Revista CEFAC

eISSN 1982-0216

DOI: 10.1590/1982-0216/20232539522 | Rev. CEFAC. 2023;25(3):e9522

Review articles

Speech perception and spoken language of children with auditory neuropathy spectrum disorder: a systematic literature review

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ABSTRACT

Purpose: to analyze the test results of speech perception and spoken language in children with hearing loss and auditory neuropathy spectrum disorder, users of hearing aids or cochlear implants.

Methods: a systematic review of the literature based on the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines. Consultation was performed in databases, considering studies from 1996 to 2021, selecting the studies that presented the results of speech perception or spoken language in children with bilateral auditory neuropathy spectrum disorder, with no structural alterations of the ear and/or vestibulocochlear nerve, or other associated impairments. Descriptive analysis was performed.

Literature Review: among 1,422 studies found, 15 were included. Variability in the sample size, types of studies, evaluation procedures and methodological questions were observed. The cochlear implants and hearing aids contributed to the development of speech perception and spoken language in children with auditory neuropathy spectrum disorder, since skills such as auditory comprehension and speech intelligibility, were achieved.

Conclusion: 15 studies suggest that cochlear implants and hearing aids may be effective for speech perception and spoken language development in children with auditory neuropathy spectrum disorder, with no other associated impairments. The need for further research with a high methodological rigor is highlighted.

Keywords: Cochlear Implants; Hearing Aids; Hearing Loss; Speech Perception; Child Language

Study conducted at Universidade de São Paulo, Hospital de Reabilitação de Anomalias Craniofaciais - HRAC/USP, Bauru, São Paulo, Brazil.

Financial support: Nothing to declare

Conflict of interests: Nonexistent

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Received on: October 10, 2022
Accepted on: March 30, 2023

INTRODUCTION

The term "Auditory Neuropathy" was first published by Starr et al. (1996)[1] to describe a possible impairment of the inner hair cells, the auditory nerve and/or the failure between the synapses of the inner hair cells and the auditory nerve[1,2]. Over the years, studies in the area have used different nomenclatures, such as "Auditory Neuropathy", "Auditory Dyssynchrony" and "Auditory Neuropathy Spectrum Disorder" – ANSD[1,3,4].

The diagnosis of auditory neuropathy is verified by the presence of Evoked Otoacoustic Emission, register of abnormal or absent Brainstem Auditory Evoked Potentials (BAEPs) in the neural response and/or the presence of Cochlear Microphonism, observed in the BAEPs or in the Electrocochleography[5,6]. In addition, when there is a hearing loss, the degree can vary from mild to profound and the difficulty in understanding speech in silence and, especially, in noise, is disproportionate to the hearing loss presented[4,7].

The variability found in the Auditory Neuropathy Spectrum (ANS) diagnostic tests demonstrates the heterogeneity of its pathophysiology[8,9]. Individuals with ANS may present alterations in the perception of sound stimuli and in the ability of temporal auditory processing, due to the failure in neural synchrony[7,10].

The use of electronic devices to access speech sounds, such as the Hearing Aid (HA) and the Cochlear Implant (CI), brings the possibility of minimizing the impact of auditory sensory deprivation and enabling the development of spoken language[11,12].

When the results provided by the HA regarding auditory speech perception are limited, the CI may be indicated, since this device partially replaces the function of the cochlea hair cells, compensating for the alteration of neural synchrony[13,14]. However, the literature reinforces the importance of considering the individuality of each case, and caution is necessary in relation to the conduct of indicating the cochlear implant[6,9,13].

A previous study systematically reviewed the existing evidence regarding the results of the auditory abilities of children using CI with auditory neuropathy spectrum disorder, verifying advances in the detection of speech, in the discrimination and recognition of words and sentences after surgery, and no differences were observed in these abilities when compared to children with sensorineural hearing loss who used cochlear implants[15].

However, further research is still needed in order to understand in more detail the effectiveness of the intervention with HA and/or CI in the performance of speech perception and spoken language of children with ANSD.

Studies that systematize the evidence produced over the years in this area may support the establishment of guidelines and scientific parameters that assist in the conduct of the auditory rehabilitation process, mainly related to the technological device to access to speech.

In view of the above, this study aimed to verify the results of speech perception and spoken language of children with hearing loss and auditory neuropathy spectrum disorder, users of hearing aids or cochlear implant.

METHODS

Research Strategy

Systematic review of the literature registered in the International Prospective Register of Systematic Reviews (PROSPERO) system, under number CRD42021273562[16], following the recommendations of the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines[17].

The review was conducted based on the guiding question: Were the cochlear implant and the hearing aid effective for the development of the perception of speech and spoken language in children with auditory neuropathy spectrum disorder?

Initially, a search was made in the Cochrane Library, Regional Portal of the Virtual Health Library (VHL) and PROSPERO databases, without verifying review studies with the proposed theme.

Thus, a bibliographic search was carried out in national and international journals indexed in the electronic databases: PubMed/MEDLINE, Web of Science, SCOPUS, Embase, Scientific Electronic Library Online (SciELO), VHL and Brazilian Digital Library of Theses and Dissertations (BDTD).

Health Sciences Descriptors (DeCS) in Portuguese and Spanish and their English counterparts in the Medical Subject Headings (MeSH) were used, as well as keywords related to the theme of the study. The descriptors and words were combined using the Boolean operators "OR" and "AND", as shown in Chart 1.

