Title

Bilateral cochlear implantation or bimodal listening in the paediatric population: retrospective analysis of decisive criteria.

Authors

Cleo MC Dhondt[a], Freya KR Swinnen[b], Ingeborg JM Dhooge[a,b]

[a] Department of Ear Nose Throat, Ghent University, De Pintelaan 185 (1P1), B – 9000, Ghent, Belgium

[b] Department of Otorhinolaryngology, Ghent University Hospital, De Pintelaan 185 (1P1), B – 9000, Ghent, Belgium

E-mail: Cleo.Dhondt@UGent.be; Freya.Swinnen@UGent.be; Ingeborg.Dhooge@UGent.be

Corresponding author

Cleo Dhondt, Department of Ear Nose Throat, Ghent University, Ghent, Belgium

E-mail: Cleo.Dhondt@UGent.be; Phone: 00 32 9 332 28 89

Postal address: Ghent University Hospital department Ear Nose Throat, De Pintelaan 185 (1P1), B – 9000 Ghent, Belgium

Conflicts of interest: None

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Abstract

Introduction: In children with bilateral severe to profound hearing loss, bilateral hearing can be achieved by either bimodal stimulation (CIHA) or bilateral cochlear implantation (BICI). The aim of this study was to analyse the audiologic test protocol that is currently applied to make decisions regarding the bilateral hearing modality in the paediatric population.

Methods: Pre- and postoperative audiologic test results of 21 CIHA, 19 sequential BICI and 12 simultaneous BICI children were examined retrospectively.

Results: Deciding between either simultaneous BICI or unilateral implantation was mainly based on the infant's preoperative Auditory Brainstem Response thresholds. Evolution from CIHA to sequential BICI was mainly based on the audiometric test results in the contralateral (hearing aid) ear after unilateral cochlear implantation. Preoperative audiometric thresholds in the hearing aid ear were significantly better in CIHA versus sequential BICI children ($p < 0.001$ and $p = 0.001$ in unaided and aided condition, respectively). Decisive values obtained in the hearing aid ear in favour of BICI were: An average hearing threshold measured at 0.5, 1, 2 and 4 kHz of at least 93 dB HL without, and at least 52 dB HL with hearing aid together with a 40% aided speech recognition score and a 70% aided score on the phoneme discrimination subtest of the Auditory Speech Sounds Evaluation test battery.

Conclusions: Although pure tone audiometry offers no information about bimodal benefit, it remains the most obvious audiometric evaluation in the decision process on the mode of bilateral stimulation in the paediatric population. A theoretical test protocol for adequate evaluation of bimodal benefit in the paediatric population is proposed.

Keywords

Cochlear implant; bimodal; simultaneous bilateral cochlear implantation; sequential bilateral cochlear implantation; pediatric.

1. Introduction

Nowadays, cochlear implantation (CI) is the golden standard in auditory rehabilitation for patients with bilateral severe to profound sensorineural hearing loss. Compared to the rehabilitation with acoustic hearing aids, a CI is more often provided unilaterally [1, 2]. However, compared to monaural stimulation, bilateral stimulation results in more natural hearing, reduced listening effort and improved quality of life [3, 4]. Providing auditory input in both ears is expected to improve speech perception in noise by a combination of the head shadow effect, binaural summation and binaural squelch. The head shadow effect is a bilateral effect, requiring two functional ears. Binaural summation and binaural squelch presume the central auditory system to combine the auditory cues from both ears. In addition, interaural time and level differences available through bilateral auditory stimulation support spatial hearing and sound source localisation in the horizontal plane [5-7]. Stimulation of both ears also prevents neural degeneration resulting from auditory deprivation [8]. Bilateral hearing seems to be of particular importance in children, as research has proved that unilateral hearing loss may be accompanied by behavioural problems, academic difficulties and delays in speech and language development [9, 10].

In patients with bilateral severe to profound hearing loss, bilateral hearing may be achieved by either bilateral cochlear implantation (BICI) or bimodal stimulation. BICI has the advantage that the ear with the best postoperative performance is certainly stimulated electrically [2, 3, 11, 12]. However, the outcome is restricted by the limitations in speech processing strategies of the devices. After all, the electric auditory CI signals predominantly comprise spectral envelope information, whereas the temporal fine structure of sound is discarded. This spectral envelope encoding is sufficient for speech perception in quiet, but for more demanding speech understanding situations the temporal information adds value [13-15].

In bimodal stimulation, electric and acoustic hearing are combined using a CI in one ear and appealing to the residual acoustic hearing in the other ear, if necessary amplified with a hearing aid [6, 12, 16]. This approach includes three major advantages. First, there is no need for a second surgery. Therefore, supplementary costs are avoided and risks concerning both anaesthetics and potential vestibular damage are reduced [2, 11, 12]. Secondly, the contralateral ear remains intact so that it can be engaged for possible new treatments for hearing loss in the future such as stem cell therapy and hair cell regeneration [1, 2, 12, 16]. Finally, in bimodal stimulation, the high-frequency electric hearing is complemented by the low-frequency acoustic input in the contralateral ear, which comprises spectro-temporal information that is lacking in the electric signal [11, 16-18]. This is especially beneficial for segregating voice sources, perceiving voicing information in consonants and perception of sound quality, melody and music [17-20]. However, bimodal stimulation is only a valuable alternative in patients with functional residual hearing [2, 6, 12, 16].

Both bimodal stimulation and BICI are considered effective approaches to provide bilateral hearing, since the majority of recent studies agree that no significant differences in speech perception, language development and localisation ability are found between bimodally stimulated patients and BICI users [16, 17, 21-24]. However, their speech perception in noise and localisation abilities remain poor compared to bilateral normal hearing listeners. The two devices, being a hearing aid and a CI or two CIs, function independently and are not aligned in terms of timing and intensity of the signal presentation, which hampers the central processing of auditory input arriving in both ears. Therefore, the benefit of bilateral compared to monaural stimulation in both bimodal and BICI listeners on speech perception in noise and localisation tasks is principally attributed to the head shadow effect, and the real benefit of binaural processing of acoustic cues is questioned [25].

In young children with bilateral profound hearing loss due to meningitis and in patients with Usher syndrome, BICI is advocated [2-4, 12]. Apart from these exceptions, BICI only seems to be considered if the use of a contralateral hearing aid results in insufficient bimodal benefit [2, 6, 12, 17]. The question remains how to determine this bimodal benefit, especially in young children, and to define what is considered sufficient in this regard. As no worldwide standard criteria are currently available concerning BICI candidacy, most CI centres are inclined to appeal to the unilateral candidacy criteria, using, for example, pure tone audiometry [1, 4]. This method is of questionable validity because the expectations of unilateral CI cannot be compared to the desirable outcome of bilateral hearing [1].

