current levels.

CI surgery in infants under 6 months and ECoG for hearing preservation should be more closely considered and investigated.

**ONGOING RESEARCHES**

- Bilateral cochlear implantation in children below 12 months
- Extended data on a larger number of infants, including those fitted with CIs below 6 months of age, both in term of audiological, language and cognitive outcomes and cost.
- Long-term hearing preservation in ECoG-assisted CI surgery
- Hearing preservation in CI surgery with different electrode arrays
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