

Development of children's auditory skills in the first year after unilateral and bilateral cochlear implant

Desenvolvimento de habilidades auditivas de crianças no primeiro ano após o implante coclear unilateral e bilateral

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ABSTRACT

Purpose: To identify the development of initial auditory skills in children with unilateral and bilateral cochlear implant during the first year of use. **Methods:** Retrospective longitudinal study of medical records of children who received cochlear implant under the age of four, separated into two groups. The first, composed of children implanted unilaterally and the second, by children implanted and simultaneously. Data on the IT-MAIS scale (Infant-Toddler Meaningful Auditory Integration Scale) were collected before surgery and three months, six months and one year after surgery. These results were compared with the existing Clinical Markers. In addition, the family's classification was noted, according to the Family Involvement Assessment Scale. **Results:** The data of 29 children with bilateral cochlear implant and 30 children with unilateral cochlear implant were evaluated. The IT-MAIS score of the two groups was similar to the Clinical Markers. Regarding the Family Involvement Assessment Scale, there was a difference, showing that families in the bilateral group were more involved. **Conclusion:** In the first year of use of the cochlear implant, the development of children implanted bilaterally did not differ from the development of children implanted unilaterally, suggesting that initial auditory skills are likely to develop with unilateral auditory input.

Keywords: Hearing Loss; Deafness; Cochlear Implants; Child; Inventories and Questionnaires; Auditory perception

RESUMO

Objetivo: identificar se as diferenças no desenvolvimento da aquisição das habilidades auditivas iniciais em crianças após o implante coclear, unilateral ou bilateral, podem ser evidenciadas durante o primeiro ano de uso. **Métodos:** estudo longitudinal retrospectivo de levantamento de prontuários. Foram incluídas crianças que receberam o implante coclear antes dos 4 anos de idade, separadas em dois grupos. O primeiro, composto por crianças implantadas unilateralmente e o segundo, por crianças implantadas bilateral e simultaneamente. Foram coletados os dados referentes à escala IT-MAIS (*Infant-Toddler Meaningful Auditory Integration Scale*) antes da cirurgia e três meses, seis meses e um ano após a cirurgia. Esses resultados foram comparados com os marcadores clínicos já existentes. Além disso, as famílias foram classificadas segundo a Escala de Avaliação de Envolvimento Familiar. **Resultados:** foram avaliados os resultados de 29 crianças com implante coclear bilateral e 30 crianças com implante coclear unilateral. A pontuação obtida no IT-MAIS dos dois grupos foi semelhante aos marcadores clínicos. Quanto à Escala de Avaliação de Envolvimento Familiar, houve diferença, mostrando que as famílias do grupo dos bilaterais estavam mais envolvidas. **Conclusão:** no primeiro ano de uso do implante coclear, o desenvolvimento da aquisição das habilidades auditivas iniciais das crianças implantadas bilateralmente não diferiu do desenvolvimento de crianças implantadas unilateralmente, sugerindo que as habilidades auditivas iniciais são passíveis de desenvolvimento com entrada auditiva unilateral.

Palavras-chave: Perda Auditiva; Surdez; Implante Coclear; Criança; Inquéritos e Questionários; Percepção auditiva

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INTRODUCTION

The literature provides good support for the recommendation of cochlear implant (CI) for children with severe to profound bilateral sensorineural hearing loss who do not benefit from the use of hearing aids (HA)⁽¹⁾. Likewise, the literature provides several materials that broadly address the benefits of bilateral cochlear implants, especially the improvement of sound localization⁽²⁾ and perception in noisy environments⁽³⁻⁵⁾.

When there is no useful residual hearing in either ear, the literature also shows that the acquisition of oral language and the development of auditory skills could be faster in bilateral cochlear implants, when compared to unilateral cochlear implants^(2-4,6-9). But, as it is not simple to follow the acquisition of these skills, the assessment of these young children is a challenge for professionals. Thus, monitoring the development of auditory and language skills is more effective in clinical practice through standardized questionnaires that have scores expected by the time of cochlear implant use to document this progress.

Based on the analysis of the first five years of hearing device use in children with unilateral implants before 36 months of age, Comerlatto⁽¹⁰⁾ determined the clinical markers for the development of auditory and oral language skills in children with cochlear implants. The author used questionnaires applied to parents and speech perception tests with a minimum battery of the CI assessment protocol and used in several studies in the literature⁽¹¹⁾, such as the Infant-Toddler Meaningful Auditory Integration Scale (IT-MAIS)^(12,13), which has good results, reliability and validity⁽¹⁴⁾. This scale is a useful validation tool for documenting the progress of children's early auditory skills after implantation through approximately four years of CI use⁽¹¹⁾.

