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Review articles

Evaluation and results of children with Autism Spectrum Disorder using cochlear implants: an integrative literature review

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ABSTRACT

Purpose: to synthesize the evidence of available studies in the literature regarding the benefit of the cochlear implant in children with additional diagnosis of autism spectrum disorder and to verify the protocols used to validate the abilities of auditory perception and oral language of this population.

Methods: an integrative literature review, searching in LILACS, MEDLINE/PubMed and SciELO databases and in the Google Scholar. Studies in Portuguese and English that assessed auditory and/or spoken language skills of children using cochlear implants with autism spectrum disorder were included.

Literature Review: 16 studies were included. In 72.18% of cases, autism spectrum disorder was diagnosed when the child was already using a cochlear implant. Studies have shown limited benefit from cochlear implants for the studied population. Parents need to be oriented regarding their expectations about the use of the device.

Conclusion: the benefit of using a cochlear implant for children with an additional diagnosis of autism spectrum disorder is limited and lower than the results obtained by children who do not have additional diagnoses. There is no standardized protocol for assessing auditory and language skills in this population.

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

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INTRODUCTION

Cochlear implants (CI) promote the access to speech sounds among children with severe or profound hearing impairment, enabling them to develop the auditory perception of speech and oral language[1], with quality of life similar to that of their normal-hearing peers[2]. However, the results of such an intervention may feature broad heterogeneity, limitations, and a slower pattern of development, due to some factors, such as the additional presence of disabilities other than hearing impairment[3,4].

The occurrence of Autism Spectrum Disorder (ASD) among children with profound hearing impairment is not rare[5,6], and the number of CI users have been increasing among this population[7]. The challenges for the professionals of the CI team may encompass the audiological diagnosis[8], the individual assessment of the CI due to the large variability of the prognosis[9], cochlear implant programming[10], the confirmation of the intervention benefits by means of standardized tests[9,11], and the speech-language therapy[8,12]. Currently, part of those difficulties may occur later as many children undergo the CI surgery before the diagnosis of ASD.

While ASD signs are usually recognized during the second year of age[13], the newborn hearing screening (NHS) enables the audiological diagnosis for hearing impairment in the first months of age, which reduces the age for hearing-aid fitting and, consequently, the CI surgery, which can be performed, in many cases, before twelve months old[14]. In addition, ASD symptoms can be masked by the hearing impairment, delaying the age of the diagnosis even more[9].

Regarding this theme, two literature reviews were published in the past two years. The evidence search for one of them was carried out in September 2019[15], and the other in May 2020[16]. In one of the studies[15], seven articles were included, which assessed 66 children with ASD, CI users, and the authors concluded that the CI might benefit this population. However, in the most recent literature review, published in 2022[15], the authors included 24 studies, which assessed 159 participants, and concluded that the results of the CI use among children with ASD are highly variable and significantly worse if compared to children without ASD. However, they pointed out that parents of such children reported positive experiences. In the review published in 2022, the authors adopted studies not available in English for full reading as the exclusion criterion. Thus, Brazilian studies were probably excluded, and consequently, Brazilian reality on the theme might not have been contemplated. There are study limitations, considering the use of different methodologies. In addition, both studies[15,16] do not discuss the protocols used for assessing hearing and language skills among that population, in spite of mentioning them in the result tables.

Learning the performance achieved by children who make use of cochlear implants with additional diagnosis of ASD, as well as the protocols used for the assessment of hearing and language skills among that population is essential for family guidance, support and advisory and for the proper therapeutic planning of the hearing rehabilitation process.

Therefore, considering the findings and speech-language pathologists' reports in the area of hearing rehabilitation, who refer to the increase in the demand and questions related to the use of CIs among children diagnosed with ASD, it was verified the need of a review to map the limitations and gaps in the scientific knowledge on the theme; fundamentally, for the contribution to the evidence-based practice within Brazilian reality.

Thus, the current review aimed to synthesize the evidence of studies available in the literature regarding the CI benefit among children with additional diagnosis of ASD, and verify the protocols used to assess the skills of auditory perception and oral language among this population in the therapeutic process.

METHODS

It is an exploratory descriptive research by means of an integrative literature review, following the steps[17]: (1) elaboration of the guiding question, (2) search in the literature and data collection, (3) critical analysis of the included studies, and (4) discussion of the results.

Eligibility criteria

PICOS acronym classification was considered to answer the following guiding question: "In what ways does the development of the auditory perception and speech skills occur among deaf children with additional diagnosis of ASD after the use of CI, and how are such skills assessed in this population?".

