

Case reports

Cochlear implant and autism spectrum disorder: a case report

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ABSTRACT

Autism Spectrum Disorder is not an adverse condition for cochlear implantation, but the goals and expectations about the effects on hearing and language outcomes are different from the group of children without other impairments. The objective of this study was to analyze the development of auditory and oral language skills of a child with autism who underwent early cochlear implantation surgery and was included in an auditory (re) habilitation program using the aural-oral method. There was no benefit from the device for the development of oral language, and the child was referred to another communicative method. After 2 years and 9 months using the cochlear implants, there was little benefit from the use of the device for the development of auditory skills, with the child evolving to recognize only his own name. The patient quit using the device after three years of its activation.

Keywords: Cochlear Implantation; Child; Autism Disorder; Auditory Perception; Language Development

A study conducted at Centro Especializado em Reabilitação - Centro Educacional de Audição e Linguagem/ Ludovico Pavoni (CER II - CEAL/LP), Brasília, Distrito Federal, Brazil.

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INTRODUCTION

The term “autism” has undergone several changes along the time. Currently, the 5th edition of the Diagnostic and Statistical Manual of Mental Disorders - (DSM-5)¹ suggests that individuals whose behavioral outcomes entail persistent disabilities in communication and social interaction, as well as stereotypes and restrictions of interests and patterns of activities be classified as subjects with Autism Spectrum Disorder (ASD). The signs of ASD have been present since childhood and restrict or hinder the daily functioning of the individual. Additionally, the disorder may feature three levels of severity¹.

Hearing impairment (HI) is defined as total or partial hearing loss, which can be congenital or acquired. It is classified according to the type (sensorineural, conductive or mixed) and degree (mild, moderate, severe or profound). Fundamentally, people with severe or profound bilateral sensorineural hearing impairment, who do not evidence any benefits from the use of hearing aids, may be referred to a Cochlear Implant (CI)².

Considering that about 40% of children with hearing impairment may feature one or more comorbidities^{2,3}, the referral criteria to the CI have been expanded in order to benefit part of that population, including children with the associated diagnosis of ASD.

Studies conducted in international cochlear implant programs, such as the ones from the University of Michigan in the United States of America (USA), and in Dublin (Ireland), point to the ASD occurring in about 1-3% of those children^{4,5}.

Despite the ASD is not a risk factor for the CI, the goals and expectations on the effects in hearing and language outcomes in that population are different from the group of children with profound hearing loss without any other comorbidities, and their oral communication cannot be a probable realistic goal in these cases⁶. However, studies point to the variety of results regarding hearing and language development found among the population of children with ASD, users of CI⁷⁻⁹.

Considering that the ASD prevalence has increased considerably without any satisfactory explanations^{10,11}, and the number of CI users has also increased among that population⁴, it deems essential to know the benefits and limitations in the use of that device among those children for guidance, reception and planning of the therapeutic process of hearing rehabilitation.

Therefore, this study aimed at analyzing the development of auditory and oral language skills in a child presented with ASD who underwent an early surgery of cochlear implant, included in a program of hearing rehabilitation by means of the aural-oral approach.

CASE PRESENTATION

This study was approved by the Ethics Research Board of the Centro Universitário Unieuro, Brazil, under number 4.274.341 and Certification of Presentation of Ethical Appreciation (CAAE) number 37139420.6.0000.5056.

Data collection was carried out at the Specialized Center in Rehabilitation – Educational Hearing and Language Center/ Ludovico Pavoni (Centro Especializado em Reabilitação - Centro Educacional de Audição e Linguagem/ Ludovico Pavoni CER II - CEAL/LP), in the city of Brasília, Federal District, Brazil. A longitudinal, retrospective study was conducted by means of the patient’s clinical records. The results in the evaluation protocols of auditory and oral language protocols, applied in different periods of the CI use, were analyzed. There was a break of only 20 days in the institution services due to the COVID-19 pandemic during the collection step. The child’s in-presence treatment re-started after that time.

It is a male child, diagnosed with profound bilateral sensorineural hearing loss at six months of chronological age. He started making use of -hearing aids in both ears at seven months old, same age that he was referred to speech-language therapy, grounded in the aural-oral approach, twice a week, sessions of 45 minutes each, apart from a weekly session, 50 minutes each, with an occupational therapist, who worked on his sensory integration. All sessions were individual.

The child underwent surgery for placement of simultaneous bilateral cochlear implant at 12 months of chronological age. The activation of both devices was held at 15 months of chronological age.

At 01 year and 06 months old, suspicion of the presence of other impairments began, apart from his hearing loss. Restricted interest in objects was observed; poor exploration of his toys; none or little communication intent; poor eye contact; lack of proper imitation behavior; preference for fitting toys and repetitive activities; repulse to some textures and use of the other as an instrument.