The use of questionnaires with parents and/or family members also shows the importance of family involvement, both for observation of expected auditory behaviors and for auditory rehabilitation. In this sense, the literature shows that children from families with a high degree of involvement have better language development and children from families with a low degree of involvement usually have language delay^(15,16).

In Brazil, unilateral cochlear implants have been performed since the 1990s, in addition to being offered by the Brazilian Unified Health System (SUS) since 2000. However, the performance of bilateral surgery was only approved in December 2014, through Ordinance No. 2776 of the Ministry of Health⁽¹⁷⁾. As a result, cochlear implant centers that have been operating since the beginning still have children with unilateral implants who, at the time, could have been recommended to perform bilateral CI. However, there are still children who only receive unilateral CI recommendation due to specific etiologies or audiological criteria.

The hypothesis of this study is that children with bilateral implants performed simultaneously would have a faster development in the acquisition of initial auditory skills in the first year after surgery, when compared to children implanted unilaterally with similar residual hearing.

This study aimed to investigate whether differences in the acquisition of initial auditory skills in children after unilateral or bilateral cochlear implantation can be evidenced during the first year of use.

METHODS

This is a retrospective longitudinal study that is part of the project approved by the Research Ethics Committee of the institution, under the protocol No. 48247615.1.0000.0068, opinion No. 1.215.074. As the study was carried out from the survey of medical records, the Informed Consent Form (ICF) was not necessary. The research sample consisted of children with cochlear implants, divided into two groups. The first group (Group 1) included children with unilateral implants, while the second group (Group 2) included children with bilateral implants performed simultaneously in the cochlear implant department of the Hospital das Clínicas da Universidade de São Paulo – School of Medicine, from 2014 to 2017.

The researchers adopted the following sample selection criteria:

- Unilateral or simultaneous bilateral cochlear implant user, provided that the implantation surgery has been performed up to 4 years of age, since the literature reports that the CI provides better results when performed within the critical period for auditory development and of language^(6,18);
- Total insertion, considering one or two electrodes outside the cochlea (provided that the same stimulated frequency range is maintained);
- Effective use of the device for at least seven/eight hours/day. In this sense, the literature reports an average of 9.86 hours (with a standard deviation of +/-3.43 hours)⁽¹⁹⁾, considering the first year of using the sound processor and the adaptations to the use of the hearing device. Average hours of use per day were analyzed by recording hours of use, when available on the sound processor, or by asking parents about daily use, battery life, and device care.

On the other hand, the researchers excluded from the sample children diagnosed with cochlear malformation, those who underwent cochlear reimplantation surgery in the first year of use, who had other underlying conditions or who had insufficient data recorded in their medical records.

Procedures

The following data were collected from the medical records of both groups:

- Child's age;
- Etiology of deafness;
- Better auditory threshold in pre-surgical audiometry (if only the result of the Brainstem Evoked Response Audiometry was available in the medical record with no result, a value of 130 dBHL (decibel hearing level) was considered for statistical analysis);
- Age when started using hearing aids before CI surgery;
- Better auditory threshold in the frequencies tested in the pre-surgical audiometry in free field with the hearing aid (when the response was absent for the maximum

intensity output of the audiometer, a value of 130 dBHL was considered for statistical analysis);

- IC brand;
- Three-tone average of 500 Hz, 1000 Hz and 2000 Hz of free field audiometry with the cochlear implant, considered as a marker of adequacy in the programming of the sound processor and access to speech sounds, as it is the most used assessment in the routine in the group of CI, which is available in most medical records;
- Data collected by the IT-MAIS questionnaire in the preoperative moments and in all follow-up visits during the first year of CI use;
- Family category classification during the first year of CI use.

All children with cochlear implants were evaluated within the protocol of the CI group⁽²⁰⁾. The applied questionnaires are part of the institution's protocol battery and are used in a standardized way by the team, which includes experienced speech-language pathologists in the area of cochlear implant.