Chart 1. Search strategy

Search strategy	
English	((Hearing Aids) **OR** ("Hearing Aids") **OR** (Hearing Aid) **OR** ("Hearing Aid") **OR** (Cochlear Implants) **OR** ("Cochlear Implants") **OR** (Cochlear Implant) **OR** ("Cochlear Implant") **OR** (Cochlear Implantation) **OR** ("Cochlear Implantation")) AND ((Auditory Neuropathy) **OR** ("Auditory Neuropathy") **OR** (Auditory Neuropathies) **OR** ("Auditory Neuropathies") **OR** (Auditory Dyssynchrony) **OR** ("Auditory Dyssynchrony") **OR** (Auditory Dys synchrony) **OR** ("Auditory Dys synchrony") **OR** (Auditory Dys-synchrony) **OR** ("Auditory Dys-synchrony") **OR** (Auditory Neuropathy Spectrum Disorder) **OR** ("Auditory Neuropathy Spectrum Disorder")) AND ((Speech Perception) **OR** ("Speech Perception") **OR** (Auditory Perception) **OR** ("Auditory Perception") **OR** (Child Language) **OR** ("Child Language") **OR** (Language Development) OR ("Language Development") **OR** (Verbal Learning) **OR** ("Verbal Learning") **OR** (Language Development Disorders) **OR** ("Language Development Disorders") **OR** (Language Disorders) **OR** ("Language Disorders") **OR** (Rehabilitation of Speech and Language Disorders) **OR** ("Rehabilitation of Speech and Language Disorders"))
Portuguese	((Auxiliares de Audição) **OR** ("Auxiliares de Audição") **OR** (Implantes Cocleares) **OR** ("Implantes Cocleares") **OR** (Implante Coclear) **OR** ("Implante Coclear")) AND ((Neuropatia Auditiva) **OR** ("Neuropatia Auditiva") **OR** (Neuropatias Auditivas) **OR** ("Neuropatias Auditivas") **OR** (Dessincronia Auditiva) **OR** ("Dessincronia Auditiva") **OR** (Desordem do Espectro da Neuropatia Auditiva) **OR** ("Desordem do Espectro da Neuropatia Auditiva")) AND ((Percepção da Fala) **OR** ("Percepção da Fala") **OR** (Percepção Auditiva) **OR** ("Percepção Auditiva") **OR** (Linguagem Infantil) **OR** ("Linguagem Infantil") **OR** (Desenvolvimento da Linguagem) **OR** ("Desenvolvimento da Linguagem") **OR** (Aprendizagem Verbal) **OR** ("Aprendizagem Verbal") **OR** (Transtornos do Desenvolvimento da Linguagem) **OR** ("Transtornos do Desenvolvimento da Linguagem") **OR** (Transtornos da Linguagem) **OR** ("Transtornos da Linguagem") **OR** (Reabilitação dos Transtornos da Fala e da Linguagem) **OR** ("Reabilitação dos Transtornos da Fala e da Linguagem"))
Spanish	((Audífonos) **OR** ("Audífonos") **OR** (Audífono) **OR** ("Audífono") **OR** (Implantes Cocleares) OR ("Implantes Cocleares") **OR** (Implante Coclear) **OR** ("Implante Coclear") **OR** (Implantación Coclear) **OR** ("Implantación Coclear")) AND ((Neuropatía Auditiva) **OR** ("Neuropatía Auditiva") **OR** (Neuropatías Auditivas) **OR** ("Neuropatías Auditivas") **OR** (Desincronía Auditiva) **OR** ("Desincronía Auditiva") **OR** (Espectro de Desórdenes de la Neuropatía Auditiva) **OR** ("Espectro de Desórdenes de la Neuropatía Auditiva")) AND ((Percepción del Habla) **OR** ("Percepción del Habla") **OR** (Percepción Auditiva) **OR** ("Percepción Auditiva") **OR** (Lenguaje Infantil) **OR** ("Lenguaje Infantil") **OR** (Desarrollo del Lenguaje) **OR** ("Desarrollo del Lenguaje") **OR** (Aprendizaje Verbal) **OR** ("Aprendizaje Verbal") **OR** (Trastornos del Desarrollo del Lenguaje) **OR** ("Trastornos del Desarrollo del Lenguaje") **OR** (Trastornos del Lenguaje) **OR** ("Trastornos del Lenguaje") **OR** (Rehabilitación de los Trastornos del Habla y del Lenguaje) OR ("Rehabilitación de los Trastornos del Habla y del Lenguaje"))

Study Selection Criteria

For the selection of primary studies, the following criteria were considered:

- Types of studies: those in Portuguese, English or Spanish, published in the period from 1996 to 2021. The delimitation of studies starting from 1996 is justified by the fact that it was in this year the term "auditory neuropathy" and its description was first published. When publication was available, dissertations and theses were replaced by published work.
- Population: children with bilateral auditory neuropathy spectrum disorder, inserted in the context of spoken language, with no structural alterations of the ear or vestibulocochlear nerve (CN VIII) or other associated impairments, such as syndromes.
- Intervention: use of the bilateral hearing aid or unilateral or bilateral cochlear implant or use of the unilateral cochlear implant with contralateral (bimodal) hearing aid.
- Clinical outcomes: results expressed as a percentage of correct answers or classifications of scales that demonstrate the performance in the perception of speech and/or spoken language.

Data Analysis

The studies were extracted from the databases and organized using the EndNote software (online version), and repeated work was manually excluded.

The evaluation took place in two stages, by the Rayyan selection platform.

