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ORIGINAL ARTICLE

Hearing rehabilitation in cerebral palsy: development of language and hearing after cochlear implantation☆,☆☆

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KEYWORDS
Cerebral palsy;
Cochlear implants;
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Abstract
Introduction: Auditory rehabilitation in children with bilateral severe-to-profound sensorineural hearing loss with cochlear implant has been developed in recent decades; however, the rehabilitation of children with cerebral palsy still remains a challenge to otolaryngology and speech therapy professionals.
Objective: To verify the effectiveness of cochlear implants in the development of auditory and language skills in children with cerebral palsy.
Methods: A prospective analytical study. The evaluation of auditory responses to speech test was applied to the children in this study at regular intervals following implantation. Standardized tests that assess and quantify the development of auditory and language skills were administered and speech therapy video records and speech therapy files were analyzed. All children went through individually tailored intensive audiological rehabilitation programs following cochlear implantation.
Results: Two participants had gradual auditory and language development when compared to other participants who reached advanced levels in hearing and oral language classifications.

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Conclusion: The use of the Cochlear implant enabled participants to reach advanced stages of hearing and language skills in three of the five participants with cerebral palsy in this study. This electronic device is a viable therapeutic option for children with cerebral palsy to help them achieve complex levels of auditory and language skills.

PALAVRAS-CHAVE
Paralisia cerebral;
Implante coclear;
Perda auditiva

Reabilitação auditiva na paralisia cerebral: desenvolvimento da audição e linguagem após implante coclear

Resumo
Introdução: A reabilitação auditiva em crianças com deficiência auditiva neurossensorial severa a profunda bilateral com o Implante Coclear foi consagrado nas últimas décadas, contudo, ainda permanece um desafio para a otorrinolaringologia e a fonoaudiologia a reabilitação do portador de paralisia cerebral.
Objetivo: Verificar a efetividade do Implante Coclear no desenvolvimento das habilidades auditivas e de linguagem em crianças com paralisia cerebral.
Método: Estudo analítico prospectivo. Foram aplicados testes padronizados que avaliam e quantificam o desenvolvimento das habilidades auditivas e de linguagem. Foram analisadas as filmagens das terapias fonoaudiológicas e os registros descritos ao término de cada sessão de terapia.
Resultados: As crianças analisadas apresentaram desenvolvimento auditivo e de linguagem satisfatório quando comparado às demais crianças que alcançaram níveis mais complexos nas categorias de audição e evolução significativa no desenvolvimento da linguagem oral.
Conclusão: O uso do Implante Coclear favoreceu o alcance de etapas avançadas das habilidades de audição e linguagem em três das cinco crianças com paralisia cerebral desse estudo. Esse dispositivo eletrônico tem sido uma opção terapêutica viável para que crianças com paralisia cerebral alcancem etapas complexas no que se refere às habilidades auditivas e de linguagem.

Introduction

Cerebral palsy (CP) is a non-progressive motor disorder resulting from brain impairment in the early stages of child development. The basic neurological symptoms are characterized by motor disorders that develop over time, causing delay or disruption of sensory motor development, with insufficient postural mechanism, presence of reflexes at times when they should be inhibited, alterations in muscle tone, and incapacity to perform movements.[1]

Possible disorders of higher cortical functions can generate important impact on activities of daily living. Moreover, language acquisition may be delayed and the child with CP may exhibit changes in articulation, speech, fluency, and prosody. Its clinical manifestations may change over the course of development due to brain plasticity, particularly in the immature brain. Due to this plasticity, uninjured areas of the brain can assume some of the functions of the damaged areas.

In addition to motor impairment, other disabilities may be present, such as hearing, visual and cognitive deficits, as well as language, behavioral, and learning alterations.[1]

The literature shows several common etiological agents for both CP and sensorineural hearing impairment. Among them are congenital infections, hyperbilirubinemia, prematurity, low birth weight, perinatal hypoxia, and cytomegalovirus, among others.