The aim of this retrospective study was (a) to evaluate the audiologic test protocol that is currently applied in deciding between bimodal stimulation and BICI in the paediatric CI population in our centre and (b) to determine which factors and audiologic test results are influencing the decision.

2. Materials and methods

2.1. Subjects

From September 1997 until the start of this retrospective study in October 2016, 276 patients have been implanted and followed at the department of Otorhinolaryngology in the Ghent University Hospital. Only patients younger than 12 years of age on the 9th of December 2009 were included in this study, since from that date onwards BICI is reimbursed to patients up to 12 years of age in Belgium. Additionally, patients needed to be stimulated bilaterally, i.e. with BICI or bimodally, from a young age onwards, i.e. before the age of 18 months.

Fifty-two paediatric patients met these inclusion criteria and were divided into three groups. The first group (CIHA) consisted of 21 bimodal listeners (12 males; 9 females) with a mean age of 10.1 years (SD: 4.1). The mean age of implantation was 4.3 years (SD: 3.0). The 19 (9 males; 10 females) sequential BICI users (Seq BICI) switched from bimodal to BICI condition and had a mean age of 9.6 years (SD: 3.7). They received the first implant at a mean age of 3.3 years (SD: 3.0) and the second at a mean age of 5.6 years (SD: 3.5) The third group consisted of 12 children (8 males; 4 females) with a mean age of 3.1 years (SD: 1.6) who received CIs in both ears simultaneously (Sim BICI) at a mean age of 1.0 years (SD: 0.4). The aetiology of the hearing loss is summarised in Table 1. Occurrence of multiple disorders (psychomotor or cognitive retardation, delayed speech and language development, vestibular, respiratory, cardiac, feeding, muscle tension and/or visual disorders) was reported in nine Seq BICI patients, seven CIHA patients, and three Sim BICI patients and showed no statistically significant difference between subject groups ($p > 0.05$, Fisher's exact test). All included patients signed an informed consent form. The study design was approved by the Ghent University Hospital Medical Ethical Committee.

2.2. Audiologic tests

2.2.1. Middle ear evaluation

In order to preclude temporary middle ear pathologies (e.g. middle ear effusion, tympanic membrane perforation), middle ear status was examined by micro-otoscopy every six months. Tympanometry (TympStar, Grason Stadler Inc., MN, USA) was performed before every audiologic measurement. High-frequency tympanometry (1000 Hz) was used in infants younger than nine months of age. From the age of three months, a 226 Hz probe stimulus was applied.

2.2.2. Auditory Brainstem Response (ABR)

Hearing thresholds were determined objectively by means of ABR testing. Wave V thresholds were examined using the Eclipse EP25 (software Otoaccess version 1.2.1, Interacoustics, Assens, Denmark) using insert phones calibrated according to ISO-389 reference values (E-A-RTONE Insert Earphone 3A ABR, 3M Company, Indianapolis, IN, USA). In clinical practice, besides click stimuli, toneburst stimuli are commonly used. In this database only thresholds using click stimuli were included as these provide a general overview of the child's hearing status. Assessment and interpretation of the measurements was performed by an audiologist out of a fixed team of four audiologists with at least five years of experience in the neonatal and paediatric audiologic diagnostics.

2.2.3. Subjective hearing evaluation

Subjective hearing evaluation included pure tone audiometry, speech audiometry and phoneme discrimination. These tests were performed in the same double-walled sound-attenuated audiometric test room. Depending on the measurement condition, stimuli were presented through headphones (TDH-39, Interacoustics, Assens, Denmark), insert phones (E-A-RTONE Insert Earphone 5A, 3M Company, Indianapolis, IN, USA) or a free-field loudspeaker in front of the listener (Canton Elektronik GmbH, Weilrod, Germany), all calibrated according to ISO-389 reference values. Since 2012, the PC-based audiometer Equinox 2.0 with Otoaccess software version 1.2.1 (Interacoustics, Assens, Denmark) was used. Before, audiometry was performed with the AC 40 clinical audiometer (Interacoustics, Assens, Denmark). Audiologic assessment in the paediatric population was executed and interpreted by two audiologists out of a fixed team of four audiologists with at least five years of experience in the paediatric field. Depending on the cooperation and concentration abilities of the child, some measurements were split up into multiple short sessions.

2.2.3.1. Pure tone audiometry

Pure tone audiometry was executed using pure tone stimuli presented through insert phones or headphones (in unaided condition) or using warble tones presented in free field through a loudspeaker (in aided condition). Depending on the child's age, Behavioural Observation Audiometry (BOA), Visual Reinforcement Audiometry (VRA), Instrumentation Conditioned Reflex Audiometry (ICRA), as well as standard pure tone audiometry were employed. In case of VRA or ICRA, conditioning preceded the test procedure and was regularly repeated throughout the test to check the child's attentiveness to the auditory stimuli. Thresholds above the technical limits of the equipment were registered as 120 dB HL. The degree of hearing loss was represented by the BIAP (Bureau International d'Audiophonologie), which is the average hearing threshold measured at 0.5, 1, 2 and 4 kHz.

2.2.3.2. Speech audiometry

Speech audiometry was performed in aided condition with Dutch monosyllabic word lists. According to the age and the speech and language skills of the subjects, Göttinger I (3-4y), Göttinger II (5-6y), NVA child and NVA lists were administered as speech stimuli [26]. In the majority of cases, the ICA (Indice de Capacité Auditive) was assessed. Word lists were therefore successively presented at 70, 55 and 40 dB SPL.

2.2.3.3. Speech-in-noise (SPIN) testing

SPIN testing was performed with speech and noise presented from the loudspeaker in front of the listener. A different test setup was applied depending on the age, the acquired speech and language skills, and the cooperation level of the child. The signal-to-noise ratio, the presented word list and the examined condition (with CI, with hearing aid alone or in bimodal mode) varied among subjects.

2.2.3.4. Auditory Speech Sounds Evaluation (A§E) – phoneme discrimination

The A§E phoneme discrimination test (Otoconsult, Antwerp, Belgium) was used as an additional test to examine functional hearing [27]. Stimuli were presented through the loudspeaker in front of the listener at 70 dB HL, as the phoneme discrimination was only assessed in aided condition. In very young children, the methods of VRA and ICRA were implemented in order to maximize their cooperation. Conditioning preceded the test and was repeated throughout the test procedure to check the child's attentiveness.

2.3. Data analysis

Statistical analysis was performed using SPSS version 20 (SPSS Inc., Chicago, IL, USA). A significance level of 0.05 was used. Since included variables were not normally distributed, nonparametric tests were applied. Comparison between the three subject groups was done by the Kruskal-Wallis test. The Mann-Whitney U test was used for between-groups comparison with Bonferroni correction (α=0.017) for multiple comparisons. Finally, in the Seq BICI children, the Wilcoxon signed-rank test was applied to compare the pre- and postoperative test results in the second implanted ear.