- P = Participants (deaf children with additional diagnosis of ASD);
- I = Intervention (use of the CI);
- C = Comparison (pre and post-CI surgery compared with children, users of CI, without diagnosis of associated impairments);

- O = Outcome (assessment of the hearing perception and/or oral language skills);
- S = Study design (primary analytical observational studies, including crosscut, longitudinal, cohort, case-control designs; primary descriptive observational studies, including sectional, prevalence, and crosscut designs; peer-assessed articles, theses and dissertations).

Articles were included, as follows: available in full, free access and/or by means of the Virtual Private Network (VPN); articles in Portuguese and English; studies assessing auditory and/or language skills after the CI among children with additional diagnosis of ASD. As exclusion criteria, were adopted: other review studies, studies which assessed children with other comorbidities, without separated results for those with ASD, brief messages, letters to the editor and event annals.

Sources of information and search strategies

The search strategy was elaborated by combining key words and proper truncations for each electronic database: Latin American and Caribbean Health Sciences Literature (LILACS), MEDLINE via Public Medicine Library (PubMed), and Scientific Electronic Library Online (SciELO). Gray literature was also used as source of information by means of search in the Google Scholar, restricted to the first ten pages for each combination, as they were the most relevant ones (Chart 1).

Chart 1. Search strategies

Database/Search Tool	Search
Pubmed/Medline	("Child"[All Fields] OR "Child"[MeSH Terms] OR "Children"[All Fields] OR "Child, Preschool"[MesH Terms] OR "Child, Preschool"[All Fields] OR "Preschool Child"[All Fields] OR "Preschool Children"[All Fields]) AND ("Cochlear Implants"[MeSH Terms] OR "Cochlear Implants"[All Fields] OR "Cochlear Implantation"[MeSH Terms] OR "Cochlear Implantation"[All Fields] OR "Cochlear Implant"[All Fields] OR "Cochlear Implantations"[All Fields] OR "Cochlear Prosthesis Implantation"[All Fields]) AND ("Autistic Disorder"[MeSH Terms] OR "Autistic Disorder"[All Fields] OR "Autism Spectrum Disorder"[MeSH Terms] OR "Autism Spectrum Disorder"[All Fields] OR "Kanner Syndrome"[All Fields] OR "Infantile Autism"[All Fields] OR "Autism"[All Fields] OR "Early Infantile Autism"[All Fields] OR "Autistic Spectrum Disorders"[All Fields] OR "Autistic Spectrum Disorders")
LILACS	("Child" OR "Children" OR "Criança" OR "Niño" OR "Enfant") AND ("Cochlear Implants" OR "Cochlear Implantation" OR "Cochlear Implant" OR "Implante Coclear" OR "Implantação Coclear" OR "Implantación Coclear" OR "Implantation Cochléaire") AND ("Autistic Disorder" OR "Autism Spectrum Disorder" OR "Infantile Autism" OR "Autism" OR "Transtorno Autístico" OR "Autismo" OR "Autismo Infantil" OR "Trouble autistique") AND (db:("LILACS"))
SciELO	#1 (((TS=(child)) OR TS=(children)) OR TS=(criança)) OR TS=(crianças)) OR TS=(niño) AND #2 ((((TS=(cochlear implants)) OR TS=(cochlear implant)) OR TS=(cochlear implantation)) OR TS=(implante coclear)) OR TS=(implantación coclear) AND #3 ((((TS=(autistic spectrum disorder)) OR TS=(autism spectrum disorder)) OR TS=(autism)) OR TS=(transtorno autístico)) OR TS=(autismo infantil)
Google Scholar	"Child" OR "Children" OR "Crianças" OR "Criança" AND "Cochlear Implants" OR "Cochlear Implantation" AND "Autistic Disorder" OR "Autism" OR "Autism Spectrum Disorder" "Criança" OR "Crianças" AND "Implantes Cocleares" OR "Implante Coclear" AND "Autismo" OR "Desordem do Espectro Autista" OR "Transtorno do Espectro Autista"

The retrieved records were managed by the EndNote® Web software (https://myendnoteweb.com). In the same software, the duplicate identification was performed. Subsequently, the records were manually saved for the selection step.

Selection of the evidence sources

Search was conducted by the authors independently, according to the established inclusion and exclusion criteria. A fourth reviewer, experienced in the area of hearing rehabilitation, solved any disagreements between them regarding the studies to be included.

