Audiology

Role of bimodal stimulation for auditory-perceptual skills development in children with a unilateral cochlear implant

Ruolo della stimolazione bimodale nello sviluppo delle abilità percettivo-uditive nei bambini con impianto cocleare monolaterale

P. MARSELLA, S. GIANNANTONIO, A. SCORPECCI, F. PIANESI, M. MICARDI, A. RESCA
Audiology-Otology Unit and Cochlear Implant Referral Center, “Bambino Gesù” Pediatric Hospital, Rome, Italy

SUMMARY

This is a prospective randomised study that evaluated the differences arising from a bimodal stimulation compared to a monaural electrical stimulation in deaf children, particularly in terms of auditory-perceptual skills development. We enrolled 39 children aged 12 to 36 months, suffering from severe-to-profound bilateral sensorineural hearing loss with residual hearing on at least one side. All were unilaterally implanted: 21 wore only the cochlear implant (CI) (unilateral CI group), while the other 18 used the CI and a contralateral hearing aid at the same time (bimodal group). They were assessed with a test battery designed to appraise preverbal and verbal auditory-perceptual skills immediately before and 6 and 12 months after implantation. No statistically significant differences were observed between groups at time 0, while at 6 and 12 months children in the bimodal group had better scores in each test than peers in the unilateral CI group. Therefore, although unilateral deafness/hearing does not undermine hearing acuity in normal listening, the simultaneous use of a CI and a contralateral hearing aid (binaural hearing through a bimodal stimulation) provides an advantage in terms of acquisition of auditory-perceptual skills, allowing children to achieve the basic milestones of auditory perception faster and in greater number than children with only one CI. Thus, “keeping awake” the contralateral auditory pathway, albeit not crucial in determining auditory acuity, guarantees benefits compared with the use of the implant alone. These findings provide initial evidence to establish shared guidelines for better rehabilitation of patients undergoing unilateral cochlear implantation, and add more evidence regarding the correct indications for bilateral cochlear implantation.

KEY WORDS: Cochlear implant • Bimodal stimulation • Deaf children • Auditory-perceptual skills

INTRODUCTION

Cochlear implants (CI), together with the introduction of universal newborn hearing screening programs, have represented a revolution in the treatment of paediatric severe-to-profound sensorineural hearing loss. In fact, children who receive an early diagnosis of deafness, proper speech
Advantages of bimodal stimulation

rehabilitation and undergo early cochlear implantation are able to develop auditory and linguistic skills at par with their hearing peers [1-11]. In Italy, the current shared guidelines state that cochlear implantation is recommended in children older than 12 months with bilateral profound sensorineural hearing loss who do not get significant benefit from conventional hearing aids [12]. Little further information is given about bilateral cochlear implantation, other than its recommendation in selected cases, such as in deaf-blind subjects and in case of deafness from meningitis, where there is a substantial risk of rapid cochlear ossification [13, 14].

Next to these two hearing solutions (unilateral and binaural electrical hearing), there is actually a third option, namely the chance to stimulate the ear opposite to the implanted side using a conventional hearing aid (binaural hearing though a bimodal stimulation). Surprisingly, in normal clinical practice (NCP) no stimulation of the contralateral ear is recommended in patients undergoing unilateral cochlear implantation. The decision to not stimulate the contralateral ear with a conventional hearing aid is due to multiple factors: in some cases, the monaurally implanted child refuses to wear the hearing aid on the other side, given the little perceived benefit compared with the electrically stimulated side; in others, parents themselves disregard the application of the contralateral hearing aid, considering it unnecessary after cochlear implant surgery; finally, the absence of scientific evidence about the effective benefit of a contralateral acoustic stimulation authorises the clinician, as well as parents, to minimise this aspect and maintain the belief that restoration of a monaural hearing through a CI is sufficient to ensure a proper development of perceptual-language skills. This belief is also supported by the treatment given to patients who, following trauma or infection, lose their hearing in only one ear (acquired unilateral deafness) and who, in NCP, receive no indications for the application of a conventional hearing aid.

However, several recent studies have demonstrated that restoration of a binaural hearing facilitates sound localisation and improves speech comprehension in noisy environments compared to monaural stimulation alone [15-18]. According to these findings, the ideal goal would be to restore binaural hearing in every circumstance [19]. Therefore, patients undergoing unilateral CI might achieve acceptable binaural processing by bimodal stimulation rather than or before receiving a second implant in the contralateral ear, whose cost/benefit ratio is yet to be fully demonstrated [20]. Some studies have shown that bimodal stimulation improves the auditory-perceptual abilities of adults with usable residual hearing in the non-implanted ear [17]. However, little is known about the comparison between auditory-perceptual performances of prelingually deaf children (0-3 years) with a unilateral CI and those of age-matched peers who benefit from a bimodal stimulation [21].