3. Results

3.1. Audiometric thresholds in the three test groups before first implantation

In the CIHA children, the median (preoperative) BIAP was 100 dB HL (interquartile range (IQR): 91-110 dB HL) in the first implanted ear (Ear 1) and 88 dB HL (IQR: 79-98 dB HL) in the contralateral ear (Ear 2). The Seq BICI children showed a median preoperative BIAP of 108 dB HL (IQR: 100-115 dB HL) in Ear 1 and 99 dB HL (IQR: 88-110 dB HL) in Ear 2. Finally, in the Sim BICI children, we found a median BIAP of 120 dB HL (IQR:107-120 dB HL) in Ear 1 and 120 dB HL (IQR: 95-120 dB HL) in Ear 2.

Preoperative audiometric test results in the three defined groups are summarised and compared in Table 2. Statistical comparisons revealed statistically significant differences for ABR thresholds and BIAP thresholds of both ears (Kruskal-Wallis test, $p < 0.05$). Between-groups comparisons revealed significantly higher ABR thresholds for Ear 2 and BIAP thresholds for both ears in the Sim BICI children compared to the CIHA group (see Table 2 for p values; Mann-Whitney U test with Bonferroni correction). In addition, significant differences were found in the (preoperative) BIAP thresholds of Ear 2 between the CIHA and the Seq BICI children (Mann-Whitney U test with Bonferroni correction).

3.2. Deciding between bimodal stimulation or sequential BICI after first implantation

Table 3 provides an overview of the available test results, playing a role in the decision regarding sequential bilateral implantation versus continued bimodal listening. In 85.7% of CIHA patients (18/21) and 63.2% of Seq BICI patients (12/19), at least one aided discrimination test (speech audiometry or A§E phoneme discrimination) was executed in the implanted Ear 1. Aided discrimination testing in the contralateral non-implanted Ear 2 was executed in 71.4% of CIHA patients (15/21) and 42.1% of Seq BICI patients (8/19). SPIN testing was performed in 9.5% of CIHA patients (2/21) at the age of 5;11 and 11;8 years, respectively. In the Seq BICI group, this was the case in 15.8% (3/19), at the ages of 8;2, 10;9 and 11;3 years.

After CI in Ear 1, no significant differences could be demonstrated in aided audiometric thresholds or speech discrimination with the CI between the CIHA and Seq BICI group (Mann-Whitney U test). Comparison of the audiometric test results in Ear 2 between the CIHA and Seq BICI group resulted in significant differences. Statistical analysis demonstrated that unaided and aided BIAP thresholds ($p < 0.001$ and $p = 0.001$, respectively), aided ICA scores ($p = 0.024$) and aided phoneme discrimination scores ($p = 0.015$; Mann-Whitney U test) were significantly better in the CIHA group compared to the Seq BICI group. Figures 1A and 1B show the boxplots of the unaided and aided BIAP results in Ear 2 for both the CIHA group and the Seq BICI group before implantation in this ear was considered. Similarly, boxplots of the aided ICA scores and the aided A§E discrimination scores obtained with Ear 2 in both groups are displayed in Figures 1C and 1D. Decisive values between both listening modes were determined retrospectively by visual deduction and are indicated by dashed lines on the graphs.

3.3. Final evaluation of hearing outcome

After implantation in Ear 2, the Seq BICI children obtained a significantly improved aided BIAP (median: 29 dB HL; IQR: 21-32 dB HL) compared to the preoperative aided BIAP with hearing aid (median: 58 dB HL; IQR: 51-69 dB HL) (Wilcoxon signed-rank test, $p = 0.001$).

Comparison of these pre- and postoperative aided thresholds was made in 15 patients and is represented in Figure 2A.

Pre- and postoperative aided outcomes for speech audiometry and A§E phoneme discrimination in the Seq BICI children are depicted in Figures 2B and 2C, respectively. Paired comparisons of the ICA in Ear 2 with hearing aid (median: 23%; IQR: 20-47%) and the ICA in Ear 2 with CI (median 72%; IQR: 58-79%) revealed a statistically significant improvement (Wilcoxon signed-rank test, $p = 0.028$). Aided A§E phoneme discrimination scores in Ear 2 also improved significantly from hearing aid (median: 57%; IQR: 29-71%) to CI condition (median: 94%; IQR: 86-100%) (Wilcoxon signed-rank test, $p = 0.046$).

Comparison of ICA scores in Ear 2 between the three groups of bilateral stimulated children is illustrated in Figure 3. Overall comparison showed a significant difference in speech perception outcomes between the three groups (Kruskal-Wallis, $p < 0.05$), but *p* values were not significant when performing the Mann-Whitney U test with Bonferroni correction for between-groups comparisons.

4. Discussion

Although the surplus value of bilateral hearing in the paediatric population is well-documented and generally accepted, the choice for simultaneous BICI, sequential BICI or bimodal listening is not always straightforward. A retrospective analysis reveals which factors and test results have been of interest in the selection process for BICI in our centre, and the final outcome is evaluated. A theoretical test protocol that could be applied in the decision process between bimodal stimulation and sequential BICI is proposed.

4.1. Simultaneous BICI

In infants with bilateral severe to profound hearing loss, the aetiology of the hearing loss and the degree of residual hearing seem to be important factors in the decision for simultaneous

BICI. This is preferred over sequential BICI in infants with limited or no residual hearing since it promotes normal-like symmetric development of the central auditory pathways and offers the greatest benefit of binaural hearing [28]. However, this does not imply that all children with limited or no residual hearing are simultaneously bilaterally implanted by default. The aetiology of the hearing loss is always taken into account. In pathologies with a highly unpredictable postoperative auditory outcome, simultaneous BICI is seldom performed. In bilateral hearing loss due to an unknown aetiology, and in hearing loss associated with a cochlear nerve abnormality or multiple disorders, clinicians would prefer to await the evolution of auditory performance with the first implant before considering contralateral implantation [29, 30]. The present study seems to confirm the stated hypothesis, since simultaneous BICI was performed in only two of all patients with an unknown aetiology (n=17), and in none of the patients with auditory neuropathy/auditory dyssynchrony (n=4) or cochlear hypoplasia (n=1). On the other hand, the choice for BICI might be quite straightforward in patients with a stable, nonsyndromic bilateral severe to profound hearing loss, for example, caused by *GJB2* mutations (connexin 26, Cx26). In such cases, the decision between bimodal listening or BICI is mainly depending on the degree of residual hearing or the bimodal benefit. In our centre, an important number of Cx26 patients (55.6%) were simultaneously bilaterally implanted. In bilateral deafness caused by meningitis and associated with an increased risk of bilateral ossification of the cochlea, the decision for simultaneous BICI is straightforward as well. Research has shown that in these cases, surgery is advisable at an early stage, prior to the onset of cochlear ossification [31]. In the present study, simultaneous BICI was performed in three out of four meningitis patients.