The cochlear implant (CI) is a high-technology electronic device developed to perform the function of damaged or absent cochlear hair cells, and to provide electrical stimulation of the remaining auditory nerve fibers. The CI does not cure deafness, but provides a sense of hearing with the required quality for the perception of speech sounds.[2]

Currently, the CI is considered a viable therapeutic option in cases of children with CP and severe/profound bilateral sensorineural hearing loss who have not shown benefits with the use of hearing aids.[3–7]

There are other aspects to consider, when other conditions are present in the child in addition to hearing loss. Each disability that is added to the deafness, will present distinct clinical features that will influence both the diagnostic evaluation and the rehabilitation of the hearing impairment. Among other factors, hearing results will depend on the child's potential for his or her overall development.[6]

The most important benefit provided by CI is the possibility of perception of higher frequency speech sounds. This allows the child to recognize speech sounds more easily, and oral language acquisition occurs faster and with less stress.[2]

The sooner the brain receives meaningful sounds, the better the conditions for it to produce good results, because of its functional plasticity and the resultant decrease in sensory deprivation.

The work with CIs in children has transformed and improved the speech therapy process due to the improved ability to perceive speech sounds from the electrical stimulation. The possibility of such prostheses to provide access to auditory experiences, such as the patterns of linguistic auditory code, effectively influences the early years of life, which determine the child's constitution.[8]

The auditory system is the natural pathway to learn speech, and auditory skills are essential for the development of oral language and speech production; thus, effective work is necessary for them to develop. This work should occur within a meaningful linguistic context for the child, as a natural result of incidental learning in situations of daily living.[9]

It is through hearing that infants acquire language. The auditory stimulus can come from several sources: the human voice, household sounds, toy sounds, and music.[10]

Standardized tests and classifications of development are important as descriptors of the development of a child with hearing loss, such as the: (1) Infant-Toddler Meaningful Auditory Integration Scale (IT-MAIS); (2) MacArthur-Bates Communicative Development Inventory (CDI); (3) Classification of auditory skills; and (4) Classification of language skills.

IT-MAIS was proposed by Zimmerman-Phillips in 1997 and is a test adapted for children younger than 4 years that evaluates responses to speech and environmental sounds that are mediated exclusively by the auditory sensory pathway. This scale has also been used to assess post-surgical outcomes of children with CI. The test consists of ten closed questions that must be answered by parents or guardians.

The CDI: Words and Gestures is a tool used in speech therapy to evaluate and monitor the language development of young children aged 8–16 months. This tool is administered to the parents or guardians in an interview. In the Portuguese version, the inventory assesses the child's development of lexical comprehension and production. It is divided into three parts: the first corresponds to the first words, and the second corresponds to actions and gestures. The first part is subdivided into: A (first signs of comprehension), B (comprehension of 28 sentences), C (starting to speak), and D (vocabulary list). Item D is subdivided into 22 categories with a total of 415 words. The second part of the test is divided into: A (first communicative gestures), B (games and routines), C (actions with objects), D (pretending to be the parents), E (imitation of other adults' activities), and F (actions with an object in the place of another). The third part corresponds to general information about the child.

Tables 1 and 2 describe the skills by classifications of hearing and oral language skills.

This study aimed to analyze, through standardized testing, the classifications of development, and clinical observations, the improvement of auditory and language skills in children diagnosed with CP and profound bilateral sensorineural hearing loss using CIs, aiming to assess the effectiveness of the CI as a therapeutic resource in this population.

Table 1 Summary of classification of auditory skills proposed by Geers (1994)[11].

Classification	Auditory skills
0	This child does not detect speech in normal conversation.
1	Detection: This child detects the presence of speech signal.
2	Perception pattern. This child detects the presence of the speech signal.
3	Starting word identification.
4	Word identification through vowel recognition.
5	Word identification through consonant recognition.
6	Word identification in open-set speech recognition.

Table 2 Classification of oral language skills proposed by Bevilacqua et al. (1996)[12].