In this scenario, our work aims to evaluate the differences arising from a bimodal stimulation compared to a monaural stimulation in children. The main outcome measure is auditory-perceptual skills development, which represent the basic and essential prerequisites to development of language. The secondary outcomes are pure tone thresholds in free field and basic perceptive milestone achievement. It is expected that patients with bimodal stimulation develop preverbal and verbal auditory-perceptual skills faster than patients with only a CI and no contralateral stimulation. Finding an advantage for bimodal stimulation would be an important reference for the management of postoperative rehabilitation of children receiving one CI, as well as the basis for investigating the potential benefits of the binaural development of acoustic networks in order to achieve adequate perceptual and communication skills in children with pre-lingual hearing loss.

Materials and methods

Population

The entire protocol was reviewed and approved by the Ethics Committee of the institution (protocol code 811/2014) and in accordance with the Declaration of Helsinki. Written informed consent was obtained from each patient’s parents.

We enrolled patients aged 12 to 36 months with similar demographic and audiological characteristics. All suffered from idiopathic or genetic severe-to-profound bilateral sensorineural hearing loss with some residual hearing, i.e. an unamplified pure tone threshold ≥70 and ≤90 dB HL for frequencies 0.25-0.5 kHz; subjects with an unamplified pure tone threshold ≥90 dB HL for frequencies 0.25-0.5 kHz were excluded from the study. Each patient had already been included in a speech and language rehabilitation programme. Other exclusion criteria were indication for simultaneous bilateral cochlear implantation (e.g., meningitis, deaf-blind children), presence of a concomitant cognitive delay, cochlear malformation, hypo-aplasia of the acoustic nerve and the impossibility to return for follow-up visits.

We enrolled 39 children, among those attending our Centre of Audiology and Otology and waiting for a CI (mean age 23.60 ± 6.24 months; 21 males and 18 females). Each patient underwent unilateral cochlear implantation, all with a perimodiolar array (Cochlear® CI512 electrode or Nucleus Freedom). Children were then divided by means of a simple randomisation into two groups using an Excel® Random Numbers Generator function: those who received and wore only a CI (unilateral CI Group, or Group 1: 21 patients; 11 males and 10 females; mean age at implantation 23.01 ± 5.96 months), and those who were instructed to use both the implant and a conventional hearing aid in the contralateral ear (Bimodal Group, or Group 2: 18 pa-
tients; 10 males and 8 females; mean age at implantation 24.28 ± 6.65 months). All children used bilaterally digital hearing aids prior to implantation; after being assigned to Group 1, subjects had to interrupt the use of the hearing aid in the non-implanted ear as a result of randomisation. Patients in Group 2 used a powerful digital hearing aid, optimised to its best fitting thanks to periodic adjustments by a hearing healthcare professional with extensive experience in paediatric audiology. Since guidelines about specific bimodal fitting protocols are yet to be provided, in Group 2 the hearing aid was regularly fitted by their HA provider, whereas CI fitting was provided by trained professionals within the CI Centre.

**Test battery**

Patients were assessed with a test battery designed to appraise preverbal and verbal auditory-perceptual skills. The entire battery was administered at time 0 (T0, i.e. before unilateral cochlear implantation), and after 6 (T1) and 12 months (T2) of CI use (transcontralateral hearing aid, where required).

- **Auditory gain/benefit testing:** the auditory gain/benefit of the CI with or without the contralateral hearing aid was evaluated in terms of free-field hearing threshold in, i.e. the average threshold for the frequency range 0.5-3 kHz (PTA 0.5-3 kHz), according to the Committee on Hearing and Equilibrium guidelines of the American Academy of Otorhinolaryngology, Head & Neck Surgery. Each child underwent 2 to 5 play audiometry sessions and electrophysiological testing (Auditory Brainstem Responses, ABR) to obtain the best optimised Ci use thanks to periodic adjustments by a hearing healthcare professional with extensive experience in paediatric audiology. Since guidelines about specific bimodal fitting protocols are yet to be provided, in Group 2 the hearing aid was regularly fitted by their HA provider, whereas CI fitting was provided by trained professionals within the CI Centre.