4.2. Bimodal listening or sequential BICI: audiologic protocol

In case the decision on simultaneous BICI is not straightforward, a more conservative approach is advised in which contralateral implantation is only considered if bimodal stimulation results in insufficient benefit [2, 6, 12, 17]. However, the main research question remains how to determine this benefit in clinical practice. The goal of this study was to provide an overview of the test protocol applied in our centre and to investigate which audiologic test results are decisive regarding the bilateral stimulation mode.

Three components are distinguished in the theoretical audiologic test protocol: evaluation of the monaural auditory performance, the bilateral auditory performance and the bimodal gain. Concerning the monaural hearing performance, auditory detection by means of pure tone audiometry seems to remain the most obvious audiometric evaluation, as this test was executed in a larger amount of patients compared to the tests evaluating functional hearing such as A§E phoneme discrimination and speech audiometry. Although pure tone audiometry in the paediatric severely hearing-impaired population may be a time-consuming and laborious procedure requiring experienced audiologists, extensive conditioning of the child, multiple test sessions and age-appropriate vision and motor skills of the child, it remains more feasible compared with A§E phoneme discrimination or speech audiometry. In addition, speech audiometry demands a certain level of cognitive development and language acquisition. The latter is often delayed or impaired in profoundly hearing-impaired children. Since the behaviourally obtained hearing thresholds have a poor sensitivity and specificity in the paediatric population, they should be cross-checked with objective measurements such as ABR or auditory-steady-state responses (ASSR). However, these techniques are infrequently used in the regular follow-up of toddlers or pre-schoolers due to the frequent need for anaesthesia and the difficulties to measure aided thresholds. Another objective technique to confirm the subjective thresholds without the necessity of sedation is the registration of Cortical Auditory Evoked Potentials (CAEP) [2]. CAEP testing can be executed reliably in young infants using tonal and speech stimuli in both aided and unaided condition [32]. Research has shown that P1 latency potentials measured by CAEP in hearing impaired children differ from the potentials found in children with normal hearing. If effective, auditory rehabilitation by means of conventional hearing aids or CI involves a gradual disappearance of these differences [33]. Therefore, CAEP testing appears to be a promising approach for evaluating functional hearing and hearing aid success objectively in children who are very young and/or difficult to test behaviourally [32]. However, CAEP measurements are also affected by many factors, such as sleep state and motor activity, which are difficult to be controlled for in infants and young children.

Besides the evaluation of monaural auditory performance, bilateral performance should be examined as well. Insufficient bilateral performance in bimodal condition could be an argument in favour of contralateral implantation. In this respect, SPIN testing should be included, as better speech perception in noise is one of the main advantages of bilateral hearing [6, 7, 24]. Our retrospective analysis revealed that SPIN was tested in only 9.5% and 15.8% of the CIHA and the Seq BICI subjects, respectively. As the youngest subject in whom SPIN testing was executed, was 5;11 years old, these low rates could be related to the aforementioned required levels of cognitive processing and language development. Additionally, determining reliable audiometric thresholds and evaluating speech perception in quiet in children may already be that time-consuming and exhausting that SPIN testing is often omitted. The evaluation of sound localisation and listening effort are also indispensable in the evaluation of bilateral auditory performance [4, 5]. However, these tests are currently not implemented in our decision-making evaluation between bimodal listening and bilateral CI. Sound localisation can be examined from the age of four years, as described by Van Deun et al. [34]. A dual task paradigm, which is feasible at school age, could be applied to evaluate listening effort [35-38].

The third component of the test protocol should comprise an evaluation of the bimodal gain. As already mentioned in the introduction, this is the most difficult component of the test protocol, as it lacks clarity in literature. It seems evident that the evaluation of bimodal gain should comprise an evaluation of the audiologic advantages of bimodal stimulation. These include better segregation of voice sources, better perception of sound quality, melody and music, and the preservation of low-frequency spectro-temporal information required for better speech perception in noise and low-frequency pitch perception [17-20]. However, evaluation of these bimodal advantages in the paediatric population is not evident. Therefore, in many CI centres, evaluation of these skills is not included in the test protocol. A survey by Schwartz et al. [1] demonstrated that less than half of the CI centres used hearing performance in background noise and even less than ten percent used localisation tasks as methods to determine candidacy for BICI.

Choosing between either continued bimodal listening or evolving to sequential BICI does not seem significantly influenced by auditory CI performance after first implantation, but rather by unaided and aided audiologic test results in the non-implanted ear between test groups. In an attempt to define decisive values in this respect, the boxplots represented in Figures 1A-D were applied. Regarding the unaided BIAP threshold measured in Ear 2, in 75% of the CIHA subjects, a BIAP threshold below 93 dB HL was recorded, whereas in 75% of the Seq BICI patients, a BIAP threshold above this value was measured. Similarly, 52 dB HL can be determined as threshold regarding the aided BIAP. A score of 70% on the A§E phoneme discrimination test and an ICA score of 40% can be defined as dividing values as well.

An important additional consideration in the decision between simultaneous BICI, sequential BICI or continuing bimodal listening is the appropriate functioning of the vestibular system. Since a few years ago, we have implemented a vestibular function evaluation (comprising the Cervical Vestibular Evoked Myogenic Potential test at least) as a standard assessment prior to and after CI in the paediatric population. Interpretation of these vestibular test results is beyond the scope of this study, but it is hypothesized that clinicians could advise against contralateral implantation in case a vestibular response in the implanted ear is absent after CI due to a pre-existing absent response or vestibular damage caused by the surgical procedure. Impairment of the contralateral vestibular system after contralateral implantation would imply a total loss of the vestibular function, causing an invalidating impact on the child's daily functioning [39].

In the Appendix, a test protocol is proposed that should provide a complete representation of the audiologic performance in bimodally stimulated patients. Note that this test protocol is a theoretical proposal. Restrictions in time, therapy loyalty, motivation and other influencing patient characteristics are not taken into account. Therefore, the development of a practical time-effective test protocol, resulting in a complete reflection of the audiologic performance of bimodally stimulated patients should be the focus of further research. Furthermore, a more objective and numeric definition (in terms of test results) of 'insufficient' bimodal gain is urgently required.

4.3. Outcome evaluation

In the Seq BICI patients included in this study, choosing contralateral implantation did result in the best audiologic outcome (Fig. 2A-C). However, it is noteworthy that conclusions concerning the elimination of possible bimodal advantages are lacking as these are currently not evaluated.