Classification	Language development
1	Does not speak, only produces undifferentiated vocalizations.
2	Speaks only isolated words.
3	Construct simple sentences, with two or three words.
4	Constructs sentences with four or five words.
5	Child is fluent in oral language.

Methods

The study was approved by the Research Ethics Committee of Fundação de Ensino e Pesquisa em Ciências da Saúde FEPECS (Brasília, DF, Brazil), under protocol number 480/09. Caregivers provided written informed consent.

The research was characterized as a longitudinal, analytical, and prospective study. Five children younger than 6 years, one female and four males, were included in the study.

All children had a diagnosis of CP associated with prelingual bilateral profound sensorineural hearing loss and had undergone CIs. All participants attend speech therapy sessions with therapeutic approach based on the aural–oral method twice weekly. Three participants attend speech therapy in a specialized philanthropic institution and the two others in private speech therapy clinics. Both establishments are located in Brasília/DF – Brazil. Table 3 describes the data of the study participants.

Data collection was conducted through speech therapy files, with the analysis of the following documents: speech-language records made at the end of each therapy session following activation of the CI, video recordings of the speech-language therapy sessions, standardized tests, and development classifications that assess the performance of auditory skills: the IT-MAIS, the MacArthur CDI, and the classifications of hearing and language). The IT-MAIS and CDI standardized tests were reported by the

Table 3 Characterization of the study participants.

Subjects	Participant A	Participant B	Participant C	Participant D	Participant E
Chronological age[a]	4 years and 11 months	4 years and 5 months	3 years and 11 months	3 years and 7 months	5 years and 6 months
Type of palsy	Diplegia	Diplegia	Hemiplegia	Quadriplegia	Hemiplegia
Degree of cognitive impairment	Marked	[b]	Moderate	[b]	[b]
Type and degree of hearing loss	Profound bilateral sensorineural	Profound bilateral sensorineural	Profound bilateral sensorineural	Profound bilateral sensorineural	Profound bilateral sensorineural
Time of speech therapy using the aural–oral method	4 years and 3 months	3 years and 2 months	2 years and 2 months	1 year and 10 months	3 years and 6 months
Chronological age at the time of CI surgery	3 years and 8 months	2 years and 6 months	3 years and 1 month	1 year and 7 months	2 years and 7 months
City in which the high-complexity center was located where the CI surgery was performed	Brasília (DF) Private institution	Natal (RN) Public institution	Bauru (SP) Public institution	Natal (RN) Public institution	Natal (RN) Public institution
Implanted ear	Right	Right	Left	Bilateral	Right
Brand and model of speech processor and internal component of CI	Advanced Bionics Platinum HiRes® 90 K	Cochlear Freedom Nucleus® 24 K	Advanced Bionics Harmony HiRes® 90 K	Cochlear Freedom Baby Nucleus® 24 K	Cochlear Freedom Nucleus® 24 K
Brain auditory age with the CI[a]	14 months	20 months	9 months	22 months	2 years and 9 months

CI, cochlear implant.
[a] At data collection.
[b] Children B, D, and E did not have their cognitive development assessment attached to their records, but speech therapy records suggest that their cognitive development appears to be better than in children A and C.

parents/guardians of study participants. These records were accessed after obtaining permission from the participants' parents or guardians and the heads of the aforementioned institutions, who signed the informed consent.

Results

The results of the IT-MAIS and CDI tests and the classifications of hearing and speech of participants A, B, C, D, and E are shown in Tables 4–8, respectively.

In participant A, it is observed that nine months after the tests were first administered, there was an improvement of 37.5% in the IT-MAIS, and an increase of 25 words that were understood and 20 words that were spoken, both in the CDI. In development of auditory and language skills after using the CI for 24 months, the child showed few significant advances.

Participant B experienced an increase of 32.5% in IT-MAIS. Progress in the hearing and language classifications was an evidence of better performance in hearing comprehension and an increase in the linguistic repertoire.

