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Research Article

Hearing and Oral Language Skill Development in Children with Unilateral and Simultaneous Bilateral Cochlear Implants in the First Year of Device Use

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Abstract

This longitudinal study aimed to verify the advantages of simultaneous bilateral stimulation for the development of hearing and oral language skills over unilateral stimulation in children using cochlear implants, in the first year of electronic device use. The study included twenty children divided into two matched groups (n=10): children using unilateral cochlear implant (UCI), and those using simultaneous bilateral cochlear implant (BCI). The IT-MAIS or MAIS and MUSS questionnaires were used for evaluated language and auditory performance at three, six, and twelve months after activation of the electrodes. A gradual increase in auditory speech perception and oral language development was seen over the first year of device use in both groups; however, there was no significant difference between the two groups. In conclusion, in the first year of cochlear implant use, children using UCI and those using simultaneous BCI showed similar development of auditory perception of speech and oral language.

ABBREVIATIONS

CI: Cochlear Implant; IT-MAIS: Infant-toddler Meaningful Auditory Integration Scale; MAIS: Meaningful Auditory Integration Scale; MUSS: Meaningful use of Speech Scales; UCI: Unilateral Cochlear Implant; BCI: Bilateral Cochlear Implant; AB: Advanced Bionics

INTRODUCTION

Over the past 30 years, cochlear implants (CIs), have become clinically available, owing to the consensus among hearing rehabilitation professionals on its effectiveness in the treatment of hearing loss in children.

For decades, the indication of unilateral CI was the treatment of choice, even in the cases of congenital or acquired bilateral sensorineural hearing loss. The results showed that when restoring the auditory sensation through electrical stimulation in an ear, the child had access to speech sounds, and this condition associated with the therapeutic process, enabled the development of oral language.

However, after several years of using CIs, patients complained frequently of difficulties in speech perception in noisy environments and locating sounds, demonstrating that binaurality is an important condition, especially when considering the school-learning environment. Additionally, research with cortical auditory evoked potentials have shown that unilateral CIs in the first years of life lead to an atypical organization of the auditory cortex, possibly related to the reported difficulties.

In the first decade of the 21st century, the first reports on the indications for bilateral CI used in children were published in the international literature [1-7]. With beneficial results principally in the localization of sound and improvement of auditory perception of speech sounds in noisy situations, the indication for bilateral CIs in children has become customary in primary international CI centers.

The European Bilateral Pediatric Cochlear Implant Forum focused on this with four central themes: early implantation, bilateral implantation, the effect of sequential implantation and simultaneous implantation, and the importance of atraumatic surgery. The consensus from this European Forum is still accepted and states that after the complete and secure diagnosis of bilateral severe or profound sensorineural hearing loss, the child should receive simultaneous bilateral CI promptly to allow optimal hearing development [8].

Literature reviews [9,10], shown that there is a tendency for greater benefit with simultaneous CI compared to sequential bilateral and unilateral CI. However, the results were inconclusive and required further study with greater methodological rigor and analysis of more variables. Studies carried out after have supported the benefits of simultaneous bilateral CIs compared to unilateral through higher verbal intelligence scores correlated with speech perception in noisy environments [11], and sound localization [12].

In this context, it is not yet clear whether the critical age for performing simultaneous CI the perspective of the development of the auditory cortex and consequently the acquisition of auditory and language skills is the similar to that for unilateral cochlear implant.

In a recent study [13], it was observed that there was no significant difference in auditory speech perception in children who had CI before and after the age of 1 year, recommending implantation between 12 and 24 months. This finding also had clinical relevance when considering that simultaneous bilateral CI surgery lasted around 4 hours, with a higher possible surgical risk in children aged under 1 year. This last aspect was rejected [14], when concluding that simultaneous bilateral cochlear implantation in children aged 8 to 61 months, with a mean age of 24 months, is a safe surgical procedure with a better cost-benefit ratio.