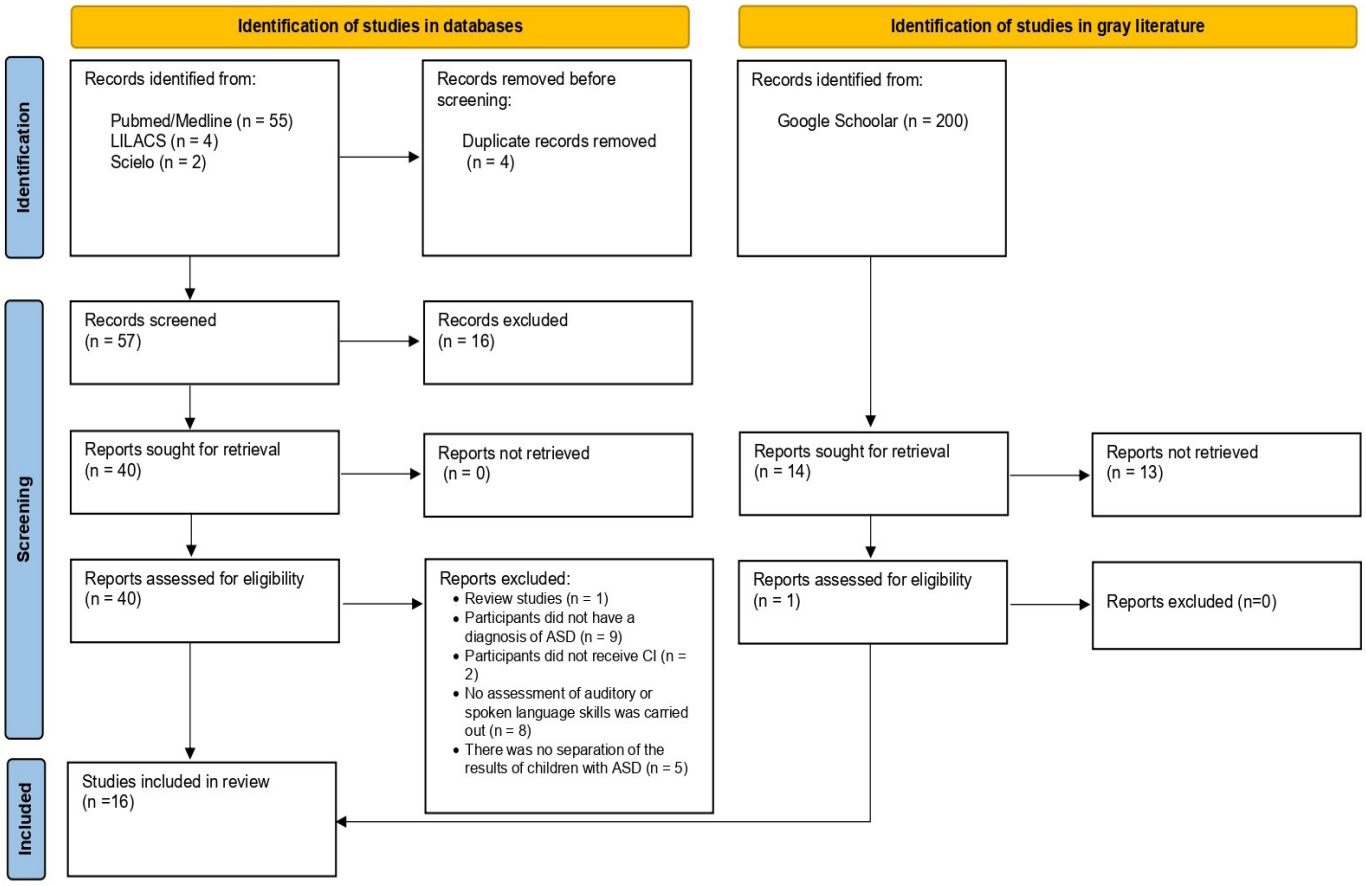
First, the publications were analyzed by title and abstract. Subsequently, the authors performed the full reading of the selected studies and came to a consensus regarding their inclusion in the integrative review.

Data analysis

A table in EXCEL was elaborated to insert the collected data by the reviewers, as follows: case studies, used tests, main results and conclusion. The identified complementary data were: year of publication, nationality and language of the published study. In this study, the assessment of the methodological quality and the classification of the level of evidence were held in the included studies.