- **Auditory perception testing,** which includes:
  - **Infant-Toddler Meaningful Auditory Integration Scale (IT-MAIS):** a structured interview schedule designed to assess the child’s spontaneous responses to sound in his/her everyday environment. The assessment is based upon information provided by the child’s parent(s) in response to 10 probes, assessing three main areas: 1) vocalisation behaviour, 2) alerting to sounds; and 3) deriving meaning from sound;
  - **Infant Listening Progress Profile (ILIP):** a profile specifically devised to monitor changes in early auditory performance of young implanted children. The profile covers a range of abilities from first response to environmental sounds, through discrimination of environmental sounds and voice, to identification of own names. As the IT-MAIS, the ILIP also investigates where and when children use their CIs or hearing aids in everyday life;
  - **Categories of Auditory Performance (CAP):** comprises a hierarchical scale of auditory-perceptual abilities, the lowest level describing no awareness of environmental sounds, through awareness and discrimination of speech sounds, and the highest level being represented by the ability to use a telephone with a known speaker;
  - **Ling Six Sound Test (SST):** explores the ability to detect six sounds (/m/, /l/, /u/, /i/, /s/, /f/) whose spectrographic characteristics provide an estimate of proper auditory perception throughout the whole speech frequency range.

The assessment of auditory-perceptual skills was obtained by a speech therapist in auditory-verbal mode, in a silent environment and in the presence of the patient’s parents. Through administration of this test battery the so-called **Basic Perceptual Milestones Achievement (BPMA)** was identified. According to these authors, the BPMA is defined as the acquisition of certain auditory skills to obtain a “minimum” score in each of the aforementioned tests at the same time: 22 points of 40 for the IT-MAIS, 12 of 16 for the ILIP, 4 of 7 for the CAP and 6 of 6 for the SST. This set of scores, in the whole, is the minimum and indispensable auditory prerequisite needed to be acquired to develop proper oral language. Conversely, a failure to achieve one of these skills after at least one year of continuous CI use is almost invariably associated with delayed language skills, even after correcting performances for auditory age.

Statistical analysis was conducted using the Epinfo software. Each outcome variable was compared across groups using a parametric statistic test (Student’s t-test); alpha error was set at 0.05.

**Results**

In the unaided condition, Group 1 children showed a PTA 0.25-0.5 kHz = 89.41 ± 2.19 dB and click-evoked auditory brainstem responses ≥107 dBHL, while Group 2 patients had a PTA 0.25-0.5 kHz = 88.41 ± 2.87 dB (p = 0.18), again having click-evoked auditory brainstem responses ≥107 dBHL.

At time 0 (pre-implant evaluation), there were no statistically significant differences (p > 0.05) between groups compared to the scores obtained for IT-MAIS (3.38 ± 0.76 vs. 4.67 ± 0.76 for Group 1 and 2, respectively; p = 0.24), ILIP (3.29 ± 0.73 vs. 5.28 ± 0.73, respectively; p = 0.06), SST (1.10 ± 0.32 vs. 1.89 ± 0.38, respectively; p = 0.12) and CAP (0.67 ± 0.19 vs. 1.17 ± 0.19, respectively; p = 0.06).

At time 1 (6 months after unilateral cochlear implantation), scores were statistically different between groups for all but one test: IT-MAIS (17.14 ± 2.04 for Group 1 vs. 21.67 ± 1.48 for Group 2; p = 0.08), ILIP (10.38 ± 0.91 vs. 13.44 ± 0.44 for Group 1 and 2, respectively; p = 0.005), SST (4.52 ± 0.41 vs. 5.78 ± 0.13, respectively; p = 0.007) and CAP (2.29 ± 0.28 for Group 1 vs. 3.39 ± 0.22 for Group 2; p = 0.003). On the other hand, the free field hearing gain did not differ between groups (PTA 0.5-3 kHz...
Advantages of bimodal stimulation

At final evaluation, 1 year after cochlear implantation (time 2), there was still a statistically significant difference, although less powerful, between groups for two of four tests. Monaurally implanted children’s auditory-perceptive skills were similar to or worse than those achieved by Group 2 children: IT-MAiS (25.71 ± 2.21 vs. 29.44 ± 1.71 for Group 1 and 2, respectively; p = 0.19), IL-IP (13.76 ± 0.65 vs. 15.39 ± 0.27, respectively; p = 0.02), SST (5.62 ± 0.2 vs 6.00 ± 0.00, respectively; p = 0.07) and CAP (3.67 ± 0.33 vs. 4.44 ± 0.17, respectively; p = 0.04). Again, the hearing gain was not statistically different between groups (PTA 0.5-3 kHz = 35.52 ± 1.37 dB for Group 1 and PTA 0.5-3kHz = 34.39 ± 1.29 dB for Group 2; p = 0.55). Figure 1 shows pure tone audiometry for each group in aided conditions at T2. Figure 2 shows the results obtained for each test/questionnaire at time 0, 6 and 12 months after cochlear implantation.