The child was diagnosed with “child autism” (ICD 10 F84.0) level 3 at age 02 years and 07 months¹. He was also diagnosed with Sensory Processing

Disorder (SPD), with significant deficit in praxis skills. However, he featured normal results in the eye test, Electroencephalogram (EEG) and skull computerized tomography scan.

He is a child who attends therapies regularly, no complications in his CI mapping.

The analyzed protocols are shown in Chart 1.

Chart 1. Analyzed protocols

Assessment	Protocol	Objective of the application
Assessment of the auditory skills	IT-MAIS ¹²	To verify the auditory skills in very small children using their parents' information.
	LittlEars® Auditory Questionnaire ¹³	To assess the auditory skills among the population of babies and children with auditory age of until two years, based on their parents' observation.
Assessment of the oral language development	MUSS ¹⁴	To assess behaviors of the child's speech production in his/her daily life by means of parents' interview.

Captions: IT-MAIS = Infant Toddler Meaningful Auditory Integration Scale; MUSS = Meaningful Use of Speech Scale

Longitudinal analysis on the advancement of the child's development was also conducted regarding his auditory and language skills by categories (Hearing Category¹⁵ and Language Category¹⁶).

RESULTS

No evolution was observed in his development of spoken language until 2 years and 3 months using the CI, as the scores in the MUSS¹⁴ and in the language classification by categories¹⁶ did not change by that time.

Regarding the auditory development, only development in sound detection was observed at the time of CI use of 1 year and 9 months, when the child developed to auditory performance category 2¹⁵, that is, he was able to differentiate words by suprasegmental features, such as duration and tone. The identification of the first words occurred after over 2 years and 3 months of CI use, when the recognition of his own name began (Table 1).

Table 1. Results of the applied tests for the assessment of the auditory and oral language skills

Date of the application	Age	Time using the CI	Auditory Category	Language Category	IT-MAIS	MUSS	LittlEars
September/2019	1y9m	6m	1	1	22.5%	0%	0/35 (critical value)
June/2020	2y6m	1y3m	1	1	22.5%	0%	12/35 (critical value)
December/2020	3y	1y9m	2	1	37.5%	0%	10/35 (critical value)
June/2021	3y6m	2y3m	2	1	37.5%	0%	11/35 (critical value)
December/2021	4y	2y9m	3	1	37.5%	0%	11/35 (critical value)

Captions: y = years; m = months; CI=Cochlear Implant; IT-MAIS = Infant-Toddler Meaningful Auditory Integration Scale; MUSS = Meaningful Use of Speech Scales.

Free-field audiometric test results with the CI evidenced minimum auditory responses at 25 dBHL for frequencies from 500 Hz to 4000 Hz.

Child refusal on the use of the CI was observed, discontinuing the use of the device. Datalogging information held in December 2021 (chronological age 4 years and 2 years and 9 months of CI use) reported

only 4 daily hours making use of the device. In the current case, non-acceptance was not associated to discomfort by professionals, but flight behavior. In December 2022, the patient was reported to quit using the CI processors, both sides. No benefit of the CI was observed to keep eye contact.

DISCUSSION

The aural-oral approach is a set of goals, techniques, strategies, conditions and procedures that prioritizes the construction of the spoken language by means of auditory via and interactive language situations¹⁷.

Despite the presence of audibility with the use of the CI, and the attendance to language-speech therapy services, the child had little evolution regarding the development of his auditory skills.

In a study⁷ conducted with 27 children, users of CI, six of them with ASD, the authors reported that there was not significant correlation between the improvement in audiometric thresholds and the results of questionnaires on auditory skills. Thus, the evaluation of the auditory thresholds with the CI, in free field, is not a good indicator to predict the benefit of the device in deaf children with associated diagnosis of ASD. Moreover, difficulties in attention, social interaction and repetitive behaviors featured by that population may hinder this type of assessment¹⁸.

Despite studies¹⁹ report the benefit of the CI to deaf children diagnosed with ASD, the limitation of the benefits need to be considered. Similar to the observed in the case reported in the current study, other studies do not refer to improvement in the eye contact after the activation of the CI^{5,8}, even though there are family reports of interaction improvement^{15,20}.

Poor evolution was observed in the IT-MAIS¹² score in 01 year and 03 months of the CI use, without any changes from that time, keeping the score of 37.5% until 02 years and 09 months using the device. In a study²¹ carried out in the United States, the authors also observed that 40% of the participants with ASD, users of CI, did not feature any improvement in the score of that test, even though they showed improvement in the thresholds with the device. It is a poorer result than expected for the length of time using the CI, considering that children without comorbidities should achieve 100% score in the test with 19 months using the device²².