LITERATURE REVIEW

Two hundred and sixty-one studies were found. After the application of the selection criteria, 16 studies[7,9,10,18-30] were included in this integrative review (Figure 1).



Captions: n=number of studies; CI=cochlear implant; ASD=autism spectrum disorder
Source: www.prisma-statement.org

Figure 1. Study selection flowchart

From the 16 included studies, most of them were conducted in the United States of America (USA) (n=4)[7,18,25,28]. Only one study[21] has Brazilian authorship. It is a doctorate thesis developed in the Postgraduation Program in Speech-Language Pathology of a public university.

The studies assessed two to 398 participants, with one to 30 participants diagnosed with ASD. The mean age of the assessed children who underwent the CI surgery was 34 months. It was still observed that from 157 children with ASD assessed in the studies, in at least 72.62% of the cases (n=114), the diagnosis of ASD was concluded when the child already made use of the CI (Chart 2).

The main results of the studies are in Table 1.

Chart 2. Characteristics of sources of evidence

Year	First author	Country	n		Average age at CI	ASD Diagnosis	
			Total	ASD		Before CI	After CI
2021	Jenks[18]	USA	30	30	41	09	21
2021	Mancini[19]	Italy	22	22	34	05	17
2018	Mesallam[20]	Saudi Arabia	50	09	39	NI	NI
2019	Scarabello[21]	Brazil	30	10	29	02	08
2018	Motegi[22]	Japan	13	04	33	01	03
2016	Lachowska[10]	Poland	06	06	22	01	05
2016	Valero[23]	UK	22	22	30	0	22
2016	Mikic[24]	Serbia	14	04	15	0	04
2015	Eshraghi[25]	USA	30	11	18	02	09
2014	Meinzen-Derr[9]	Serbia	24	14	29	0	14
2013	Robertson[26]	Ireland	10	10	42	02	08
2013	Ozdemir[27]	Türkiye	12	03	37	NI	NI
2008	Johnson[28]	USA	02	01	52	0	01
2006	Daneshi[29]	Iran	398	04	NI	NI	NI
2004	Donaldson[7]	USA	07	05	48	03	02
2000	Hamzavi[30]	Austria	10	02	61	NI	NI

Caption: ASD=Autism Spectrum Disorder; CI=Cochlear Implant; n=number of participants; NI=the information is not included in the study; UK=United Kingdom; USA=United States of America.

Table 1. Main results of the studies included in the review

Year	First author	Tests*	Main results		Conclusion
			Hearing skills	Oral language	
2021	Jenks	PBK; CNC; MAIS	Thirty-three percent evidenced open-set auditory recognition after 4 or 5 months using the CI; 13.33% achieved speech recognition of monosyllables in a closed-set format, while 40% did not evidence any score changes in the MAIS scale, even improving their hearing thresholds by making use of the CI.	Thirty per cent used oral language after the CI; 13.33% spoke being helped by signs, and 13.33% use Sign Language; 13.33% used AAC, and 26.66% did not use any formal ways of communication.	Language development is significantly affected by the ASD and, consequently, few children achieve the use of oral language and school inclusion. However, the use of CI is supported for its potential to improve hearing and language skills, and in the interaction, at least in part of that population.
2021	Mancini	CAP; CL; MAIS; IT-MAIS; MacArthur Bates; CDI; PPVT; Common Protocol of Evaluation in Rehabilitation Audiology; TROG-2	13.6% did not evidence any benefits making use of the CI, in terms of auditory recognition; 31.8% were able to recognize environmental sounds or words; and 22.5% achieved the ability of speech understanding.	45.5% did not evidence any benefits regarding oral language; 72.7% did not achieve oral language or only spoke isolated words, and 18.2% could articulate simple phrases.	CI can benefit deaf children with ASD, although their results regarding speech perception and language development are worse than the findings among implanted children with hearing loss, but no comorbidities. Limited improvement in language skills was found in most children with more severe ASD.
2018	Mesallam	MAIS; MUSS	Average scoring in MAIS: 18.88%; the group with ASD had worse result of speech perception than the other children with assessed comorbidities.	Average scoring in MUSS: 7.33%; 8 out of 9 assessed children did not use the oral language to communicate after the CI.	The benefits of the CI in deaf children with ASD are limited and cannot be compared to the results in children without additional disabilities.
2019	Scarabello	MUSS; ABFW (vocabulary); PPVT; MacArthur; OCC; DDST I; Language Categories	Not assessed	Performance worse than expected in the expressive vocabulary of the ABFW test, with a minimum of 0% in the verbal designation of words until 5 years of age, when there was average scoring of 9% in the "means of transport" category. All participants scored 0 in the receptive vocabulary. Most of them (n=8) remained in the Language Category 1 – absence of speech. No participants achieved over 3 element sentence building. Mean of 18.7% in the MUSS.	Communicative performance of children with hearing loss and ASD below the expected, despite discrete improvement. There is worse performance in this population than in the population user of CI without additional disabilities.
2018	Motegi	Enjoji Scale of Infant Analytical Development	Not assessed	Children with ASD, HI and ID showed significant delay in language development and greater delay in development of emotional and social behavior when compared with children of groups with ID only.	There is development delay characteristic in children with additional comorbidities, users of CI.
2016	Lachowska	Response to music; Ling 6 Sound Check; Response to own name; Test of onomatopoeic words; Parents' questionnaire with questions on hearing behavior	Three out of 6 children showed speech recognition of their own names, according to their parents. There was no hyperacusis using the CI. Parents did not observe any improvement in the children's visual contact or anxiety. Only one child, the oldest, could undergo the free-field audiometric test.	Only one out of 6 assessed children, the oldest one, used isolated words to communicate. The other children only uttered screams.	Traditional methods for skill assessment of children without comorbidities, who make use of CI, are insufficient to assess children with ASD, users of CI.
2016	Valero	CAP; CL	Fifty-nine percent of the children rejected the CI, resulting in its discontinued use at some moments, and 27.2% quit using the CI. Complete rejection of the device was reported at a mean age of seven, ranging from 2 months to 10 years after the CI surgery. Among those, 2% reported hyperacusia.	Most of the children communicated by means of more than one method (e.g.: speech, signs and/or AAC); 27.2% made exclusive use of non-verbal communication; 18% used speech as their main way of communication; 41% were able to utter some phrases or words to communicate.	Further research is required to develop and/or change ASD assessment in children who make use of CI. Moreover, longitudinal studies of speech expression/perception in children with ASD before and after the implantation are required.