Although not significant in this study, Sim BICI patients seemed to achieve higher monaural speech perception scores compared to the monaural speech perception scores in Ear 2 of Seq BICI patients (Fig. 3). It is generally accepted that a long inter-implant interval in sequential BICI has a negative impact on auditory performance with the CI and on linguistic development, due to asymmetric development of the central auditory pathways [40]. Since our Seq BICI patients consistently used acoustic amplification before receiving their second implant, the impact of the inter-implant delay might have been restricted.

5. Conclusion

Bilateral hearing in the severe to profound hearing impaired paediatric population could be achieved by either bimodal listening or BICI, either simultaneous or sequential. The decision between both approaches is influenced by multiple factors of which the aetiology of the hearing loss and the amount of residual hearing are the most important. In practice, an estimation of residual hearing and bimodal benefit in the paediatric population is often based on pure tone audiometry and to a lesser extent on speech audiometry and A§E phoneme discrimination. As the latter require higher cognitive processing and good conditioning, respectively, they can only be executed reliably in older patients. In this respect, CAEP testing appears to be a promising approach for evaluating hearing aid success objectively in children who are very young and/or difficult to test behaviourally. Additionally, tests evaluating other bilateral, binaural and bimodal (e.g., music appreciation) advantages should be a part of the test protocol. However, it can be questioned whether this is feasible and/or relevant in the paediatric population.

The retrospective study design, in combination with rather small subjects groups and missing data require a cautious interpretation of the results of this study. Future research with larger and more equally divided subject groups is warranted to allow more general conclusions.

Acknowledgements

The authors thank the parents of the children included in this study for giving their consent to use the test results. Additionally, we want to thank Birgit Philips, Lotte Rombaut, Stephen Vlaeminck, Sandrien Van Vooren and Stefanie Krijger, as well as the colleagues from the rehabilitation centres for their assistance in collecting data.

References

[1] S.R. Schwartz, S.D. Watson, D.D. Backous, Assessing candidacy for bilateral cochlear implants: A survey of practices in the united states and canada, Cochlear implants international. 13 (2012) 86-92.

[2] R.A. Tange, W. Grolman, W.A. Dreschler, What to do with the other ear after cochlear implantation, Cochlear implants international. 10 (2009) 19-24.

[3] E. Offeciers, C. Morera, J. Muller, A. Huarte, J. Shallop, L. Cavalle, International consensus on bilateral cochlear implants and bimodal stimulation, Acta Otolaryngol. 125 (2005) 918-919.

[4] B.R. Peters, J. Wyss, M. Manrique, Worldwide trends in bilateral cochlear implantation, Laryngoscope. 120 Suppl 2 (2010) S17-44.

[5] M.L. Hawley, R.Y. Litovsky, H.S. Colburn, Speech intelligibility and localization in a multi-source environment, J. Acoust. Soc. Am. 105 (1999) 3436-3448.

[6] M. Mok, K.L. Galvin, R.C. Dowell, C.M. McKay, Spatial unmasking and binaural advantage for children with normal hearing, a cochlear implant and a hearing aid, and bilateral implants, Audiol. Neurootol. 12 (2007) 295-306.

[7] L.W. Welsh, J.J. Welsh, L.F. Rosen, J.E. Dragonette, Functional impairments due to unilateral deafness, Ann. Otol. Rhinol. Laryngol. 113 (2004) 987-993.

[8] S.A. Gelfand, S. Silman, Apparent auditory deprivation in children: Implications of monaural versus binaural amplification, J. Am. Acad. Audiol. 4 (1993) 313-318.

[9] J.L. Culbertson, L.E. Gilbert, Children with unilateral sensorineural hearing loss: Cognitive, academic, and social development, Ear Hear. 7 (1986) 38-42.

[10] J.E. Lieu, Speech-language and educational consequences of unilateral hearing loss in children, Arch. Otolaryngol. Head Neck Surg. 130 (2004) 524-530.

[11] B.C. Papsin, K.A. Gordon, Bilateral cochlear implants should be the standard for children with bilateral sensorineural deafness, Curr. Opin. Otolaryngol. Head Neck Surg. 16 (2008) 69-74.

[12] Y.S. Yoon, Y.R. Shin, Q.J. Fu, Clinical selection criteria for a second cochlear implant for bimodal listeners, Otol. Neurotol. 33 (2012) 1161-1168.

[13] R.V. Shannon, F.G. Zeng, V. Kamath, J. Wygonski, M. Ekelid, Speech recognition with primarily temporal cues, Science. 270 (1995) 303-304.

[14] Z.M. Smith, B. Delgutte, A.J. Oxenham, Chimaeric sounds reveal dichotomies in auditory perception, Nature. 416 (2002) 87-90.

[15] F.G. Zeng, K. Nie, G.S. Stickney, Y.Y. Kong, M. Vongphoe, A. Bhargave, C. Wei, K. Cao, Speech recognition with amplitude and frequency modulations, Proc. Natl. Acad. Sci. U. S. A. 102 (2005) 2293-2298.

[16] T.Y. Ching, E. van Wanrooy, H. Dillon, Binaural-bimodal fitting or bilateral implantation for managing severe to profound deafness: A review, Trends Amplif. 11 (2007) 161-192.

[17] H.E. Cullington, F.G. Zeng, Comparison of bimodal and bilateral cochlear implant users on speech recognition with competing talker, music perception, affective prosody discrimination, and talker identification, Ear Hear. 32 (2011) 16-30.

[18] R.J. van Hoesel, Contrasting benefits from contralateral implants and hearing aids in cochlear implant users, Hear. Res. 288 (2012) 100-113.

[19] M.F. Dorman, R.H. Gifford, A.J. Spahr, S.A. McKarns, The benefits of combining acoustic and electric stimulation for the recognition of speech, voice and melodies, Audiol. Neurootol. 13 (2008) 105-112.

[20] Y.Y. Kong, G.S. Stickney, F.G. Zeng, Speech and melody recognition in binaurally combined acoustic and electric hearing, J. Acoust. Soc. Am. 117 (2005) 1351-1361.

[21] R.Y. Litovsky, P.M. Johnstone, S.P. Godar, Benefits of bilateral cochlear implants and/or hearing aids in children, Int. J. Audiol. 45 Suppl 1 (2006) S78-91.

[22] S. Nittrouer, C. Chapman, The effects of bilateral electric and bimodal electric--acoustic stimulation on language development, Trends Amplif. 13 (2009) 190-205.

[23] E.C. Schafer, A.M. Amlani, A. Seibold, P.L. Shattuck, A meta-analytic comparison of binaural benefits between bilateral cochlear implants and bimodal stimulation, J. Am. Acad. Audiol. 18 (2007) 760-776.