In order to assess the initial auditory skills, the IT-MAIS scale was used, developed by Zimmerman-Phillips et al.⁽¹²⁾ and adapted to Brazilian Portuguese by Castiquini and Bevilacqua⁽¹³⁾. It should be noted that this scale aims to assess the development of early auditory skills in children with hearing impairment in their routine, based on the use of the hearing device. The scale questions address the following auditory skills: vocalization, detection and attention to sounds, discrimination of environmental and speech sounds, and ability to attribute meaning to sounds. All these skills are observed during the first years of development of the child with the cochlear implant⁽¹⁰⁾.

The scale consists of ten questions that are asked in the form of an interview with parents or caregivers of children. Then, the responses are scored by the evaluator by the frequency of occurrence of the child's behavior. The score ranges from 0 to 4, where 0=never (0%); 1=rarely (25%); 2=occasionally (50%); 3=often (75%); and 4=always (100%). The maximum score for the questionnaire is 40 points, which corresponds to 100%.

In this study, the questionnaire score was obtained in the preoperative period and in the follow-up visits at three months, six months and one year of cochlear implant use.

Clinical markers of the development

The results of the two groups were compared using the clinical markers of the development in the IT-MAIS, proposed by Comerlato⁽¹⁰⁾. The aforementioned study assessed the development of 230 children with unilateral implants up to 3 years of age, using the IT-MAIS, MUSS (Meaningful Use of Speech Scale) and Hearing and Language Categories. The study sample was divided into three groups: children who received their cochlear implants before 18 months of age (group 1); between 19 and 24 months (group 2) and between 25 and 36 months (group 3) and determined the clinical markers of the development for each scale.

Family Category

The assessment of the quality of the family's involvement in the rehabilitation process was essential for the interpretation of the children's progress. For this purpose, the researchers used the Family Involvement Rating, which was developed by Moeller⁽¹⁵⁾ and translated into Brazilian Portuguese by Ribeiro⁽²¹⁾. The Moeller scale was completed and scored by the speech-language pathologist at the implant center by summing the information provided by the family and when available the speech-language pathology report of auditory rehabilitation.

The Family Involvement Rating assesses issues such as family adjustment, participation in sessions and effectiveness in communicating with the child through a score of five categories: 1=limited participation 2=below average participation; 3=average participation; 4=good participation; and 5=ideal participation (Chart 1).

Demographic data from IT-MAIS and family category were analyzed between groups and statistically analyzed using the Mann-Whitney U test for independent samples. The analysis of variance (ANOVA test) was used only in the comparison between the hearing thresholds of the right and left ears between the groups.

RESULTS

The researchers evaluated the outcomes of 29 children with simultaneous bilateral cochlear implants and 30 children with unilateral cochlear implants. There was no difference for the variables studied between the two groups of children using cochlear implants (Table 1).

As for the Family Involvement Rating⁽¹⁵⁾, there was a statistical difference for the Moeller category between the unilateral group and the bilateral group, which shows that families in the group of children with bilateral implants are possibly more involved and committed to stimulation (Table 1).

Very similar IT-MAIS scores were obtained between the groups, both for the values of clinical markers of the development for the same age group at implantation of Group 2⁽¹⁰⁾, considering the difference in standard deviation, as well as between the groups. The only difference was in the pre-surgical IT-MAIS, which suggests that children with unilateral cochlear implants would have some previously developed auditory skills (Table 2).

DISCUSSION

The study aimed to investigate whether the acquisition of initial auditory skills in children undergoing simultaneous bilateral cochlear implant would be different from that observed in children receiving unilateral cochlear implant in the first year after surgery. In this sense, with these results, the objective would be to guide the family earlier in terms of expectations.

As for the patients included in the research, it should be noted that the surgery was performed before the 2014 ordinance⁽¹⁷⁾ in the group of children with unilateral implants, as well as the patients in the study that defined the Clinical Benchmarks of the Development, proposed by Comerlato⁽¹⁰⁾. Therefore, even though these children fit the recommended criteria for bilateral

Chart 1. Family Involvement Rating Scale^(9,16)