However, other important conditions must be considered when simultaneous CIs are indicated, such as children with multiple disabilities or auditory neuropathy, which cases the benefits with the CI are variable. In this situation, the sequential bilateral CI with short interval-interimplant must be the treatment of choice, because is the ultimate limiting disability affecting the child's outcome. Additionally, assuming that simultaneous bilateral CI should be performed promptly, the family may not get the required time to assimilate the diagnosis and understand the obligations related to the CI; for example, the maintenance of two electronic devices. The quality of the intervention in the period between the diagnosis and surgical stage of CI reflects the family's adherence to treatment. Thus, this study aimed to verify the advantages of simultaneous bilateral stimulation for initial hearing and oral language skill development when compared to unilateral stimulation in children with CIs in the first year of use of the electronic device. It also aimed to contribute to establishing a consensus on CI to guide health agencies and implant centers in developing the best practices for the treatment of hearing loss.

MATERIALS AND METHODS

This prospective longitudinal study was carried out at the Cochlear Implant Section of the Hospital of a Public University, Brazil, approved (CAAE: 61745916.1.0000.5441), by the institutional Ethics Committee. Written consent was obtained from the parents/guardians before data collection.

The sample was selected according to the following inclusion criteria: children with pre-lingual hearing loss who had an indication of CI surgery by an interdisciplinary team from the Cochlear Implant section; full insertion of the electrodes, regardless of the device manufacturer; effective use of electronic devices; speech therapy at least once a week for more than 12 months of CI use. Children with neurological impairment and/or a diagnosis of auditory neuropathy spectrum disorder (ANSD), were excluded from the study. All patients underwent magnetic resonance imaging using a Phillips device with 1.0 T magnetic fields.

A total of 20 children with congenital bilateral sensorineural hearing loss who underwent CI surgery participated in this study, divided into two groups: the unilateral CI (UCI) group, consisting of 10 children of both sexes, using unilateral CIs, with an average age of 29.30 ± 10.26 months during CI activation without the effective use of individual hearing aids in the contralateral ear; and the bilateral CI (BCI), group, consisting of 10 children of both sexes, using bilateral CIs with the simultaneous activation of the electrodes, with an average age of 28.20 ± 10.00 months during CI activation. The groups were matched according to their chronological age at electrode activation and the manufacturer and model of internal and external components. Table 1 shows the demographic data of the participants in this study.

Procedures

The children were evaluated at three time points after 3, 6, and 12 months of CI activation. To assess auditory speech perception, the Portuguese adaptation to infant-toddler meaningful auditory integration scale (IT-MAIS) [15], was used for children under 4 years, and meaningful auditory integration scale (MAIS) [16], for those older than 4 years. Both these scales comprise 10 simple questions relating to the child's auditory behavior in different everyday situations, within three different areas of the development of hearing skills: changes in vocalization associated with the use of the device, attribution of meaning to sound, and recognition of environmental sounds.

To evaluate language development, the Portuguese adaptation of oral language assessment questionnaire-meaningful use of speech scales (MUSS) [17], was used. The MUSS questionnaire was used for children between 2 and 5 years of age and was composed of 10 questions related to oral language in daily life situations, in three areas: vocal control, the use of spontaneous speech, and the use of communication strategies in daily situations.

These scales had 10 questions. Each question had a 5-point scale, with scores ranging from 0 (zero) to 4, as follows: 0 = never, 1 = rarely, 2 = occasionally, 3 = often, 4 = always. The result was calculated by the summation of the total number of points accumulated in each question (0–lowest to 4–highest), with the possibility of obtaining a maximum of 40 points. This score was transformed into a percentage, where 100% was the maximum score.

All parent-report scales were applied to parents or guardians in an interview format by the researcher responsible.

Data analysis

Descriptive data analysis was performed using the mean, standard deviation, minimum, and maximum for age at electrode activation (months), and IT-MAIS/MAIS and MUSS in percentages at 3, 6, and 12 months. The Kolmogorov-Smirnov normality test was used to verify the distribution of differences to identify the tests for inferential analysis of the data. The results of the IT-MAIS/MAIS and MUSS questionnaires in both groups showed a normal distribution. Therefore, the Student's t-test was used to compare the IT-MAIS/MAIS and MUSS questionnaire scores at the three different time points. For comparison of the slopes between the unilateral and bilateral CI groups, a mixed effects regression analysis was used. The level of statistical significance used was 5%.

