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Auditory hyper-responsiveness in autism spectrum disorder, terminologies and physiological mechanisms involved: systematic review

Hiper-responsividade auditiva no transtorno do espectro autista, terminologias e mecanismos fisiológicos envolvidos: revisão sistemática

Keywords

Hyperacusis
Hypersensitivity
Hearing
Autistic Spectrum Disorder
Child

Palavras-chave

Hiperacusia
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Audição
Transtorno do Espectro Autista
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ABSTRACT

Purpose: this paper aims to identify the most used terminologies to designate the disproportional behavior to sounds in the autism spectrum disorder (ASD) and its relationship with the respective tools for its investigation, as well as its occurrence and outcomes. **Research strategies:** the databases used were PubMed, PsycINFO, Web of Science, Scielo and Lilacs. The keywords used were “autism”, “hyperacusis” and “auditory perception”, with the following combinations: “autism AND hyperacusis” and “autism AND auditory perception”. **Selection criteria:** individuals diagnosed with ASD of any age group; available abstract; papers in English, Spanish and Brazilian Portuguese; case series, prevalence and incidence studies, cohort and clinical trials. **Data analysis:** we analyzed studies with individuals diagnosed with ASD of any age group; reference in the title and/or summary of the occurrence of disproportional behavior to sounds, accepting the terms hyper-responsiveness, hypersensitivity and hyperacusis; summary available; papers in English, Spanish and Brazilian Portuguese; series of cases, prevalence and incidence studies, cohort and clinical trials. **Results:** Of the 692 studies resulting from the consultation, 13 studies could achieve the established requirements. **Conclusion:** The term auditory hypersensitivity was the most commonly used to designate disproportional behavior to sounds, followed by hyperacusis. There was no relationship between the terms and the respective research tool, and the questionnaires were the most used to designate the referred behavior, whose reported frequency was from 42.1% to 69.0%. The auditory behavior tests when performed showed the involvement of the auditory, afferent and efferent neural pathways.

RESUMO

Objetivo: identificar as terminologias mais utilizadas para designar o comportamento desproporcional a determinados sons (CDS) no TEA e sua relação com as respectivas ferramentas para sua investigação, assim como sua ocorrência e desfechos. **Estratégia de pesquisa:** Foram utilizadas as bases de dados: PubMed, PsycINFO, Web of Science, Scielo e Lilacs. As palavras-chave utilizadas foram “autism”, “hyperacusis” e “auditory perception”, com as seguintes combinações: “autism AND hyperacusis” e “autism AND auditory perception”. **Crerios de seleção:** Foram incluídos os trabalhos com diagnóstico de TEA, de qualquer faixa etária; resumo disponível; Artigos em inglês, espanhol e português brasileiro; série de casos, estudos de prevalência e incidência, coorte e ensaios clínicos. **Análise dos dados:** Foram analisados estudos com sujeitos com diagnóstico de TEA de qualquer faixa etária; referência no título e/ou resumo da ocorrência do CDS, aceitando os termos hiper-responsividade, hipersensibilidade e hiperacusia; resumo disponível; artigos em inglês, espanhol e português brasileiro; série de casos, estudos de prevalência e incidência, coorte e ensaios clínicos. **Resultados:** Dos 692 estudos resultantes da consulta, foram identificados 13 que atendiam aos requisitos estabelecidos. **Conclusão:** O termo hipersensibilidade auditiva foi o mais empregado para designar o CDS, seguido da hiperacusia. Não houve relação entre os termos e a respectiva ferramenta de investigação, sendo os questionários os mais utilizados para designar o referido comportamento, cuja frequência relatada foi de 42,1% a 69,0%. Os testes auditivos, quando realizados, mostraram o envolvimento das vias neurais auditivas, aferente e eferente.

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INTRODUCTION

The literature characterizes Autism Spectrum Disorder (ASD) as a continuum of changes in social communication, with heterogeneous manifestations that affect development with different levels of severity, causes early losses in socialization and communication, as well as presents restricted and stereotyped behaviors and interests^(1,2). Disorders such as sensory hypo and hyper-responsiveness have always been frequent in the population with ASD, but only after launching the fifth edition of the *Diagnostic and Statistics Manual of mental disorders* (DSM-5), they were reported as common manifestations⁽³⁻⁶⁾.

Studies investigating auditory sensory disturbances in ASD have two strands. The first strand aimed at a better understanding of the mechanisms of temporal listening and speech perception in noise^(7,8); the second one aimed at analyzing the response during presentation of a given acoustic stimulus, characterized by a disproportionate behavior according to sound characteristics (DBS)⁽⁹⁾.

In the first strand, the studies showed that individuals with ASD presented longer responses to the perception of temporal variations of acoustic stimuli, as well as inferior results in conditions of hearing in the face of noise⁽⁸⁾. These results characterize the presence of auditory processing disorder⁽¹⁰⁾, a clinical entity that is also commonly described in other conditions that affect development, such as dyslexia, specific language disorder, dysfluency and attention deficit/hyperactivity disorder (ADHD), among others^(11,12). It is important to emphasize that the behavioral tests of auditory processing evaluation require the individual to understand the instructions of the test and its cooperation, motor and/or verbal response, depending on the test, which requires the precise “timing” of response⁽¹⁰⁾, and these abilities are impaired in ASD.