In the first stage, a blind evaluation by two reviewers selected the studies with potential relationship with the theme addressed by reading the titles and abstracts. In the second stage, two reviewers through blind evaluation analyzed the full text of the studies in which inclusion or exclusion was not possible only by reading the title and abstract, so as to define which studies fulfilled the selection criteria.

When there were disagreements among the judges regarding the inclusion or exclusion of the studies in the previous stages, another judge was called as a tie-breaking criterion. The third judge was able to analyze the title, abstract and full text (when it was not possible to include or exclude the study only by reading the title and abstract).

A protocol was used to analyze the research resulting from the previous stages, containing the following information: authors, year of publication, objectives, population, procedures for assessing speech perception and/or language, main results, level of evidence of the studies and risk of bias. One author organized these data, which were analyzed by another author and, if there were disagreements, a discussion was held to reach consensus.

To determine the level of evidence, the classification adapted by Cox (2004) was used[18].

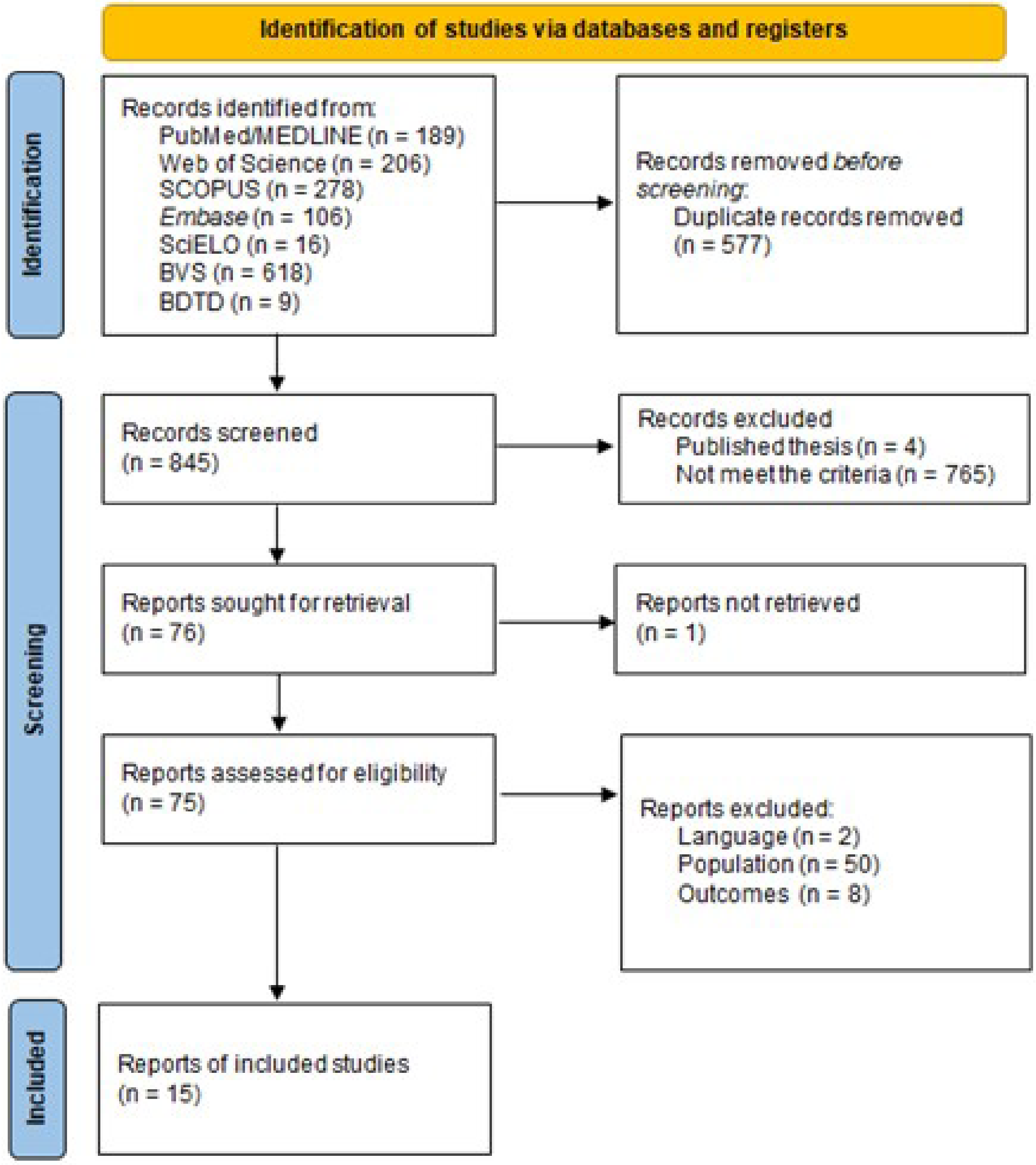
To assess the risk of bias, the critical appraisal tools appropriate for this type of study from the Joanna Briggs Institute were used[19]. The risk of bias was characterized according to the frequency of "yes" answers, which meant adequacy of the study. The risk of "high" bias corresponded to the frequency of "yes" responses less than or equal to 49%, "moderate" between 50% and 69% and "low" greater than 70%. These tools were applied by the same reviewers who evaluated the articles in the previous stages, through consensus and, in case of disagreements, a discussion was held to reach consensus.

It is worth noting that the literature review has been updated in order to contemplate all possible studies that were published by the end of 2021.

LITERATURE REVIEW

Considering the importance of monitoring the results of the intervention, both with the hearing aid and with the CI, this study aimed to analyze the results of tests of speech perception and spoken language of children with auditory neuropathy spectrum disorder. This objective aims to answer the guiding question of the study, which analyzes the effectiveness of the devices in this population.