RESULTS AND DISCUSSION

This study aimed to verify auditory and oral language development in children who had bilateral CI with simultaneous activation in the sensitive period at 3 months, 6, and 12 months after electrode activation compared to those with unilateral CI.

Table 2 shows the descriptive statistical analysis of the scores of the IT-MAIS/MAIS and MUSS questionnaires at 3, 6, and 12 months of CI use for both groups.

There was no statistically significant difference between the UCI and BCI groups when comparing the IT-MAIS/MAIS mean scores at 3 ($p = 0.198$), 6 ($p = 0.297$), and 12 months ($p = 0.384$) and the MUSS scale mean scores at 3 ($p = 0.215$), 6 ($p = 0.392$), and 12 months ($p = 0.399$), after CI activation.

Figure 1 shows the change in the IT-MAIS/MAIS and MUSS scores for both groups during the evaluation period.

The IT-MAIS/MAIS and MUSS questionnaire scores increased significantly during the evaluation period for the UCI group (IT-MAIS/MAIS: $b = 3.12$, $t = 3.02$, $p = 0.01$; MUSS: $b = 1.53$, $t = 3.22$, $p = 0.003$), and for the simultaneous BCI group (IT-MAIS/MAIS: $b = 3.36$, $t = 2.48$, $p = 0.02$; MUSS: $b = 1.4$, $t = 3.58$, $p = 0.001$). The rate of change over time was similar between the groups for both tests ($p > 0.05$) (Figure 1).

The results indicated a gradual improvement in the scores of the IT-MAIS/MAIS and MUSS questionnaires in both groups at the three assessment times; however, there was no statistically significant difference between them during the first year of CI use. Thus, simultaneous bilateral stimulation compared with unilateral stimulation in the first year of use did not negatively influence the development of early auditory skills, pre-verbal behaviors, and the onset of oral language acquisition. It is noteworthy that the protocols evaluating this initial phase for young children are scarce, and most of them evaluate the children under the view of parents and professionals.

Our findings corroborate those in the previous study [18], who suggested that there is no difference between the two types of intervention, unilateral or simultaneous bilateral implantation in the first year of device use, from the parents'

Table 1: Demographic data, characterizing the sample according to the type of implantation, etiology/risk factor for hearing loss, age at electrode activation, electronic device manufacturer, internal component model, and speech processor.

Groups	Etiology / Risk factors	Age CI activation (Months)	CI device	Internal component model	Speech processor
UCI 1	Parental Consanguinity	36	Med-El	Sonata Ti 100	Opus 2
UCI 2	Unknown	24	Med-El	Sonata Ti 100	Opus 2
UCI 3	Parental Consanguinity	33	AB	Hires 90K MS	Harmony
UCI 4	Parental Consanguinity	21	Med-El	Sonata Ti 100	Opus 2
UCI 5	Unknown	52	Cochlear	Nucleus CI24	Nucleus 5
UCI 6	Toxoplasmosis	19	Cochlear	Nucleus CI24	Nucleus 5
UCI 7	Unknown	20	AB	Hires 90K MS	Naída
UCI 8	Unknown	23	Med-El	Sonata Ti 100	Opus 2
UCI 9	Unknown	29	Cochlear	Nucleus CI24	Nucleus 5
UCI 10	Unknown	36	AB	Hires 90K MS	Naída
BCI 1	Unknown	36	Med-El	Sonata Ti 100	Opus 2
BCI 2	Meningitis	23	Med-El	Sonata Ti 100	Opus 2
BCI 3	Unknown	29	AB	Hires 90K MS	Naída
BCI 4	Meningitis	21	Med-El	Sonata Ti 100	Opus 2
BCI 5	Unknown	49	Cochlear	Nucleus CI24	Nucleus 5
BCI 6	Family history	19	Cochlear	Nucleus CI24	Nucleus 5
BCI 7	Unknown	16	AB	Hires 90K MS	Naída
BCI 8	Unknown	24	Med-El	Sonata Ti 100	Opus 2
BCI 9	Family history	28	Cochlear	Nucleus CI24	Nucleus 5
BCI 10	Cytomegalovirus	37	AB	Hires 90K MS	Naída

Caption: CI: Cochlear Implant; UCI: Unilateral cochlear implant; BCI: Bilateral cochlear implant; AB = Advanced Bionics.