Year	First author	Tests*	Main results		Conclusion
			Hearing skills	Oral language	
2016	Mikic	CAP; SIR	In children diagnosed with ASD later, auditory skills developed slowly. Depending on their individual skills, at 6 years of age, some children were able to identify environmental sounds or discriminate speech sounds.	The speech intelligibility in children with ASD was classified as category 2 at best, with none or very little progress until age 6, despite intensive speech-language therapy.	Communication skills were strongly affected by a degree of expression of autism characteristics. Accurate and valid triage instruments for babies and toddlers are further necessary to reduce the detection age of ASD in children with congenital deafness, and they must be included in the pre-surgical procedures of CI.
2015	Eshraghi	Early Speech Perception Test; Multisyllabic Lexical Neighborhood Test; PBK; Questionnaire to parents	Sixty-seven percent of the children advanced for the recognition of simple phrases or hearing understanding, while 33% progressed to speech detection.	Sixty percent of the children were able to communicate using simple phrases and some sentences, while 33% remained in category 1 (utterances).	The use of the CI may favor expressive and receptive language in children with hearing impairment and ASD, even if those children do not develop their skills as the others without comorbidities.
2014	Meinzen-Derr	Revised Gesell Developmental Schedules; PLS-4; CELF; VABS	Barely a third of the children were considered oral communicators.	The scores of receptive language ranged from 19 to 22%; the children did not show the same development as users of CI without comorbidities.	It is fundamental to consider language and communication delays in children who make use of the CI, and not assume that they are simply results of hearing loss in all situations. With the commonly observed disorders in children with deafness and ASD, focusing on a single approach (e.g.: oral language) is not enough to help communicative development among that population.
2013	Robertson	CAP; SIR	Six out of 10 assessed children made consistent use of the CI, 2 made inconsistent use, and 2 of them discontinued its use. One child achieved simple /familiar phrase understanding after seven years of CI use.	Six out of 10 children remained non-verbal after the CI. Only one became user of the oral language.	The results after the CI in children with ASD may vary, and it is possible to benefit from the device with time and proper support. Some may reject it completely. In case of ASD suspicion or confirmation before the CI, it is important adequate counselling to parents regarding their expectations.
2013	Ozdemir	MAIS; LiP	Three out of four children with autism evidenced limited use of the CI, while one did not use the device. Progress was observed in auditory test results after 24 months of its implantation, with similar performance among children with limited use of the device, and lower score for the child who did not use the CI.	Not assessed	Factors like ASD may result in limited or no use of the CI. In such cases, it is essential the joint work of therapist, family and school in order to achieve more effective results on the development of hearing, language and quality of life.
2008	Johnson	RDLS, MacArthur-Bates; CDI;10-minute recording in free playful situation with mother and 5 minutes by themselves	Not assessed.	The child already presented utterances before the CI, with the support of communicative signs. At 5 years and 4 months of chronological age, after 12 months using CI, scores of receptive and expressive language equivalent to age 23 months were observed, in addition to greater involvement in symbolic playful activities.	Children with multiple disabilities feature unique challenges for result assessment after the CI. Interactive parent-child tasks are a valid representation of the interaction skills in children with hearing loss and other comorbidities.
2006	Daneshi	Persian auditory perception test for the hearingimpaired	Limited improvement and not statistically significant differences in speech perception were identified in the group of children with CI before and after the use of the CI.	Not assessed	The idiosyncrasies involving the prognosis of children with varied disabilities associated to deafness are expressive, and point to limitation in their speech development, posing challenges in the aspects of evaluation, planning and implementation of rehabilitation.