Figure 1 shows the diagram of the results of this search.



Source: Page et al. (2020)[17]

Figure 1. Diagram of systematic review results

The research identified 1,422 studies in the databases used. After the duplicates were removed, 845 papers were screened. Of these, four dissertations/theses had already been published, being disregarded from the analysis and 765 studies did not contemplate the selection criteria.

After reading the title and abstract, 76 studies showed a relationship with the proposed theme and in only one the full text was not recovered, totaling 75 studies included for the second stage. After full analysis, 60 studies were excluded for different reasons, such as language, population and clinical outcomes, and 15 were included in this study. Taking into account the period elapsed between the first description of auditory neuropathy and the current advances in audiological diagnosis and intervention, it can be inferred that the number of studies found is relatively low.

Chart 2 shows the synthesis of the characteristics of the included studies.

Chart 2. Summary of included studies

AUTHOR/YEAR	OBJECTIVE	POPULATION	AGE AT EVALUATION	SPEECH PERCEPTION PROCEDURE	CLINICAL OUTCOME PROCEDURE	MAIN FINDINGS	LEVEL OF EVIDENCE
Trautwein et al. (2000)[13] Case report	To present the potential benefits of CI in a case of a child with ANSD.	1 boy that received unilateral CI Profound hearing loss.	4 years and 3 months.	*Ling's sounds *The Early Speech Perception test (ESP) *Test of Auditory Comprehension	Not provided	The child showed significant improvement in speech perception after implantation. When compared with 10 children who had cochlear hearing loss and used CI, their performance in the skills evaluated was similar.	5
Lin et al. (2005)[23] Case report	To report the case of a Mandarin speaking child with ANSD, who received the CI.	1 boy that received unilateral CI Profound hearing loss	3 years	*Mandarin Auditory Perception Test Battery (MAPTB)	Not provided	There was a significant improvement in speech perception skills after the CI.	5
Melo et al. (2008)[30] Case-control study	To compare the performance of speech production of two groups of children using CI, one group with sensorineural hearing loss and another group with ANSD, considering the time of CI use and the chronological age of the child at the time of evaluation.	10 CI users (5 children with ANSD paired with 5 children with sensorineural hearing loss). Profound hearing loss	2 years and 2 months to 5 years and 7 months.	*Evaluation procedure of speech perception in children with profound hearing impairment from five years of age *List of words as assessment procedure of speech perception for hearing impaired children	* Naming of figures of the ABFW Test – phonology and spontaneous speech test	The CI brought benefits to both groups of children, and no differences in performance in speech production were observed, which was higher for participants with a shorter period of sensory deprivation.	4
Rance et al. (2008)[26] Case-control study	To evaluate the speech perception skills of children with ANSD who received the CI and compare with a group of children with ANSD who use hearing aids and a group of children with sensorineural hearing loss, who have CI.	20 children with ANSD (10 users of unilateral, bilateral or contralateral CI and 10 users of bilateral HA) and 37 children with sensorineural hearing loss, with unilateral, bilateral CI or with contralateral HA. Severe/profound loss of CI group with ANSD. Mild and severe loss of the HA group with ANSD.	ANSD + CI group: 89.6 months. ANSD + HA group: 94.2 months. Sensorineural hearing loss group: 92.6 months	*Consonant-nucleus-consonant (CNC) words	*Diagnostic Evaluation of Articulation and Phonology test	There was a significant advance in the speech discrimination of children with ANSD, and the performance of children using CI and HA was similar in this ability. However, both groups presented speech perception results lower than those obtained by children with sensorineural hearing loss.	4
Fukushima et al. (2009)[35] Case series	To present the clinical course of children with the presence of otoacoustic emissions, hearing loss and cochlear implantation.	2 users of unilateral CI. Severe/profound hearing loss.	8 to 10 years.	*Monosyllabic speech perception tests	* Speech intelligibility test	The CI was effective and contributed to the development of the language of the cases presented.	5
Carvalho et al. (2011)[31] Cross-sectional study	To evaluate the auditory performance and the characteristics of the Electrically Evoked Compound Action Potential (eCAP) in the Auditory Nerve, in a group of children with ANSD who use CI.	18 children with ANSD using CI. Profound and severe hearing loss.	Not provided	* Procedure for the Evaluation of Profound Hearing-Impaired Children * List of Words as a Procedure for Evaluating the Speech Perception for Hearing Impaired Children	Not provided	The CI was effective for the development of auditory skills in 94% of the participants. No changes were observed in the threshold and amplitude characteristics of the eCAP for the two stimulation frequencies tested, and the electrical stimulation was able to compensate for the alteration of neural synchrony resulting from the ANSD.	4
Fei et al. (2011)[22] Case report	To present the case of an implanted patient, including preoperative evaluations and a series of postoperative evaluations, to provide preliminary clinical evidence of the efficacy of CI in patients with ANSD.	1 boy implanted unilaterally. Profound hearing loss.	4 years.	*Meaningful Auditory Integration Scale (MAIS) *Mandarin early speech perception test (MESP) *Categories of Auditory Performance (CAP)	*Speech Intelligibility Rating (SIR)	There were benefits for the patient presented, in relation to auditory sensitivity, speech recognition and communication, after the period of seven months of CI use.	5
Kim et al. (2013)[24] Case report	To report a case of a child with ANSD and CI.	1 boy who received unilateral CI. There is no reported degree of hearing loss.	2 years and 7 months.	*Phonetically Balanced Kindergarten (PBK): *Word List and the Common Phrases	*Peabody Picture Vocabulary Test (PPVT) *Expressive One-Word Picture Vocabulary Test (EOWPVT)	The cochlear implant can be an effective method for auditory habilitation in patients with auditory neuropathy, by restoring neural synchrony and allowing the use of auditory information, favoring the development of communication	5