Participant C, seven months after the first IT-MAIS testing, showed an increase of 50% in the questionnaire score. This percentage may indicate that the CI has enabled better auditory perception for this child. At 24 months of brain auditory age, the mother replied in the CDI that the child aurally understood approximately 147 words. However, according to the records of observations made during diagnostic speech therapy sessions, the child showed no understanding for the words mentioned in therapeutic situations. One can assume that the mother overestimated the child's hearing comprehension. Likewise, there was no significant progress in the hearing and language classifications.

Participant D, at the last IT-MAIS testing, showed an increase of 35% in the test score when compared to the first testing. Six months after the first standardized tests were applied, the child reached the maximum classification of auditory skills and, concomitantly, showed a better performance in hearing comprehension and use of new words in spontaneous speech. These findings suggest that the use of CI has been effective for this child, which will result in an increasingly more confident attitude regarding the auditory sensory pathway.

Table 4 Results of IT-MAIS and CDI tests and classifications of hearing and language of participant A.

Auditory age with CI	IT-MAIS	CDI (comprehension)	CDI (linguistic repertoire)	Classification of hearing	Classification of language
1 month	25%	–	–	1	1
6 months	32.5%	20 words	–	1	1
10 months	60%	25 words	13 words	2	1
14 months	62.5%	31 words	16 words	3	1
24 months	62.5%	44 words	20 words	3	1

IT-MAIS, Infant-Toddler Meaningful Auditory Integration Scale; CDI, MacArthur-Bates Communicative Development Inventory.

Table 5 Results of IT-MAIS and CDI tests and classifications of hearing and language of participant B.

Auditory age with CI	IT-MAIS	CDI (comprehension)	CDI (linguistic repertoire)	Classification of hearing	Classification of language
1 month	32.5%	–	–	2	1
8 months	55%	83 words	31 words	3	2
12 months	62.5%	216 words	53 words	4	2
20 months	65%	247 words	86 words	5	3

IT-MAIS, Infant-Toddler Meaningful Auditory Integration Scale; CDI, MacArthur-Bates Communicative Development Inventory.

Table 6 Results of IT-MAIS and CDI tests and classifications of hearing and language of participant C.

Auditory age with CI	IT-MAIS	CDI (comprehension)	CDI (linguistic repertoire)	Classification of hearing	Classification of language
1 month	12.5%	–	–	2	1
8 months	22%	23 words	–	2	1
16 months	55%	140 words	3 words	3	1
24 months	62.5%	147 words	11 words	3	1

IT-MAIS, Infant-Toddler Meaningful Auditory Integration Scale; CDI, MacArthur-Bates Communicative Development Inventory.

Table 7 Results of IT-MAIS and CDI tests and classifications of hearing and language of participant D.

Auditory age with CI	IT-MAIS	CDI (comprehension)	CDI (linguistic repertoire)	Classification of hearing	Classification of language
14 months	40%	58 words	54 words	3	2
20 months	55%	103 words	86 words	6	3
22 months	67.5%	171 words	139 words	6	3
30 months	67.5%	307 words	243 words	6	3
38 months	75%	342 words	289 words	6	5

Table 8 Results of IT-MAIS and CDI tests and classifications of hearing and language of participant E.

Auditory age with CI	IT-MAIS	CDI (comprehension)	CDI (linguistic repertoire)	Classification of hearing	Classification of language
2 months	27.5%	15 words	6 words	2	2
4 months	65%	43 words	14 words	3	3
11 months	90%	107 words	32 words	4	3
16 months	97.5%	154 words	77 words	4	3
20 months	100%	236 words	121 words	4	3
34 months	100%	454 words	378 words	6	4

IT-MAIS, Infant-Toddler Meaningful Auditory Integration Scale; CDI, MacArthur-Bates Communicative Development Inventory.

As a result of moving to another house, participant D only joined the specialized speech therapy service in which this research was developed after 15 months of CI activation. The researcher did not have access to the records of the institution where the child previously underwent speech therapy. Thus, it was not possible to assess previous data.