The BPMA was also evaluated. In Group 1, 6 of 21 children (28.57%) reached BPMA after 6 months of CI use, while another 10 patients (47.63%, total 72.20%) reached the BPMA after 1 year from the hook up; the remaining 5 children (23.80%) did not obtain the scores required for each of the tests even at T2. In Group 2, 9 of 18 children (50%) showed a BPMA at T1, the other 8 (44.45%, total 94.45%) at T2 and only 1 patient (5.55%) did not get the scores required for the BPMA (Fig. 3).

Discussion

Auditory-perceptual skills represent a cornerstone for language development in children. These are essential for auditory feedback, proper voice self-monitoring and the subsequent acquisition of oral language. In the hearing-impaired child, the auditory-perceptual skills need to be reset/acquired through CIs and/or hearing aid use and activated by speech therapy and rehabilitation training to families.

Given the burgeoning interest of the scientific community for the auditory-perceptual skills related to language development in children, in this prospective randomised study we sought for a “springboard”, namely the acquisition of a minimum level of skills such as to expect verbal language to develop in cochlear implant patients. The first milestone is the detection of all of Ling Six Sounds, thus confirming the access to all speech frequencies. Continued use of the implant and the inclusion of the child in a stimulating sound environment (as assessed by the IT-MAiS and ILIP) allow proper exposure to the world of sounds, which is crucial for verbal language learning; the same applies to the ability to discriminate among different environmental sounds (explored by the CAP) which enable acquiring the auditory prerequisites necessary for language development.

In our clinical practice, we considered that the BPMA, that is the simultaneous achievement of certain scores in each of the test from our battery, represents the starting point to develop a proper verbal language.

Until a few years ago, the scientific literature unanimously agreed in considering only one ear to be completely sufficient for the acquisition of auditory-perceptual skills and the subsequent development of language in children with preverbal hearing loss. Thus, it did not seem worth the effort to have guidelines based on solid scientific evidence about the management of the ear contralateral to the implant. However, this lack of evidence has created great heterogeneity in the postoperative management of unilaterally implanted children: in most cases, the issue is still assigned to the clinical orientation of each tertiary care centre or left to the initiative of the parents of the
Fig. 2. Box and whisker plots for each of the two groups for each test/questionnaire administered. Statistically significant differences are represented by * (p < 0.05) and ** (p < 0.01).
Advantages of bimodal stimulation

child, but there are still many in the field who do not encourage bimodal stimulation, believing it may be even confusing, for the two different strategies of stimulation of the acoustic nerve (electric vs. acoustic).

In fact, it is known from the literature that unilateral deafness does not undermine hearing acuity in normal listening, having no influence on the tonal threshold in free field conditions in a quiet environment. This is consistent with our results, since free field hearing thresholds between the two study groups did not differ significantly in spite of contralateral hearing aid use. However, when a residual hearing is present, a conventional acoustical amplification provides an advantage in terms of signal processing, since starting from an initial assessment (which did not differ significantly between groups, confirming the enrollment of two homogeneous cohorts of patients) T1 and T2 evaluations documented significantly better scores in Group 2. One possible explanation could be that binaural stimulation of the auditory system through bimodal stimulation promotes the central integration of the stimulus and supports the acquisition of auditory perceptual skills. Thus, “keeping awake” the contralateral auditory pathway, though not crucial in determining auditory acuity, guarantees perceptual benefits compared with no use of the device. The difference between the two groups in the percentage of children who reached the BPMA 12 months after the initial assessment compared to the evaluation at time 1 is reduced, probably because of a phenomenon of test saturation, as a “ceiling effect”.

Analysing the individual data of children who did not reach the BPMA 12 months after the initial assessment, poor adaptation to the cochlear implant, defined as a discontinuous use or limited to a few hours a day, was observed in all cases. An interesting consideration is the fact that most of these children belonged to Group 1. In this scenario, we assume that the parents might play a role: if family members are convinced of the potential benefits of binaural stimulation of the auditory system, they presumably encourage the use of both devices, and counter the reluctance of their children, helping increase the compliance for a continuous use of both the implant and the hearing aid to a greater extent than parents of children with only one implant. Hence, it confirms the importance of raising awareness as well as informing and involving families about the rehabilitation process of their children.

Conclusions
This study provides preliminary yet convincing data about the benefits arising from the use of contralateral hearing aid in patients with unilateral cochlear implant, consisting of faster and more numerous BPMA than children with only one CI. These findings could represent the initial evidence to establish shared guidelines for better rehabilitation management of patients undergoing cochlear implantation, and possibly provide increasingly credible evidence for bilateral cochlear implantation.

References
6. Grieco-Calub TM, Litovsky RY. Spatial acuity in 2-to-3-