AUTHOR/YEAR	OBJECTIVE	POPULATION	AGE AT EVALUATION	SPEECH PERCEPTION PROCEDURE	CLINICAL OUTCOME PROCEDURE	MAIN FINDINGS	LEVEL OF EVIDENCE
Yamaguti, (2013)[36] Case-control study	To investigate speech perception in the presence of noise in children with ANSD who use CI .	14 children with ANSD who used CI and 12 control subjects (children with sensorineural hearing loss, users of CI). Severe/profound hearing loss.	Experimental group: 111.5 months. Control group: 127.4 months.	*Hearing in Noise Test (Portuguese version)	Not provided	The performance in the ability of speech perception in noise was similar between the group of children with ANSD and the group of children with sensorineural hearing loss. The cochlear implant proved to be a viable treatment option for this population, provided that the indication criteria are followed.	4
Liu et al (2014)[32] Cohort study	To evaluate the auditory and language skills after CI in children with ANSD, with no cochlear nerve deficiency and to determine the role of age in implantation in the clinical data found.	10 children with ANSD, users of CI, divided by the age at which the implantation was performed (before or after 24 months). Profound hearing loss.	Not provided	*Categories of Auditory Performance (CAP) *Meaningful Auditory Integration Scale (MAIS)/Infant-Toddler Meaningful Auditory Integration Scale (IT-MAIS) *Monosyllabic Lexical Neighborhood Test (LNT Chinese version) *Multisyllabic Lexical Neighborhood Test (MLNT Chinese version)	*Speech Intelligibility Rating (SIR) *Meaningful Use of Speech Scale (MUSS)	The CI is a treatment option for children with ANSD, and children implanted before 24 months presented superior performance for auditory and speech skills than children implanted after 24 months.	4
Praveena et al. (2014)[27] Case report	To verify the language development of children with ANSD who use hearing aids.	3 children using bilateral hearing aids. Moderate/severe hearing loss.	2 years and 10 months to 5 years and 1 month.	Not provided	* Language Development Assessment (LDA)	Advances were observed in the development of the receptive age in relation to expressive age. This result may be characteristic of children with ANSD who use hearing aids.	5
Attias et al. (2016)[33] Case-control study	To compare the auditory result of CI between children with ANSD and children with sensorineural hearing loss, and to investigate the impact of different pathophysiology underlying ANSD and sensorineural hearing loss on objective and subjective electrical parameters of CI.	16 children with ANSD using uni or bilateral CI and 16 children with sensorineural hearing loss using unilateral or bilateral HF. Severe/profound hearing loss.	5 to 12 years.	*The Infant-Toddler Meaningful Auditory Integration Scale *Monosyllabic speech test: *Spondee word Test *Everyday sentences test	Not provided	Children with ANSD presented auditory performance similar to children with sensorineural hearing loss after CI. In addition, children with ANSD require a lower electrical current of CI stimulation than children with sensorineural hearing loss, which may be due to biological differences in cochlear conditions between these two groups.	4
Fernandes et al. (2016)[12] Case series	To analyze the speech perception of children with ANSD who use bilateral hearing aids.	4 children using bilateral hearing aids. Mild to moderate hearing loss.	9 years to 12 years and 2 months.	*List of recognition of monosyllabic words and disyllables *List of recognition of meaningless syllables: *List of sentence recognition Meaningful Auditory Integration for Young Children (IT MAIS) scale *Hearing category	* Meaningful Use of Speech Scale (MUSS) *Language categories	The auditory performance of the participants who used hearing aids was satisfactory, allowing the maximum development of auditory skills	5
Daneshi et al. (2018)[25] Cohort study	To evaluate the auditory performance and speech production of implanted children with ANSD.	136 children implanted unilaterally. Severe/profound hearing loss.	Not provided	*Categories of Auditory Performance (CAP)	*Speech Intelligibility Rating (SIR)	Children with ANSD benefit from CI. The improvement in auditory performance and speech production ability depends on the age at which the CI surgery was performed and the duration of the postoperative follow-up period.	4
Alzhrani et al. (2019)[34] Case-control study	To evaluate the auditory and speech performance of implanted children with ANSD and to compare the results obtained with implanted children with sensorineural hearing loss.	58 users of unilateral or bilateral CI: 18 com ANSD 40 controls. Severe/profound hearing loss.	Not provided	*Categories of auditory Performance (CAP)	*Speech Intelligibility Rating (SIR)	Children with ANSD, implanted early, benefit from CI, achieving auditory and speech performance similar to that of children implanted without ANSD.	4

Captions: CI: cochlear implant; ANSD: auditory neuropathy spectrum disorder; HA: hearing aid

Regarding the population of the included studies, the chronological age ranged from two years and two months to 12 years and two months, with a sample size between one and 136 participants. Regarding the degree of hearing loss, two studies included participants with mild hearing loss, two studies addressed the moderate degree, seven studies evaluated children with severe hearing loss, 12 encompassed the profound degree, one article mentioned the nomenclature "degree of severe hearing loss" and one study did not mention the degree of hearing loss.