Year	First author	Tests*	Main results		Conclusion
			Hearing skills	Oral language	
2004	Donaldson	MAIS, MacArthur; CDI; PPVT, GASP, Questionnaire with parents	The greatest difference observed by parents was their children's pleasure for music. It was reported that all 7 children uttered, responded to the sound and showed satisfaction with music, at least some times after the CI. Only 2 out of seven could answer the GASP, and one of the children achieved 100% of word recognition in the test after 2 years using the CI. MAIS score increased 30% after the CI.	Only one child used oral language. It was possible to assess expressive vocabulary in only 3 children of the 7 participants, and one of them improved after 2 years of CI use.	The gains after the use of the CI were small. However, when compared these children before and after the intervention, they showed some progress. Improvement in behaviors and interaction points to benefit in their quality of life after the CI, although it is difficult to quantify that.
2000	Hamzavi	EARS	After 1 year, they could understand "hot", "come", "no", "go", "shoe", "mom" and "dad". Three years after the surgery, one of the children achieved scores of 83.3% in the word recognition and vocabulary of 30 words.	Three months after activating the CI, one of the children started using their voice to communicate. Another child with ASD started uttering.	Children with additional diagnoses are not unfit for the CI, but not all of them are considered good candidates. Parents and children's lives can be positively changed after the CI. However, parents must have realistic expectations.

Captions: ABFW= Child Language Test; AAC=Alternative Augmentative Communication; CAP=Categories of Auditory Performance; CDI= Communicative Development; Inventories, CELF= Clinical Evaluation of Language Fundamentals; CL=Categories of Language; CNC= Consonant Nucleus Consonant Monosyllabic Word Test; HI=Hearing Impairment; ID=Intellectual Disability; DDST I=Denver Development Screening Test; EARS=Evaluation of Auditory Responses to Speech test Battery; GASP = Glendonald Auditory Screening Procedure; CI=cochlear implant; IT-MAIS= Infant Toddler - Meaningful Auditory Integration Scale; MAIS= Meaningful Auditory Integration Scale; LiP= Listening Progress Profile; MUSS= Meaningful Use of Speech Scale; CBO=Communicative Behavior Observation; PBK= Phonetically Balanced Kindergarten Test; PLS-4=Preschool Language Scales-4th edition; PPVT=Peabody Picture Vocabulary; Pre-CI=before the cochlear implant; Post-IC=after the cochlear implant; RDLS=Reynell Developmental Language Scales; SIR=Speech Intelligibility Rating; ASD=Autism Spectrum Disorder; TROG-2= Test for Reception of Grammar; VABS=Vineland Adaptive Behavior Scales.
*Tests for the assessment of auditory and oral language skills.

Tests and protocols used in the assessment

Assessment protocols for the auditory perception of speech and language development already used and standardized for children with hearing loss without additional diagnoses were observed. Thus, similar to the evaluation of children without comorbidities, the chosen protocols must consider not only the proper level of development for the age, but also the child's level of language and auditory perception[23].

In order to evaluate the hearing skills, most studies used the *Infant Toddler Meaningful Auditory Integration Scale* (IT-MAIS), and the *Meaningful Auditory Integration Scale* (MAIS)[18-20,27], the classification in Hearing Loss Categories[19,23,24], and/or speech perception testing, conducted with the child, such as the *Phonetically Balanced Kindergarden Test* (PBK)[18,25]. Regarding the evaluation of language development, the *Peabody Picture Vocabulary Test* (PPVT)[19,21], classification in Language Ability Categories[19,21,23,24,26], and the *Meaningful Use of Speech Scale* (MUSS)[20,21] were used.