Ideal Involvement (5)	Families seem to have made a good adaptation to the child's deafness, being able to put into perspective the child's conditions in the family context. Family members actively participate and attend sessions and meetings and independently seek information. They are effective advocates for children in their insertion in health and educational services. Family members become highly effective as conversation partners with children and serve as language role models consistently, as well as becoming fluent users of the child's mode of communication, being able to apply language expansion techniques. Extended family members are involved and provide additional support for the child.
Good Involvement (4)	Family members show an adjustment to the child's deafness that is above average. They regularly attend sessions and meetings. Parents play an active role (perhaps not the main one) in planning clinical and educational goals for the child. Family members serve as good language models for the child and strive to bring the techniques home. Some family members have reasonable skills in the child's mode of communication and/or in techniques for language stimulation. Efforts are made to involve extended family members.
Involvement within the Average (3)	The family struggles to understand and accept the child's diagnosis. Family members attend most sessions and meetings. Busy schedules or family stresses can limit opportunities to carry out what has been learned at home. The family may understand child care as a challenge. The family participates in planning, but, in general, mainly accepts the opinion of professionals. The family tries to protect the child, but does not use its efforts well. Family members (e.g., the mother) may take most of the responsibility for developing the child's communication needs. Family members develop at least basic skills in the child's mode of communication. They intend to use language expansion techniques, but need constant support and guidance.
Below Average Involvement (2)	The family makes an effort but suffers to accept the child's diagnosis. The family may be inconsistent in attendance at appointments. Parents can be inconsistent in placing and maintaining the child's hearing aids at home and at school. They may have some significant stress points in their lives that can affect the consistency of working from home. Caring for the child represents daily challenges for the family. Communicative interactions with the child are basic. The family lacks fluency in the way of communicating with the child.
Limitation Involvement (1)	The family faces significant stresses in life, which can affect the child's needs (e.g., domestic abuse and lack of a home). The family has a limited understanding of deafness and its consequences for the child. The involvement may be sporadic or ineffective. Communication between parent and child is limited to the most basic needs.

Table 1. Comparison of demographic data between groups (median - minimum and maximum)

Variable	Bilateral Median (minimum-maximum)	Unilateral Median (minimum-maximum)	p-value*
Age at implantation (months)	23 (10-45)	24 (15-43)	p=0.0987
Best pre-surgical hearing threshold (dBNA)	95 (70-130)	97.5 (60-130)	p=0.3694
Beginning of pre-surgical hearing aid use (months)	10 (3-34)	12 (3-27)	p=0.1549
Best pre-surgical free field auditory threshold with hearing aids -(dBNA)	80 (40-130)	73 (40-130)	p=0.7807
Moeller Category (Classification)	4 (2-5)	3 (1-5)	p=0.0111
PTA of free field hearing threshold with CI – right ear/ left ear (dBNA)	35/35	38	p=0.1876

*Significant values (p≤0.05) in the Mann-Whitney U test.

Subtitle: dBNA=decibel hearing level; PTA = pure tone average of 500, 1000 and 2000 Hertz; CI=Cochlear Implant

Table 2. Comparison of Infant-Toddler Meaningful Auditory Integration Scale values in both groups with clinical markers⁽¹⁰⁾

	Clinical Marker (G2) ⁽¹⁰⁾	Bilateral	Unilateral	p-value
	Median	Median (minimum-maximum)	Median (minimum-maximum)	
Pre-surgical	-	10% (0-62.5)	16% (0-82.5)	0.0437*
Three months	55%	45% (5-85)	42% (12.5-92.5)	0.7933
Six months	80%	72% (20-97)	59.5% (5-100)	0.2434
12 months	94.7%	85% (12-100)	83.5% (25-100)	0.5904

*Significant values (p≤0.05) in the Mann-Whitney U test.

Subtitle: G2=Group 2; %=percentage

implantation, at that time it was only possible to perform unilateral implantation.

In the characterization of the sample, both the residual hearing before implantation and the beginning of hearing aid

use can influence the results and, in this study, they were similar between the unilateral and bilateral CI groups. Likewise, the time of daily use of the devices is essential for the development of auditory skills and the effective and consistent use contributes

to the acquisition of speech and language^(22,23). In this context, the effective and consistent use may have been a bias in this study, as the data record is very accurate for analysis by the speech-language pathologist, especially in the first year of cochlear implant use, in which the number of hours of use increases after adaptation with the hearing device. In addition, studies have already shown the importance of continuous use for better speech and language development⁽¹⁹⁾. However, most of the sample used sound processors that did not record this data and, as such, the self-report of hours of use in both groups was used to carry out the study.