When analyzing the criteria related to the population included in this study, we selected the studies with children with no structural alterations of the ear or vestibulocochlear nerve (cranial nerve VIII) and with no other associated impairments, such as syndromes.

It is known that the diagnosis of the Auditory Neuropathy Spectrum Disorder may be accompanied by other comorbidities, due to risk factors, genetic factors and peri- or postnatal complications, which makes the population extremely heterogeneous[8,9,20].

In addition, it is worth noting that the term "spectrum" brought an expansion to the concept of the lesion site, which contemplates other sites of alteration, in addition to the auditory nerve[21]. Although the pathophysiological mechanism of auditory neuropathy is not yet fully understood, the literature reports that the lack of synchronization of neural discharges along the auditory pathway causes changes in the accuracy of the coding or transmission of auditory information, which may explain this condition[7,9,10].

Thus, in view of the different manifestations that individuals with ANSD may present, the referred criteria for the population were established aiming at greater homogeneity of the participants of the included studies, which may be related to the number of studies found.

Among the included studies, the number of the sample varied widely, observing case report studies[13,22-24] that evaluated the speech perception and language of children who received unilateral cochlear implant and a multicenter study[25], which proposed to evaluate the auditory performance and speech production of 136 children who received the cochlear implant between 2003 and 2015, performing a follow-up in the period of two years post-implantation. Due to the longitudinal character, the authors obtained the complete evaluations of 79 participants.

Regarding the degree of hearing loss, which ranged from mild to profound, only two studies involved participants with ANSD and mild hearing loss[12,26] and two studied children with ANSD and moderate hearing loss[26,27]. This result emphasizes the importance of neonatal hearing screening programs, which involve both evoked otoacoustic emissions and brainstem auditory evoked potentials, because the combination of both tests will avoid errors, allowing to verify babies with possible findings compatible with auditory neuropathy spectrum disorder[4,28,29].

Most of the children evaluated had severe or profound hearing loss and used cochlear implants[13,22-26,30-36]. Studies show that the cochlear implant can bring greater neural synchrony[13,24,31], becoming an alternative treatment when the hearing aid is not effective for the development of auditory skills and language[23].

However, the CI indication process should be thorough, considering that a common clinical conduct is not possible for this group of children, due to the fact that the ANSD presents different degrees of impairment along the auditory pathway, with fluctuation in auditory performance, making the decision-making process about the best rehabilitation option for this population challenging[26,31,33,34].

Two studies showed that children who use hearing aids may present benefits in speech perception skills, with regard to auditory recognition and comprehension, when evaluating children with degrees of hearing loss that ranged from mild to severe[12,26]. However, another study, which evaluated the language development of children using hearing aids with moderate to severe hearing loss, found superior performance for participants with lower degree of hearing loss, stating that amplification can help in hearing, but is not enough to modify the neural coding pattern that is altered in the different degrees of hearing loss, especially in the severe degree[27].

To this extent, it is emphasized that a careful preoperative evaluation is necessary, which takes into account different aspects, such as the individual benefits of hearing aids, the parents' perception of the child's development, the parents' expectations regarding the results of the CI and the active participation of the family in the rehabilitation process[13,22,32].

It is worth mentioning that, after the conduct is established, the success of the devices (HA and CI) for the development of auditory and language skills involves factors such as the motivation to use the device, family support, speech therapy, among others[31,37].

Regarding the clinical outcomes of speech perception, it was possible to observe that results were obtained through procedures that involved inventories answered by the guardians, scales scored by the evaluator, tests with the child him/herself or the procedures agreed upon.

Among the inventories answered by the parents/guardians, the Meaningful Auditory Integration Scale (MAIS) was applied in two studies[22,32] and the Infant-Toddler Meaningful Auditory Integration Scale (IT-MAIS) in three studies[12,32,33]. The use of these procedures is relevant, considering that the report of the parents/guardians is an important tool to evaluate the effectiveness of the devices in significant contexts of daily life, especially for young children[22].

The scores on these scales ranged from zero to 44% before CI surgery [22,33], while after surgery scores of up to 100% were achieved[12,22,32,33]. In one study[22], the results were obtained after three and seven months of use of the implant, achieving development of hearing skills, with a maximum score of 81% at the end of this period. Another study found that children implanted before 24 months of life had higher scores for the MAIS and IT-MAIS scales than children implanted after this period[32].

The early age for cochlear implant surgery is reported in the literature as a factor capable of bringing benefits to the speech perception of children with ANSD [25,32,34]. Other authors, who evaluated the auditory skills of this group, through the Categories of Auditory Performance (CAP) scale, found superior results for participants implanted early[25,34].

Regarding the tests applied with the child him/herself, the results showed that different skills were evaluated through a variety of evaluation procedures, as shown in Chart 2.

One of the main aspects considered was to understand whether there were differences in performance in speech perception between children with ANSD who used hearing aids or CI and children with sensorineural hearing loss[13,26,33,34,36]. It was noted that four studies found similar results between the groups with regard to the abilities of detection of speech sounds[13], attention to environmental and speech sounds of everyday life[33,34], auditory discrimination[13,33,34], recognition in closed and open set[13,33,34,36], sequential memory[13], background figure[13] and auditory comprehension[13,34].

However, one study found different results, in which children with ANSD who used CI or HA had lower performance in the ability to recognize phonemes, when compared to children with sensorineural hearing loss[26].