[24] J.E. Choi, I.J. Moon, E.Y. Kim, H.S. Park, B.K. Kim, W.H. Chung, Y.S. Cho, C.J. Brown, S.H. Hong, Sound localization and speech perception in noise of pediatric cochlear implant recipients: Bimodal fitting versus bilateral cochlear implants, Ear Hear. 38 (2017) 426-440.

[25] R.Y. Litovsky, Bilateral cochlear implants in children, in: L.S. Eisenberg (2nd Ed.), Clinical management of children with cochlear implants, Plural Publishing Inc., San Diego, 2017, pp. 153-175.

[26] A.J.W.J.D. Bosman, W., Realisatie van een cd voor spraakaudiometrie in vlaanderen, Logopedie en foniatrie. 67 (1995) 218-225.

[27] P.J. Govaerts, K. Daemers, M. Yperman, C. De Beukelaer, G. De Saegher, G. De Ceulaer, Auditory speech sounds evaluation (a§e®): A new test to assess detection, discrimination and identification in hearing impairment, Cochlear implants international. 7 (2006) 92-106.

[28] V. Easwar, H. Yamazaki, M. Deighton, B. Papsin, K. Gordon, Simultaneous bilateral cochlear implants: Developmental advances do not yet achieve normal cortical processing, Brain Behav. 7 (2017) e00638.

[29] C.E. Corrales, J.S. Oghalai, Cochlear implant considerations in children with additional disabilities, Curr Otorhinolaryngol Rep. 1 (2013) 61-68.

[30] N.M. Young, F.M. Kim, M.E. Ryan, E. Tournis, S. Yaras, Pediatric cochlear implantation of children with eighth nerve deficiency, Int. J. Pediatr. Otorhinolaryngol. 76 (2012) 1442-1448.

[31] M. Durisin, A. Buchner, A. Lesinski-Schiedat, S. Bartling, A. Warnecke, T. Lenarz, Cochlear implantation in children with bacterial meningitic deafness: The influence of the degree of ossification and obliteration on impedance and charge of the implant, Cochlear implants international. 16 (2015) 147-158.

[32] S. Purdy S.; Kelly, Cortical auditory evoked potential testing in infants and young children, The New Zealand Audiological Society Bulletin. 11 (2001) 16-24.

[33] M.F. Dorman, A. Sharma, P. Gilley, K. Martin, P. Roland, Central auditory development: Evidence from caep measurements in children fit with cochlear implants, J. Commun. Disord. 40 (2007) 284-294.

[34] L. Van Deun, A. van Wieringen, F. Scherf, N. Deggouj, C. Desloovere, F.E. Offeciers, P.H. Van de Heyning, I.J. Dhooge, J. Wouters, Earlier intervention leads to better sound localization in children with bilateral cochlear implants, Audiol. Neurootol. 15 (2010) 7-17.

[35] S. Choi, A. Lotto, D. Lewis, B. Hoover, P. Stelmachowicz, Attentional modulation of word recognition by children in a dual-task paradigm, J. Speech. Lang. Hear. Res. 51 (2008) 1042-1054.

[36] C.B. Hick, A.M. Tharpe, Listening effort and fatigue in school-age children with and without hearing loss, J. Speech. Lang. Hear. Res. 45 (2002) 573-584.

[37] C.S. Howard, K.J. Munro, C.J. Plack, Listening effort at signal-to-noise ratios that are typical of the school classroom, Int. J. Audiol. 49 (2010) 928-932.

[38] K.C. Hughes, K.L. Galvin, Measuring listening effort expended by adolescents and young adults with unilateral or bilateral cochlear implants or normal hearing, Cochlear implants international. 14 (2013) 121-129.

[39] A. Inoue, S. Iwasaki, M. Ushio, Y. Chihara, C. Fujimoto, N. Egami, T. Yamasoba, Effect of vestibular dysfunction on the development of gross motor function in children with profound hearing loss, Audiol. Neurootol. 18 (2013) 143-151.

[40] M. Lopez-Torrijo, S. Mengual-Andres, R. Estelles-Ferrer, Clinical and logopaedic results of simultaneous and sequential bilateral implants in children with severe and/or profound bilateral sensorineural hearing loss: A literature review, Int. J. Pediatr. Otorhinolaryngol. 79 (2015) 786-792.

[41] A. van Wieringen, J. Wouters, List and lint: Sentences and numbers for quantifying speech understanding in severely impaired listeners for flanders and the netherlands, Int. J. Audiol. 47 (2008) 348-355.

[42] R. Kang, G.L. Nimmons, W. Drennan, J. Longnion, C. Ruffin, K. Nie, J.H. Won, T. Worman, B. Yueh, J. Rubinstein, Development and validation of the university of washington clinical assessment of music perception test, Ear Hear. 30 (2009) 411-418.

[43] W.R. Drennan, J.J. Oleson, K. Gfeller, J. Crosson, V.D. Driscoll, J.H. Won, E.S. Anderson, J.T. Rubinstein, Clinical evaluation of music perception, appraisal and experience in cochlear implant users, Int. J. Audiol. 54 (2015) 114-123.

[44] W.B. Cooper, E. Tobey, P.C. Loizou, Music perception by cochlear implant and normal hearing listeners as measured by the montreal battery for evaluation of amusia, Ear Hear. 29 (2008) 618-626.

[45] B. Vaerenberg, A. Pascu, L. Del Bo, K. Schauwers, G. De Ceulaer, K. Daemers, M. Coene, P.J. Govaerts, Clinical assessment of pitch perception, Otol. Neurotol. 32 (2011) 736-741.

Appendix : Proposed test protocol

I. Middle ear evaluation

A. Micro-otoscopy

B. Tympanometry

II. Evaluation of the aided monaural auditory performance (with CI and with hearing aid, separately)

	Behavioural test	Electrophysiological test
A. Auditory detection		
	- Pure tone audiometry	- Auditory Brainstem Response
		- Auditory Steady-State Response
B. Speech sound discrimination		
	- Auditory Speech Sounds Evaluation phoneme discrimination test	- Cortical Auditory Evoked Potentials
C. Speech perception in quiet		
	- Speech audiometry in quiet	

III. Evaluation of the bilateral auditory performance (with CI alone vs with CI and hearing aid)

A. Speech sound discrimination (see above)

B. Speech perception in quiet (see above)

C. Speech perception in noise [41]

D. Sound localisation [34]

E. Listening effort [35-38]

IV. Evaluation of the bimodal gain (with CI alone vs with CI and hearing aid)

A. Segregation of voice sources [17]

B. Perception of sound quality, melody and music [42-44]

C. Preservation of the low-frequency spectro-temporal information

- Speech perception in noise [41]
- Low-frequency pitch perception [45]