Most researchers[7,10,18-21,27] included scales and/or questionnaires applied by means of an interview with the parents. That can be justified due to the difficulty in obtaining reliable results in standardized tests with that population. Many times, researchers[7,18,28,30] claimed that it was not possible to apply standardized measures to assess the development of hearing and language skills among children diagnosed with ASD, who make use of CI, due to the severity of their development delay. In a study[18] published in 2021, the authors reported that MAIS was the only protocol of speech perception capable of being applied. In another study, only two of seven assessed children were able to respond the *Glendonald Auditory Screening Procedure* (GASP)[7].

Standardized tests must be applied whenever it is possible. However, the use of standard scores cannot be informative due to the performance really below their age expectations among that population[7]. Traditional methods of assessment for CI results are not enough for the complete evaluation of the benefits for the use of the device in children with additional ASD diagnosis[10]. The observed performance cannot always be expressed in percentages, and measuring the success of that population with their CI is usually a subjective process[30]. Background information on language development about those children may contribute to better understanding on how the CI affects their oral language. Moreover, brief samples of joint

attention and symbolic playful behaviors recorded in an individual playful situation, and with their parents, may add complementary information to the one from clinical measures[28]. Therefore, the evaluation to measure the progress with the use of the device is challenging and comprehensive, considering the communicative limitations from the ASD[10].

These data point out the need of standardized measures to assess the use of CI among children additionally diagnosed with ASD[7], as well as the importance for professionals to share their knowledge on that population[10].

Reported results

In this review, it was observed that, in most cases, the ASD diagnosis was concluded when the child was already making use of the CI. This can be justified by the fact that the UNHS enables the diagnosis of hearing impairment earlier than the outcome of the typical features for ASD, or before they can be perceived. In that sense, ASD probability must always be considered by the professionals involved in the guidance of babies' parents referred to the use of the CI[24].

The CI can benefit hearing-impaired children also diagnosed with ASD, although all studies included in this review reported worse results for that population than those observed in children without additional disabilities[7,9,10,18-30]. There is a series of reasons for such differences in the results between different populations, and they include the presence of learning and communicative disorders, associated motor impairment, sensory sensitivity, among other disorders observed in children with ASD, regardless of the presence of hearing impairment[23].

The communicative development of children with ASD who make use of CI is also lower when compared with other groups featuring comorbidities. In a study[20], the authors compared the development of a group of children who used CI, with additional diagnosis of Attention Deficit/Hyperactivity Disorder (ADHD), and evidenced worse results in the group of ASD regarding the development of auditory and language skills. In another study[22], the authors observed worse results in the group of children diagnosed with ASD, users of CI, than in the group of children with intellectual disability (ID), and reported that this finding seems to reflect the ASD features, such as impaired social interaction, atypical communication and restricting, repetitive behaviors.

Actually, ASD and hearing impairment may coexist. Therefore, the possibility of an ASD diagnosis cannot be disregarded simply because the child has already been diagnosed with hearing loss. It is important to pay attention to delays in the communicative development of children who make use of a CI, not assuming that it is simply the outcome of their hearing impairment[9]. If a child with a CI does not develop his/her language properly, an evaluation to comorbidities, such as ASD, must be recommended[25]. The main characteristics of atypical communication, atypical social interaction and restrictive, repetitive behaviors should not be attributed to hearing impairment. Oral language may be delayed in hearing-impaired children, but their communicative development should not present such atypical features[9].

Part of deaf children with additional diagnosis of ASD may benefit from the use of the CI and, along the time and proper support, they may develop functional speech skills[26]. Results of studies included in this review reported that part of that population was able to communicate by means of the oral language[18], increasing their utterances[7], either achieving the enunciation of words[10,19,21] or simple phrases[19,23,25]. In spite of that, most children assessed in the studies did not develop speech, and some of them could not communicate by any formal methods, and others could communicate by other methods, such as the Sign Language and/or Augmentative and Alternative Communication. Therefore, ASD should not prevent the referral for CI[26], but it should be in mind that that population may never reach the score of pre-language deaf children, without any additional disabilities who make use of the device[30], and oral communication may not be a realistic goal[7].