These different findings in the literature show that knowing the site of the alteration along the auditory pathway is fundamental in assisting in the choice of the technological resource for auditory rehabilitation and in understanding the prognosis of the individual with ANSD. When there is a sensory impairment, a favorable prognosis is expected with the use of hearing aids, but if there is a dysfunction that also involves the neural component, the auditory responses may be unpredictable, regardless of the device used (HA or CI) and, specifically with the CI, the results may depend on the ability of the auditory nervous system to deal with electrical stimulation[12].

It is worth noting that among the tests applied with the child, only two studies evaluated the perception of speech in the presence of competitive noise. In both studies, children with CI and ANSD were compared with children with CI and sensorineural hearing loss, in relation to the recognition of sentences in noise[33,36] and spondaic words[33], in which the performance of these groups was similar.

Considering that the monitoring of the development of the auditory skills of children who use devices is an important factor to verify the effectiveness of the intervention with the HA and the CI and that this population can reach the plateau in the tests applied in silence, the evaluation with competitive noise becomes relevant, because it inserts a listening situation closer to everyday life[12,25,38].

Regarding the clinical outcomes of spoken language, the receptive and expressive aspects were evaluated, using different procedures, as shown in Chart 2.

In relation to the receptive aspect, only one study investigated this ability, through the Language Development Assessment (ADL) procedure. Three children using bilateral hearing aids, with moderate to severe hearing loss, who underwent speech therapy with emphasis on hearing and spoken language, were evaluated, verifying differences in receptive age between the pre- and post-therapy moments, which ranged from six months to three years[27]. Regarding the expressive aspect, the same study obtained results by ADL before and after the intervention, with differences in expressive age ranging from one year to one year and seven months[27].

Although the number of participants was low, this study demonstrates the importance of speech therapy, with emphasis on the development of hearing and spoken language, as children with hearing loss, using devices, need an intervention that helps in the use of hearing information obtained by hearing aids or CI for the establishment of effective oral communication[24,27].

It is worth mentioning that the main expressive aspect evaluated in the studies was speech intelligibility, in which four studies[22,25,32,34] used the Speech Intelligibility Rating (SIR) scale. One study[35] applied a speech intelligibility test based on percentage and one study[30] used the ABFW phonology test.

The literature reports that intelligibility is an aspect that can bring representative results about the child's competence for spoken language, quickly, reflecting their performance in a more natural communication situation[39,40].

All studies that evaluated speech intelligibility were performed with implanted children and verified that the intervention with CI led to an increase in the scores obtained with the scales used[22,25,30,32,34,35]. Considering that the development of spoken language in children who use hearing aids may also be influenced by the quality of the acoustic signal provided by the device, impacting on the expressive aspect, the evaluation of speech intelligibility in this group is also relevant[27].

Two studies compared speech intelligibility between children with ANSD who used CI and implanted children with sensorineural hearing loss, finding no differences in performance between the groups[30,34]. These findings demonstrate that the access to speech sounds provided by the adequate adaptation of the device can bring benefits to the spoken language of children with hearing loss.

It was also pointed out in the studies that children with higher scores for speech intelligibility had shorter sensory deprivation time and had undergone CI surgery at an early age[25,32,34]. In addition, there was an evolution in the performance of speech intelligibility, as a function of the duration of the post-adaptation follow-up period with the CI[25]. These results show the importance of early intervention and systematic monitoring of the development of this group.

Regarding the level of evidence, eight studies were classified in "level 4", which consists of descriptive studies, five case-control studies[26,30,33,34,36], two cohort studies[25,32] and one cross-sectional study[31], while seven studies were classified in "level 5", which represents the case studies[12,13,22-24,27,35]. It is possible to observe that the 15 included studies are between levels 4 and 5 of the classification proposed by Cox (2004) [18], which consist of lower levels related to the quality of the study.

Chart 3 shows the risk of bias of the studies, analyzed by the critical appraisal tools of the *Joanna Briggs Institute*[19].

Chart 3. Risk of bias of included studies

Checklist: Case Reports					
Questions	**Trautwein et al. (2000)[13]**	**Lin et al. (2005)[23]**	**Fei et al. (2011)[22]**	**Kim et al. (2013)[24]**	**Praveena et al. (2014)[27]**
1. Were patient's demographic characteristics clearly described?	Y	Y	Y	Y	N
2. Was the patient's history clearly described and presented as a timeline?	Y	Y	Y	N	N
3. Was the current clinical condition of the patient on presentation clearly described?	Y	Y	Y	Y	Y
4. Were diagnostic tests or assessment methods and the results clearly described?	Y	Y	Y	N	Y
5. Was the intervention(s) or treatment procedure(s) clearly described?	Y	Y	Y	N	Y
6. Was the post-intervention clinical condition clearly described?	Y	Y	Y	Y	Y
7. Were adverse events (harms) or unanticipated events identified and described?	NA	NA	NA	NA	NA
8. Does the case report provide takeaway lessons?	Y	Y	Y	Y	Y
Risk of bias (% "YES")	100% (Low)	100% (Low)	100% (Low)	57,40% (Moderate)	71,43% (Low)
Checklist: Case-Control Studies					
Questions	**Melo et al. (2008)[30]**	**Rance et al. (2008)[26]**	**Yamaguti (2013)[36]**	**Attias et al. (2016)[33]**	**Alzhrani et al. (2019)[34]**
1. Were the groups comparable other than the presence of disease in cases or the absence of disease in controls?	Y	Y	Y	Y	Y
2. Were cases and controls matched appropriately?	Y	Y	Y	Y	N
3. Were the same criteria used for identification of cases and controls?	Y	Y	Y	Y	Y