Participant E obtained a 72.5% increase in the IT-MAIS test responses when compared with the first test application. This child has reached satisfactory levels of performance in hearing and language skills, which can be proven by the number of spoken and understood words documented on the CDI. This performance allowed the child to reach the maximum classification of auditory skills and construct phrasal structures with four or five words, which characterizes the fifth classification of language skills.

It was not possible to perform the statistical analysis considering the clinical heterogeneity of CP and the small number of participants; the analysis would not provide accurate data for results. Consequently, one of the challenges for future research is measuring and quantifying the results of the CI in different manifestations of CP.

Discussion

All five of the participants experienced anoxia at birth associated with prematurity as the etiology of their CP. The etiology of the sensorineural hearing loss was likely related to the use of ototoxic drugs and a prolonged ICU stay in participants A, B, and C, pneumococcal meningitis in child D, and severe jaundice in participant E.

In a study of 40,000 children, low birth weight and anoxia were given as the causes of CP, but these two factors alone would not explain the existence of the different clinical pictures.[13] It is now known that there are many factors that can damage a developing brain.

A study of 67 CP patients of both genders found that 51% of the sample had hearing impairment.[14]

Because of the chronological age of the study participants, they would not ordinarily be administered the CDI. However, we decided to use this tool for evaluation because of the hearing deficit caused by the profound sensorineural hearing loss in all participants.

When there is a language disorder associated with CP, two possibilities must be considered: the first is that the associated intellectual disability and, in this case, language alterations, are worsened by the motor deficit, which would likely make verbal interactions more difficult. In the second scenario, the individual with CP has normal overall cognitive development, but with some degree of language impairment, which may be in the phonological and morphosyntactic development, or in the semantic and psycholinguistic aspects.[13] Another facet to be considered in the communication context of children with CP is speech; its components related to vocal production can be greatly affected, thereby altering language acquisition.

Children with CP may miss opportunities to enable their linguistic repertoire, as the perceptive development occurs through the body's own integrated actions to psychomotor measures, influencing the maturation process and, consequently, the development of the processing of auditory, visual, and somesthetic information.[14]

A recent study reported that in children with other disorders including CP in addition to hearing impairment, language development may be close to that of normal children if the impairment is mild. In contrast, children with more severe disorders may show a lower-than-expected development.[15]

Currently, there is considerable discussion in implant centers regarding the indication of CIs in children with other disorders associated with hearing impairment. Those that choose implantation aim to minimize the auditory sensory deprivation by improving interaction with the environment, language comprehension, and consequently, the quality of life.[15]

According to indication and contraindication criteria, at the national and international levels, disorders in addition to hearing impairment, as in the case of CP, do not contraindicate cochlear implantation.[6,7] In this sense, the participants assessed in this study met the indication criteria for the CI because the criteria considered as contraindications were not observed in these patients – there were no medical conditions that contraindicated surgery; no agenesis of the cochlea or of the auditory nerve or central auditory lesions; and no active middle ear infections. Although CP is a neurological impairment, it does not generate any impediment to the use of CI, because the affected area, in this case, refers to the motor area.[6]

The use of IC allowed the improvement in speech perception in children with additional needs, although this improvement is often significantly lower than in children who do not have additional disorders.[16]

The studies described in the literature on CI benefits in cases of CP are few and show a gradual improvement in speech perception and oral language development during the years of device use.[3–5,7,17]

Children with CP and hearing loss are very heterogeneous. The similarity among them lies in the fact that they have hearing impairment. The other characteristics of the motor and cognitive picture are dissimilar and can potentially present as variables in the evaluation process and the rehabilitation approach with CI.[6]

An important piece of information that may influence the performance of auditory skills is the chronological age of the children at the time of surgery, as this identifies the duration of brain auditory sensory deprivation that the child experienced. When comparing the five study participants, participant D received an implant at the earliest age, while participant A was the oldest when implanted.