Another important point to be discussed is that children with ASD may completely reject the use of the CI speech processor[26]. In a study[23], the authors reported that 59% of the assessed children made inconsistent use of the CI, and 27.2% of them discontinued the use of the device completely after some years. Other authors[26] also pointed that six, out of ten children assessed, made consistent use of the CI, while two of them used it in an inconsistent way, and two did not use it at all. Literature reports that diagnosis-related variables may influence the use of the CI processor by children with multiple disabilities. Additionally, it is suggested that studies include objective data in their investigations, such as the datalogging records[31].

Moreover, the possibility of hyperacusis in this population must be considered[25].

Despite the low incidence of oral language after the CI in children with ASD, parents tend to recommend its use to other families in similar situation, although claiming that their children had a performance worse than expected[7,25]. Some parents observed changes in their children's communicative behavior and greater awareness of the environment[7]. There is also the report of increased response to music[7,10], but poor improvement in the visual contact[25].

Briefly, post-CI expectations are very different for children diagnosed with ASD when compared with children without additional diagnoses[7,9,10,18-30]. In that sense, parents must be aware that the use of CI will not alter characteristic behaviors of ASD, nor will somewhat impact on its severity[7,25].

Other considerations

In a study[32] published in 2019, the authors concluded that hearing impairment was nine times more prevalent in individuals with ASD when compared with the population at large. Despite such rates of occurrence have still been under discussion, authors point out that thorough audiological screening is recommended in all suspicious cases of ASD in order not to delay the diagnosis of hearing impairment, in case of comorbidities[33].

Given the evidenced variables and the level of support, goals and expectations of performance vary for each child with ASD undergoing a CI, and they must be discussed in an individualized way with each family[7], in order to set realistic expectations. Advisory must also focus on discussing with parents aspects regarding a favorable prognosis, in case of mild degrees of ASD, absence of intellectual disability or neurological disorders, early implantation and family support[25].

In addition, the discussion is recommended in the literature, before the intervention, on the future impact of probable comorbidities, not diagnosed before the procedure, such as ASD, on all children referred to the CI, thus, preventing them from unreal expectations[7,23,25,26].

Apart from parents, professionals involved in the rehabilitation must be aware that children with multiple disabilities require rehabilitation with unique and individualized strategies[29]. In other words, despite the consideration that parents opt for the CI aiming at their children's oral language[7], focusing only on the speech approach during the rehabilitation of a child, user of CI with additional diagnosis of ASD, is usually insufficient to help improve his/her communication[9]. Therefore, other commonly used approaches for ASD rehabilitation must be applied[25]. By using strategies applied to atypical patterns of language in those children, that may build a path towards more effective communication[9]. Insisting on exclusively oral communication to that population may contribute to their delay in understanding the pre-linguistic foundations of communication, critical for their communicative enhancement[9].

This study evidences some limitations. One of them is the fact that search was not carried out in all of the available databases. Moreover, the analysis of the methodological quality of the included studies in this integrative review was not conducted. In this sense, case studies were included, as well as those, which carried out the assessment only by using questionnaires and/or scales oriented to the children's parents. Therefore, the need of further research in the area is pointed out, fundamentally in order to establish specific protocols for the assessment of children with additional diagnosis of ASD, who make use of CI, considering the idiosyncrasies of that population and the interfering variables in the results.

ASD must not prevent the referral for CI, although it is fundamental that parents and professionals establish real expectations of prognosis in such cases. In addition, parents of children without additional diagnoses must also be advised before the CI on the possibility of further diagnoses, which will influence the development of auditory, language skills in children.

This study is expected to guide the assessment of that population in rehabilitation services. Additionally, it aims to guide multidisciplinary teams regarding treatment expectations, guidance and advisory of those children's families.

CONCLUSION

It can be concluded with this study that:

- The benefit with the use of the CI by that population is limited and worse than the benefit obtained by children, users of the device, who were not diagnosed with comorbidities;
- Great part of the children, users of CI with additional diagnosis of ASD, does not advance to exclusively oral communication, and the use of the device does not minimize characteristic behaviors of ASD;
- There is not a standardized protocol for assessing auditory perception and oral language skills in children, users of CI and diagnosed with ASD;

- The assessment of hearing perception and oral language in this population must be held in an individual and adapted way, and the child's age must be considered, as well as his/her level of development;
- The use of questionnaires with parents and filming parents with their child may help the assessment process of the auditory and speech skills among that population.

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Authors' contributions

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

JMS, LCV, TMS: acquisition, analysis and interpretation of data, preparation of the article;

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