4. Was exposure measured in a standard, valid and reliable way?	NA	NA	NA	NA	NA
5. Was exposure measured in the same way for cases and controls?	Y	Y	Y	Y	Y
6. Were confounding factors identified?	N	Y	Y	Y	N
7. Were strategies to deal with confounding factors stated?	N	Y	Y	Y	N
8. Were outcomes assessed in a standard, valid and reliable way for cases and controls?	Y	Y	Y	Y	Y
9. Was the exposure period of interest long enough to be meaningful?	U	NA	NA	NA	U
10. Was appropriate statistical analysis used?	Y	Y	Y	Y	Y
Risk of bias (% "YES")	66,67% (Moderate)	100% (Low)	100% (Low)	100% (Low)	55,56% (Moderate)

Checklist: Case Series

Questions	**Fukushima et al. (2009)[35]**	**Fernandes et al. (2016)[12]**
1. Were there clear criteria for inclusion in the case series?	Y	Y
2. Was the condition measured in a standard, reliable way for all participants included in the case series?	Y	Y
3. Were valid methods used for identification of the condition for all participants included in the case series?	Y	Y
4. Did the case series have consecutive inclusion of participants?	Y	N
5. Did the case series have complete inclusion of participants?	U	U
6. Was there clear reporting of the demographics of the participants in the study?	Y	Y
7. Was there clear reporting of clinical information of the participants?	Y	Y
8. Were the outcomes or follow up results of cases clearly reported?	N	Y
9. Was there clear reporting of the presenting site(s)/clinic(s) demographic information?	NA	NA
10. Was statistical analysis appropriate?	Y	Y
Risk of bias (% "YES")	77,78% (Low)	77,78% (Low)

Checklist: Cohort Studies

Questions	**Liu et al. (2014)[32]**	**Daneshi et al. (2018)[25]**
1. Were the two groups similar and recruited from the same population?	Y	Y
2. Were the exposures measured similarly to assign people to both exposed and unexposed groups?	Y	Y
3. Was the exposure measured in a valid and reliable way?	Y	Y
4. Were confounding factors identified?	Y	Y
5. Were strategies to deal with confounding factors stated?	Y	Y
6. Were the groups/participants free of the outcome at the start of the study (or at the moment of exposure)?	U	N
7. Were the outcomes measured in a valid and reliable way?	Y	Y
8. Was the follow up time reported and sufficient to be long enough for outcomes to occur?	Y	Y
9. Was follow up complete, and if not, were the reasons to loss to follow up described and explored?	U	N
10. Were strategies to address incomplete follow up utilized?	U	N
11. Was appropriate statistical analysis used?	Y	Y
Risk of bias (% "YES")	72,73% (Low)	72,73% (Low)

Checklist: Cross Sectional Studies

Questions	**Carvalho et al. (2011)[31]**
1. Were the criteria for inclusion in the sample clearly defined?	Y
2. Were the study subjects and the setting described in detail?	Y
3. Was the exposure measured in a valid and reliable way?	Y
4. Were objective, standard criteria used for measurement of the condition?	Y
5. Were confounding factors identified?	NA
6. Were strategies to deal with confounding factors stated?	NA
7. Were the outcomes measured in a valid and reliable way?	Y
Risk of bias (% "YES")	100% (Low)

Captions: Y – Yes; N – No; U – Unclear; NA – Not applicable
Source: Joanna Briggs Institute (JBI)[19]

The risk of bias in three studies was classified as moderate, with blanks in the description of the patients' history[24], pairings of cases and controls[34], description of the method used to evaluate clinical outcomes[24], description of intervention procedures[24] and lack of identification of confounding factors, as well as strategies for resolving these factors[30,34].

There were also 12 studies identified with low risk of bias, although five studies presented methodological aspects that were not contemplated, such as: whether there was the inclusion of all participants of interest from the place where the research was developed[12,35], clear description of the demographic characteristics of the participants[27], results of speech and/or language perception before the intervention with the devices[25,32], clearly reported clinical outcomes[35], description of the reasons for the loss of participants throughout the period in which the research was developed, as well as strategies for adjusting the analysis of the outcomes, when patients did not participate in the entire study period[25,32].

Although variability was observed in relation to sample size, types of studies, evaluation procedures used and methodological aspects, the results of this literature review demonstrated that CI and HA can be effective for the development of speech perception and spoken language in children with auditory neuropathy spectrum disorder, once complex levels of these skills were achieved, such as listening comprehension and speech intelligibility. Due to the heterogeneity of the studies, it was not possible to perform a meta-analysis.

Considering the specificities of ANSD, the continuity of research that seeks to evaluate the speech perception and spoken language of this population, with standardized methods and longitudinal character, may add information about the effectiveness of hearing aids and CI and contribute to the intervention in this pathology.

CONCLUSION

Considering the heterogeneity of the auditory neuropathy spectrum, over 25 years of research, 15 studies were verified suggesting that CI and HA may be effective for the development of speech perception and spoken language of children with auditory neuropathy spectrum disorder with no other associated impairments. The need for studies with high methodological rigor that demonstrate the effectiveness of the intervention with the devices (HA and/or CI) in the performance of speech perception and language of children with ANSD, is emphasized.

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Author contributions:

FRS: conception and design of the study, data collection, analysis, interpretation of data and writing of the article;

JSZF: conception and design of the study, data collection, analysis, interpretation of data and critical review of the article for relevant intellectual content;

EMCD-P: conception and design of the study, data collection, analysis, data interpretation, critical review of the article for relevant intellectual content and final approval of the version to be published.