Table 2: Descriptive analysis of IT-MAIS/MAIS and MUSS scores.

		Groups	N	Mean (SD)	Minim.	Maxim.
Activation CI	Age (months)	UCI	10	29.30 (10.26)	19	52
		BCI	10	28.20 (10.00)	16	49
3 months	IT-MAIS/MAIS (%)	UCI	9	48.28 (23.84)	13	88
		BCI	8	63.56 (24.78)	26	100
	MUSS (%)	UCI	9	13.88 (9.10)	0	27.5
		BCI	8	19.31 (7.235)	12.0	30
6 months	IT-MAIS/MAIS (%)	UCI	10	64.50 (20.06)	33	90
		BCI	10	71.50 (15.28)	55	100
	MUSS (%)	UCI	10	20.95 (8.79)	5	30
		BCI	10	24.75 (6.91)	15	35
12 months	IT-MAIS/MAIS (%)	UCI	10	78.00 (19.03)	47.5	100
		BCI	10	85.00 (17.11)	60.0	100
	MUSS (%)	UCI	10	28.25 (10.93)	10.0	47.5
		BCI	10	32.25 (9.01)	20.0	52.5

Caption: CI: Cochlear implant; UCI: Unilateral cochlear implant; BCI: Bilateral cochlear implant; SD: Standard deviation; Minim: Minimum; Maxim: Maximum

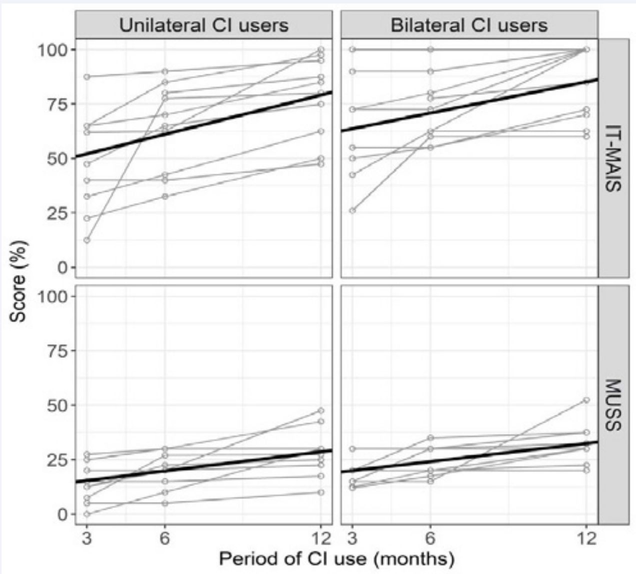

Figure 1 Change in IT-MAIS/MAIS and MUSS scores in evaluation period (Mixed effects regression analysis).

perspective. However, it is very important to emphasize that this development is influenced by different variables. In the first year of hearing, regardless of the type of electrical or acoustic stimulation, the key factors for good development are the child should effectively use the device, the acceptable condition of the device, the environment around the child should be favorable for his/her development, and the child should attend auditory verbal therapy.

The results of our study found that IT-MAIS/MAIS scores were higher in both groups than reported in the literature [19,20], and in the group with bilateral CI 21,22]. However, there was no statistically significant difference between the groups. The impact of simultaneous bilateral CI on speech and language development 23], showed that children who underwent simultaneous bilateral CI between 5 and 18 months of age, had hearing function similar to that of normal hearing peers, after 9 months of use. Further, approximately 81% of children had similar receptive language skills, and 57% of children had similar expressive language, after 12 and 48 months of use, indicating promising long-term results.

Auditory asymmetries detrimental to the processing of temporal aspects of the signal and fundamental binaural cues for locating and distinguishing sounds in noisy environment can be observed in children with unilateral CI. However, these asymmetries can be resolved when simultaneous bilateral surgery is performed, or if a second, surgery is performed within a year and half [24,25]. These second surgeries have a positive effect on speech perception [24,25]. As the age and interval between implants increase, it becomes difficult to redirect cortical structures to their primary function, hearing, because of the long period of sensory deprivation.