V. Evaluation of the vestibular function

A. Cervical Vestibular Evoked Myogenic Potential test

B. Video Head Impulse Test

C. Rotatory test

D. Caloric test

Table 1 Aetiology of hearing loss in the three subject groups

	Total		CIHA		Seq BICI		Sim BICI	
	n=52		*n*=21		*n*=19		*n*=12	
	%	(*n*)	%	(*n*)	%	(*n*)	%	(*n*)
cCMV	23	(12)	10	(2)	42	(8)	17	(2)
Cx26	17	(9)	14	(3)	5	(1)	42	(5)
Bilateral EVA	8	(4)	14	(3)	5	(1)	-	
Meningitis	8	(4)	5	(1)	-		25	(3)
AN/AD	8	(4)	10	(2)	11	(2)	-	
Premature hypoxia	2	(1)	-		5	(1)	-	
Cochlear nerve hypoplasia	2	(1)	-		5	(1)	-	
Unknown - familial	10	(5)	19	(4)	5	(1)	-	
Unknown	23	(12)	29	(6)	21	(4)	17	(2)

CIHA=bimodal listeners; Seq BICI=children with sequential bilateral cochlear implantation; Sim BICI=children with bilateral simultaneous cochlear implantation; cCMV=congenital cytomegalovirus infection; Cx26=connexin 26 gene mutation; EVA=enlarged vestibular aqueduct; AN/AD=auditory neuropathy/auditory dyssynchrony.

Table 2 Comparison of preoperative audiometric thresholds between the three subject groups before implantation in Ear 1

	Median	IQR (Q1 – Q3)	Group *p* value	Between-groups *p* values: CIHA	Seq BICI	Sim BICI
ABR Ear 1 (dB nHL)						
CIHA (n=18)	100	80 – 100		-	>0.05	0.043
Seq BICI (n=16)	100	96 – 100	**0.024***	>0.05	-	>0.05
Sim BICI (n=12)	100	100 – 100		0.043	>0.05	-
ABR Ear 2 (dB nHL)						
CIHA (n=18)	80	69 – 100		-	0.027	**0.001***
Seq BICI (n=16)	95	89 – 100	**0.001***	0.027	-	>0.05
Sim BICI (12)	100	100 – 100		**0.001***	>0.05	-
BIAP Ear 1 unaided (dB HL)						
CIHA (n=21)	100	91 – 110		-	>0.05	**0.001***
Seq BICI (n=18)	108	100 – 115	**0.004***	>0.05	-	0.035
Sim BICI (n=12)	120	107 – 120		**0.001***	0.035	-
BIAP Ear 2 unaided (dB HL)						
CIHA (n=21)	88	79 – 98		-	**0.016***	**<0.001***
Seq BICI (n=18)	99	88 – 110	**0.001***	**0.016***	-	0.048
Sim BICI (n=12)	120	95 – 120		**<0.001***	0.048	-

IQR=interquartile range; CIHA=group ending up as bimodal listeners; Seq BICI=group of children with sequential bilateral cochlear implantation; Sim BICI=group of children with simultaneous bilateral cochlear implantation; *n*=number of subjects within each test group included in the statistical analysis; ABR=Auditory Brainstem Response; BIAP= Bureau International d'Audiophonologie, which is the average hearing threshold measured at 0.5, 1, 2 and 4 kHz; Ear 1=(first) implanted ear; Ear 2=contralateral ear to Ear 1. Statistically significant *p* values (<0.05 for the Kruskal-Wallis test and <0.017 for the Mann-Whitney U test with Bonferroni correction) are indicated by (*).

Table 3 Available audiometric test results decisive for continued bimodal listening (CIHA) versus sequential bilateral cochlear implantation (Seq BICI)

	CIHA *n*=21		Seq BICI *n*=19	
Ear 1	%	(*n*)	%	(*n*)
PTA aided (CI1)	95	(20)	100	(19)
Speech audiometry aided (CI1)	81	(17)	63	(12)
Speech audiometry bimodal (CI1 + HA)	57	(12)	26	(5)
A§E aided (CI1)	71	(15)	53	(10)
Ear 2	%	(n)	%	(n)
PTA unaided	100	(21)	100	(19)
PTA aided (HA)	100	(21)	89	(17)
Speech audiometry aided (HA)	62	(13)	32	(6)
A§E aided (HA)	52	(11)	37	(7)

The upper part of the table displays the availability of audiometric test results in the implanted ear (Ear 1), obtained in aided condition (with cochlear implant, CI1). The lower part summarizes the audiometric tests undertaken in the (at that moment non-implanted) contralateral, hearing aid ear (Ear 2).
CIHA=bimodal listeners; Seq BICI=sequential bilateral cochlear implant group; PTA=pure tone audiometry; A§E=Auditory Speech Sounds Evaluation phoneme discrimination test; CI=cochlear implant, HA=hearing aid; *n*=number of subjects within each test group.