It is a known fact in preventive psychology that the younger the age of the child for an intervention, the better the results. Thus, the ideal time for CI surgery is during first 2 years of life. This is because of the greater neural plasticity, which facilitates brain reorganization to new stimuli and is complemented by the need for verbal learning in the period of normal language acquisition – from 1 to 2 years of age. This provides an effective mechanism to allow development that is appropriate to the child's developmental process, while avoiding a longer duration of auditory deprivation.[18]

Studies show that children implanted before three years of age have better performance in auditory perception for speech sounds than children implanted later.[18] In our study participants implanted before 3 years of age were case: B, at 2 years and 6 months; D, at 1 year and 7 months; and E, at 2 years and 7 months. Early childhood is the most important period for neuronal plasticity; therefore, the tendency is that these children will achieve better results with the CI.[2]

Between ages 3 and 6 the indication for the CI is more complicated, since the post-surgical improvements are more limited, due to longer auditory sensory deprivation.[2] Participants A and C were not implanted until ages 3 years and 8 months, and 3 years and 1 month, respectively.

The initiation of audiological awareness in the implanted child begins with helping her understand the meaning of the sounds she hears by making her aware of the sound source. As this occurs, she will become increasingly confident in her auditory sensory pathway. Upon activation of the CI, the child's hearing development should occur following the same stages through which children without hearing impairment go. However, one aspect should be taken into consideration in relation to participants A and C: there is cognitive impairment associated with the neurological symptoms of CP that may slow the development of auditory skill with the CI.

In one study of 60 children treated with CI, 27 children reached auditory skill development classifications 5 and 6, the most advanced levels of auditory development. Twenty-three children achieved classifications 3 and 4, while 10 children reached only classifications 1 and 2.[19]

In the present study, at 20 and 36 months of brain auditory age with CI, participants D and E reached classification 6 of hearing. At 20 months of brain auditory age, participant B reached classification 5. At 14 and 16 months of brain auditory age, participants A and C were in classification 3.

In a study with three deaf children younger than age 3, one of the tools used to monitor the development of auditory and language abilities was the CDI – Words and Gestures version.[20] Although this tool is indicated to evaluate children aged between 8 and 16 months, the author chose to use it due to the children's language gap from decreased auditory acuity. We made the same decision in our study for the children whose chronological age was 5 years and 6 months, and 3 years and 7 months.

At close to 2 years of brain auditory age, participants B, D, and E were actively developing hearing skills, particularly in relation to the more complex skill of auditory comprehension. At this time, considering his linguistic repertoire, participant D understood 172 words according to the CDI. At 2 years of age, the child had auditory memory for two words, understood a variety of sentences, discriminated descriptive sentences, followed orders in two directions, recognized by categorization, understood action sentences, understood questions, imperatives, and routine and situational statements, understood personal pronouns, understood the negative ''no'', and understood some concepts and approximately 250–300 words.[21,22]

Taking into account the brain auditory age with the CI, the study shows that in participants B, D, and E, whose speech therapy records suggest better cognitive development than those of participants A and C, the CI has helped the development of hearing skills at stages similar to those observed in children with normal hearing.

When comparing the auditory age of participant C to children with normal hearing of the same age, she has developed auditory skills, in spite of the small gap. In contrast, participant A, who has marked cognitive impairment, has developed auditory skills with a significant lag.

Children with cognitive delay can benefit from the CI, but will have limited results when compared to their peers with normal hearing without cognitive impairment.[18] This scientific finding agrees with the results observed in participants A and C of this study.

Another study of a child with a CI and CP showed that neurological alteration was not an impediment for the child to reach the more advanced classifications of auditory and language skill development.[23]

Conclusion

Although there is a paucity of studies in the literature addressing the use of CI in children with CP, this study demonstrates that the use of this electronic device has contributed to the development of auditory and language skills in the participants.

The CI has been a viable therapeutic option for children with hearing impairment associated with CP, as the device allows children to achieve more advanced stages of auditory and language skills, although at a more gradual rate.

Conflicts of interest

The authors declare no conflicts of interest.

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