This study found a progression in the IT-MAIS results over the follow-up visits, at a rate similar to the values proposed by the clinical markers⁽¹⁰⁾ in both groups (Table 2). However, it should be noted that this result does not mean that bilateral implantation does not have advantages over unilateral implantation, since only the initial skills of detection, discrimination, attention and ability to attribute meaning to sounds were evaluated. It is known that bilateral stimulation promotes the development of binaural skills that will be perceived later^(18,24), such as sound localization⁽²⁾ and speech discrimination in noise^(3,4). The longer the follow-up time of the implanted children's development, the more data can be collected to analyze the difference between the groups, which is essential, since two years or more are needed for integration and binaurality^(5,7). Misurelli et al.⁽⁹⁾ showed benefits in the bilateral cochlear implant compared to the unilateral implant in the abilities of speech recognition in noise in different positions and the selective attention of children with more than six years of use of the bilateral cochlear implant. In addition, Wie et al.⁽²⁵⁾ monitored children with bilateral cochlear implants for six years after implantation and reported that, four years later, there was no longer any significant difference between the implanted group and the normal-hearing group in general language abilities.

These protocols and other ways of measuring the effects of bilateral cochlear implant use, such as electrophysiological responses⁽²⁶⁾ with cortical auditory potential testes, or through tomography tests to observe activation of the auditory cortex⁽²⁷⁾, are not available in all IC centers, due to the high cost, specific materials and time demands.

In Brazil, cochlear implant centers that work in the Unified Health System, with a large volume of patients, need accessible, easy-to-apply and low-cost instruments to validate the benefits acquired with the use of cochlear implants. In this sense, the questionnaires are a good example of a low-cost and easy-to-apply tool, which are already part of the minimum protocol for pre- and post-surgical assessment of cochlear implants and become essential for monitoring the evolution and providing guidance to families.

The recommendation of bilateral cochlear implant in cases of proven profound deafness is increasingly present in cochlear implant centers and, for monitoring, it is essential to analyze whether the development of initial auditory skills is faster. In this study, the findings of the IT-MAIS questionnaire suggest that the initial auditory skills are similar between the groups and that it is possible to use the data to help guide and review the adjustment of the cochlear implant for better results, regardless of unilateral or bilateral use.

In addition, the results of the IT-MAIS during the first year of use could be used as a reference in both groups studied. When there is a difference in these results, if the child has very

divergent results and lower than the clinical markers, it is an indication that the development is not within the expected range.

Pianesi et al.⁽²⁸⁾ reported that the IT-MAIS values measured at six months after CI activation explained most of the variability in the onset of the First Milestones of Oral Language (FMOL). Children with better scores showed basic language skills earlier than children with lower scores. Lower-than-expected IT-MAIS scores during the first six months after CI should be considered as a warning sign of insufficient progress in oral language development. In these cases, the professional will be able to carry out the appropriate interventions and guidance to the family immediately, in all potentially modifiable factors: CI programming or rehabilitation strategy, in order to improve pre-lingual auditory-perceptive skills and avoid an unfavorable linguistic result.

In clinical practice, other questionnaires⁽²⁹⁾ can be incorporated into the evaluation battery, for greater sensitivity in monitoring the development of initial skills. This can be observed in the study by Comerlato⁽¹⁰⁾ and Pianesi et al.⁽²⁸⁾, who showed a statistically significant correlation between the categories and scales of hearing and speech and language. The data show that, as the child improves the performance in the IT-MAIS and in the Hearing Categories, they also develop the spoken language skills observed in the MUSS and in the Language Categories.

As for the results of the Family Involvement Rating, although the families in the bilateral group had a higher degree of family involvement, there was no difference in the initial auditory skills. In order to observe this relationship between language development and the involvement of families in this study^(15,16), it would also be necessary to follow up for a longer period, in addition to evaluating the evolution of the acquisition of other skills and oral language. However, despite the median involvement of parents in the group of children with unilateral cochlear implants, there was development of early auditory skills. This once again reinforces the importance of reviewing guidelines and conduct when there are indications of non-evolution in the IT-MAIS questionnaire.

In order to provide better monitoring of the evolution of children with cochlear implants, it is necessary to monitor hearing and language in different contexts⁽³⁰⁾. The IT-MAIS questionnaire should be included in the protocol to identify early auditory skills and assist in monitoring unilateral and bilateral implanted children. However, complementary assessments and other tools are also needed in patient assessment.

CONCLUSION

There was no difference in the development of initial auditory skills acquisition in the first year of cochlear implant use in children with bilateral cochlear implants compared to children with unilateral implants, which suggests that the acquisition of early auditory skills may develop with unilateral auditory aid.

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