In the current study, the child developed auditory recognition of isolated words, being able to recognize his own name. In a study held with 22 children with ASD, users of CI in Italy²³, the authors also observed that most participants (31.8%) progressed to the recognition of environmental sounds or words, not advancing to the auditory understanding of speech.

There was no observed evolution in the development of spoken language. In Brazil, children using CI with ASD also performed worse results in that item than

those implanted children without other comorbidities, with most cases remaining in the emission of vocal sounds²⁴. Data obtained in this study also corroborates international literature. In a study²³ conducted in Italy, the authors reported that 45.5% of the participants with ASD did not feature any benefits regarding the spoken language after the CI, thus, 72.7% did not develop spoken language and only 18.2% were able to utter simple phrases. In another study⁵ with a series of six cases, conducted in Poland, the authors observed that only one child used few words to communicate. In a study²⁵ held with 22 children with ASD, CI users in England, the authors reported that only three of them developed oral communication, two of them could not communicate, and the others communicated by signs or augmentative and alternative communication.

In spite of providing the access to sounds and the possibility of developing auditory skills, studies have shown statistically significant differences between groups of children with ASD, CI users, and groups of children, CI users, without other comorbidities regarding the development of auditory skills and oral language^{9,24}, with limited or scarce/no progress in some skills, even after intense aural-oral speech-language therapy⁹.

The child dropped the CI use after three years of its activation. Report of discontinued use or quitting the CI by children with additional diagnosis of ASD is not rare^{5,23,25}. Factors, such as sensory and behavioral difficulties/disorders, are believed to influence its discontinued use. Moreover, it is important to be attentive to the device programming. The configurations of the electrical current tend to be lower than the reported values in literature to children with CI without additional diagnoses²³. In the case reported here, the child was undergoing weekly therapy of sensory integration with an occupational therapist, and no intercurrent was observed after returns to program the devices. However, the cause for dropping the use of the CI cannot be securely stated.

Listening children diagnosed with ASD feature receptive and expressive vocabulary inferior to children with typical development. In addition, the greater the degree of ASD impairment, the lower the performance regarding those skills²⁶. The case reported here is about a child diagnosed with ASD, support level 3. In this sense, atypical development is expected. Similar to the variability observed in the development of children with ASD, there are also many variables involved in the development of children who make use of the CI.

In view of the results exposed here, deaf children with ASD support level 3, the use of the CI does not grant them the acquisition of oral language, even in cases of early intervention, as the lack of audibility for speech is not the only hindrance for the development of oral language.

There is a recommendation that children who make use of CI, when diagnosed with ASD, be referred to an intervention including strategies for both conditions. Thus, therapies grounded in the aural-oral approach may be combined with other types of intervention aiming at the ASD, such as the Science of the Applied Behavior Analysis (ABA), when recommended⁸.

The child described in this case report was referred to intervention with the use of the augmentative and alternative communication (AAC). Currently, the literature that describes the use of the AAC in children, CI users, with additional needs, is not robust enough to guide therapy, and further studies, which explore the potential for the use of AAC in clinical practice to users of CI, must be conducted²⁷.

A limitation in this study is the fact that it is a case study. Therefore, it was not possible to carry out statistical analyses or control intervening variables. In spite of that, in the face of the literature in the area, the obtained data, exposed here, describe, in a reliable way, one of the probable outcomes of the benefit in using the CI for the development of auditory and oral language skills in children with ASD support level 3. Further clinical studies are suggested with a larger sample and control of variables to verify the extent to which those results can be representative of the studied population.

CI and hearing rehabilitation centers must perform adaptations in their protocols and services in order to deliver therapeutic care to those children and their families. Multidisciplinary teams, including social workers and psychologists are needed to help in the evaluation as well as in the process of intervention²⁸.

The authors in this study are favorable to early intervention using auditory devices when there is proper referral, grounded in audiological and non-audiological factors in case of deafness with additional disabilities. This study is not expected to prevent the use of the CI in this population. It aims to expand the discussion on the theme and the awareness of the professionals involved in their treatment regarding the need of leveling parents' expectations, and the importance of scientific-based practice, elaborating individualized clinical planning, considering the child's potentialities, but also the intervening variables as well as the expected prognosis.

CONCLUSION

Scarce benefit by using the CI was observed for the development of auditory skills, and no evolution in the development of spoken language in 2 years and 9 months after the activation of the device in a deaf child diagnosed with ASD support level 3, undergoing early intervention and aural-oral rehabilitation.

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Authors' contributions:

VLDF: conception and design of the study, analysis and interpretation of data, preparation of the article and critical review for relevant intellectual content and final approval of the version to be presented for publication; VFM, TMD, FMM, AMSH and CCR: acquisition, analysis and interpretation of data, preparation of the article.