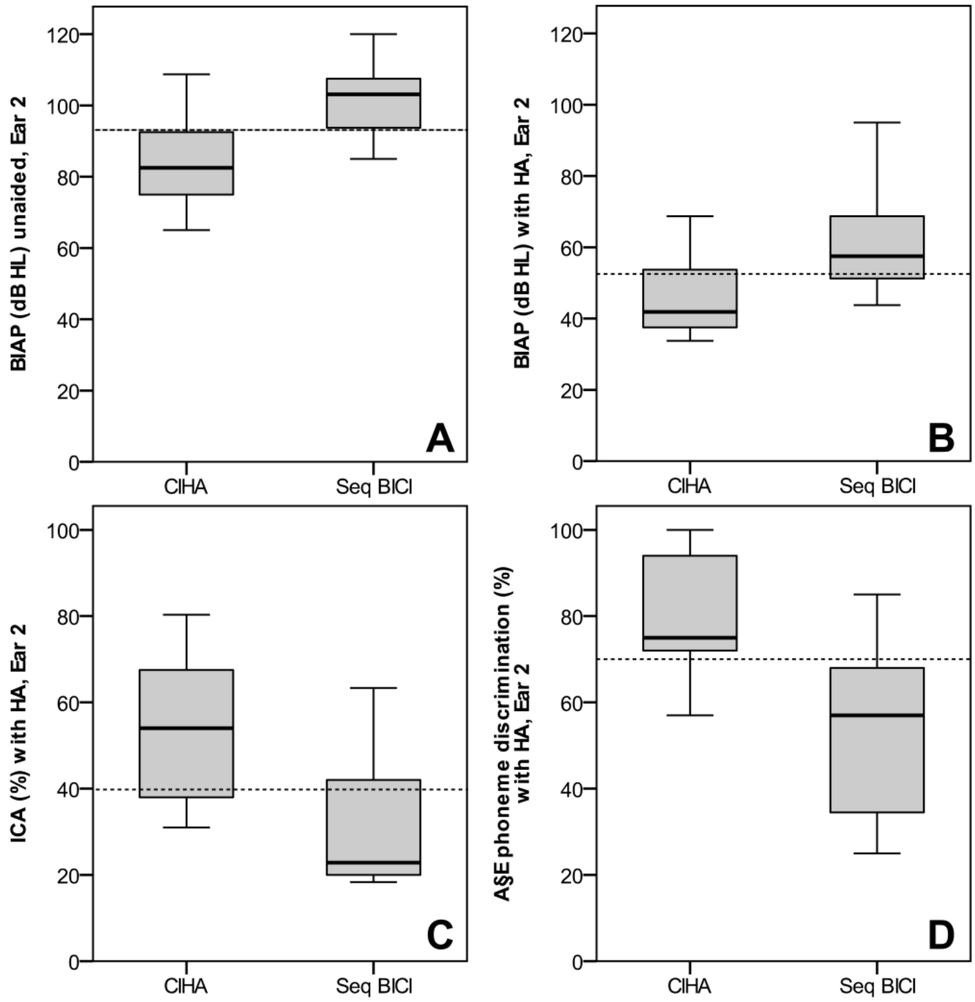

Fig. 1 Boxplots representing test results on pure tone audiometry, speech audiometry and speech discrimination in Ear 2 in CIHA and Seq BICI children: (A) the unaided BIAP threshold (dB HL), (B) the aided (with hearing aid) BIAP threshold (dB HL), (C) the aided (with hearing aid) ICA score (%) and (D) the aided (with hearing aid) A§E phoneme discrimination score (%).The dashed lines indicate a retrospectively determined cut-off value between continuing bimodal listeners (CIHA) and children evolving to sequential bilateral cochlear implantation (Seq BICI). BIAP=Bureau International d'Audiophonologie, which is the average hearing threshold measured at 0.5, 1, 2 and 4 kHz; ICA=Indice de Capacité Auditive, which is the average speech perception score at 70, 55 and 40 dB SPL stimulation level; A§E=Auditory Speech Sounds Evaluation; CIHA=group of bimodal listeners; Seq BICI=group of children with sequential bilateral cochlear implantation; HA=hearing aid.

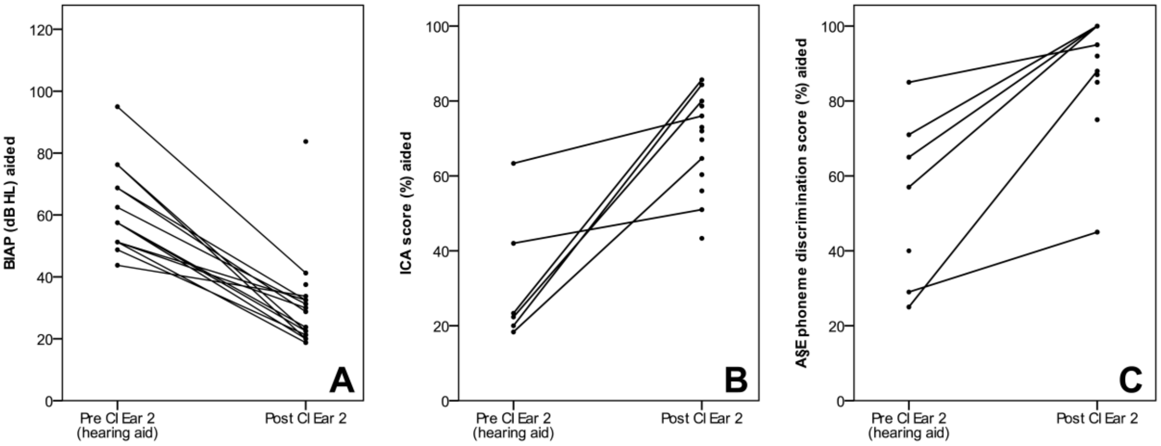

Fig. 2 Paired comparisons of test results on pure tone audiometry, speech audiometry and speech discrimination in hearing aid and CI condition in Ear 2 in sequentially bilaterally implanted patients: (A) BIAP thresholds (dB HL) in 15 patients, (B) ICA scores (%) in 6 patients and (C) A§E phoneme discrimination scores (%) in 6 patients.

The dots represent data from patients in which paired comparison was not possible, as one of both test results was missing. BIAP=Bureau International d’Audiophonologie, which is the average hearing threshold measured at 0.5, 1, 2 and 4 kHz; ICA=Indice de Capacité Auditive, which is the average speech perception score at 70, 55 and 40 dB SPL stimulation level; A§E=Auditory Speech Sounds Evaluation; Pre=preoperative; Post=postoperative; CI=cochlear implant.

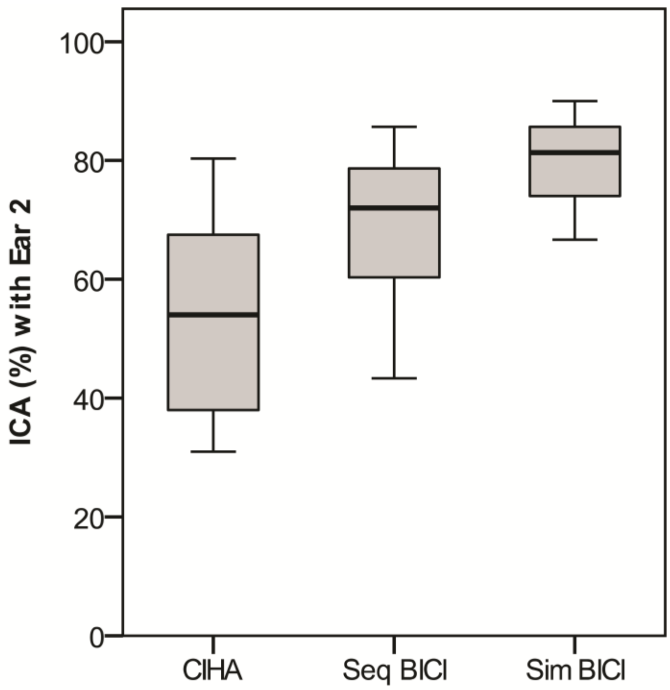

Fig. 3 Boxplot comparing ICA scores (%) in Ear 2 between CIHA (*n*=12, with hearing aid in Ear 2), Seq BICI (*n*=13, with CI in Ear 2) and Sim BICI children (*n*=3, with CI in Ear 2). ICA=Indice de Capacité Auditive, which is the average speech perception score at 70, 55 and 40 dB SPL stimulation level; CIHA=group of bimodal listeners; Seq BICI=group of children with sequential bilateral cochlear implantation; Sim BICI=group of children with simultaneous bilateral cochlear implantation.