As for language development, studies have shown the importance of early communication signals, such as the development of basic pre-lingual skills in the first year of CI use. In particular, skills such as turning towards sounds, crying with different intonations, mimicking sounds, recognizing familiar voices, and understanding words and producing them including onomatopoeic sounds, are predictive of the onset of oral language development. Our results showed that regardless of the type of unilateral or bilateral electrical stimulation, the MUSS questionnaire scores indicate that children who received the device during the sensitive period presented in the first year of use develop pre-lingual auditory skills, which may be predictive of the onset of oral language development, and are important to monitor. The delay in this phase is an indication that immediate intervention is needed, as it has a direct influence on the prognosis of the child's language development [22].

Our findings have relevant clinical applicability. Cochlear implantation in difficult cases such as ANSD, cerebral palsy, or associated neurological disorders sometimes makes the indication more challenging, since the hearing benefits obtained are unclear, given the other existing difficulties. The possibility of implementing the first CI, initiating the therapeutic process, and resuming the discussion for bilateral indication with more information about the results of electrical stimulation in the auditory system and the impact on the child's development, makes the prospect of bilateral implants more convincing. The results support the possibility of sequential bilateral CIs with a short time interval between surgeries in these specific cases, since the auditory and oral language skills were similar between the two groups studied, UCI and BCI.

Currently, simultaneous or sequential bilateral cochlear surgery between surgeries is most acceptable in the clinics as binaurality is critical for speech perception in difficult listening and musicality as well as for oral language development.

Children, who received CI both unilaterally and bilaterally before 24 months, had hearing evolution without statistically significant differences regarding developmental milestones in the first 6 months of hearing age [17]. However, the authors performed a longitudinal follow-up of these children and pointed out that with the use of hearing aid for 5 years, there were statistically significant differences between children regarding auditory development, especially for recognition ability, in which children with bilateral CIs could achieve 100% syllable word recognition and better sentence recognition performance, reflected in better acquisition of socio-linguistic skills [17].

Therefore, after some years of CI use, the type of hearing stimulation may make a difference. The benefits of simultaneous or sequential bilateral CI with a short interval between surgeries facilitate incidental learning. This is because of a greater ability to access oral language in difficult listening situations, such as the school environment, which enables the acquisition of more complex verbal skills [11]. In addition, binaural hearing also reduces auditory effort, enhances child safety, and improves socialization, which are important aspects of child development [11].

However, it is important to highlight that scientific evidence regarding the benefits of simultaneous bilateral CIs in the acquisition of auditory skills and oral language is still scarce and uncertain.

Systematic review of studies published until 2013 [26], found that the results regarding sound perception and expressive language development were statistically favorable for simultaneous bilateral CI when compared to unilateral and sequential CI, even for a short interval. The authors analyzed studies involving children who received CI before the age of 3 years as well as those that compared simultaneous and sequential bilateral CI. However, the study [26] pointed it to be preliminary data, since only a small number of individuals were involved and the studies presented low methodological rigor, with a need for a larger number of randomized controlled trials.

On the other hand, a review of literature until 2014 [9], emphasized that it is possible to achieve binaural skills for children who received simultaneous bilateral CI or short-term sequential bilateral CI, with a strong possibility of these skills being similar to those of normal listeners. However, the data is still weak because of the small number of long-term studies that analyze children with methodological rigor.

Thus, this study can contribute to verifying the auditory and oral language development in the cultural and socioeconomic context of Brazilian children who received bilateral CI with simultaneous activation, compared to unilateral CI, showing that in the first year of use, there are no differences in auditory and language development markers regarding initial skills. This finding had an implication in clinical practice, for the indication of simultaneous or sequential CI with a short period of time between surgeries.

However, binaurality, an essential skill, is achieved only in simultaneous or sequential bilateral CI with a short period of time, to aid children in achieving greater incidental learning, development of binaural auditory skills as well as more complex, memory-oriented processing aids, and verbal intelligence to foster communicative independence and academic performance.

CONCLUSION

In the first year of CI use, children using unilateral CIs and those using simultaneous bilateral CIs showed similar development in auditory perception of speech and oral language.

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