The second strand seeks to characterize, understand and investigate the occurrence of DBS, which is named with different terms, being the most common auditory hypersensitivity (AHS)^(8,13) and hyperacusis (HPA)⁽¹⁴⁻¹⁷⁾. The existence of divergence in the nomenclature to designate DBS in ASD has already been observed and previously weighted^(9,18), as well as the repercussions that such designations generate in the information conflict for the way they were studied⁽⁹⁾. In fact, Tyler et al.⁽¹⁹⁾ have already called attention to the fact that these terminologies in the field of hearing science are not synonymous.

The AHS theoretically refers to the higher sensitivity thresholds of those considered “normal”, which is impossible to measure, that is, researched⁽¹⁹⁾. The auditory thresholds considered as adequate, without hearing loss, range from -10 to + 15, 20 or 25 dB HL, depending on the age and the criterion adopted⁽²⁰⁻²²⁾, based on the calibration recommendations of the measuring instrument^(23,24). Studies that investigated comparatively the auditory sensitiveness (tonal thresholds) in individuals with and without ASD, all without hearing loss, revealed that the differences found between the two populations is insignificant^(13,15).

The HPA, in turn, refers to the complaint of discomfort and/or irritability in the face of a certain sound or sounds when other people, who share the same environment and sound exposure, do not report the same⁽⁹⁾. The use of specific, standardized and validated questionnaires/inventories⁽²⁵⁻²⁷⁾ may help to identify

the risk population for HPA. However, currently, the gold pattern for its identification is the psychoacoustic threshold of discomfort, which, in this condition, occurs at an intensity lower than usual⁽¹⁹⁾. This same test has already been recommended as a useful tool for the population with ASD⁽¹⁸⁾, assuming that DBS in ASD can be configured in the HPA.

Leekam et al.⁽²⁸⁾ reported that sensory disturbances affect up to 90.0% of individuals with ASD. According to the same authors, auditory sensory disorder has a lower frequency than visual and proprioceptive, but it is interesting to note that while the frequency of these last two modalities increased according to the severity of ASD, auditory frequency remained stable⁽²⁸⁾. The occurrence of DBS in ASD is heterogeneous, between 16.2% and 69.0%, whether the different terminologies HSA, HPA and auditory hyper-responsiveness (AHR) are computed^(8,13,14,29). It is important to note that the literature describes the occurrence of HPA in 1.9 to 9.2% of adults and elderly⁽³⁰⁾, and in 3.4% of children’s population, all without ASD⁽²⁶⁾.

Pathophysiological mechanisms of HPA, regardless of the affected population, are not yet fully known, nor of the DBS in ASD. There are some hypotheses for HPA, three of which are the most frequent. The first relates to the homeostatic plasticity of the Central Nervous System (CNS), which is responsible for the accuracy of neural coding, through the regulation and adaptation of different sound stimuli to which individuals are exposed^(31,32). The second refers to tonotopic reorganization in primary areas after damage to the receptors, and in this process, there would be an increase in the representation of certain frequencies, resulting in auditory discomfort^(33,34). The third hypothesis is related to a failure in the modulation of the efferent fibers of the olivocochlear system that protrude to the cochlea, specifically to external ciliated cells, which are responsible for regulating sound amplification⁽³⁵⁾.

The lack of homogenization in the terminology to designate the DBS in ASD has been previously pointed out⁽⁹⁾. The use of different terminologies to denominate a frequent manifestation in this condition does not allow the identification of the tool, method, context and behavior observed. It does not allow the reader to identify whether these terminologies refer to the symptom and/or clinical sign, which influences the little knowledge about the characteristic behavior of ASD.

OBJECTIVES

This review aims to elucidate how the use of the main terminologies to characterize the DBS in ASD relate to the respective research instruments. It also aims to describe the occurrence of phenomenon and outcomes of the respective mechanisms involved in this response.

Research strategy

The articles that have been studied were selected from searches in the following databases: PubMed, PsycINFO, Web of Science, Scielo and Lilacs. To carry out the search, the following descriptors were used: “autism”, “hyperacusis” and “auditory perception”, with the following combinations: “autism AND

hyperacusis' and "autism AND auditory perception", with no lower limit of date and published papers until September 2017. The descriptors used in the review process were selected by consulting the descriptors in health science.

Selection criteria

The references resulting from the search were analyzed according to the established inclusion criteria, namely: individuals diagnosed with ASD of any age group; reference in the title and/or abstract of the DBS, accepting the terms AHR, AHS and HPA; available abstract; papers in English, Spanish and Brazilian Portuguese; case series, prevalence and incidence studies, cohort and clinical trials. The selected papers were read in full and the next step was to exclude those whose method did not report the instrument used to identify the occurrence of DBS, or those that studied sensory disorders, but did not specify the modalities. The flowchart (Figure 1) shows the steps of the process mentioned.

Data analysis

Two of the authors conducted the study independently and subsequently they confronted the collected data. When there was incompatibility in the data collected, a third professional was

invited to have a common sense. The research questions were: form (e.g. questioning to relatives, questionnaires/inventories/test) to obtain information of DBS; the term used; realization or not of auditory test, if yes, which; and results.

Quality of the study assessment

In order to characterize the evidence level of the work, the Newcastle-Ottawa Scale was used⁽³⁶⁾, and this scale analyzes three main categories: sample selection, comparability of the two groups and the way the outcome is measured. The total score can range from 1 to 10, where a score greater than or equal to six indicates quality, according to the scale proposal for this study.

RESULTS

From the descriptors, 692 studies were obtained that, after analysis by the inclusion and exclusion criteria, 13 studies were selected, corresponding to 1.8% (13/692) of the initial sample. Table 1 shows the selected works with the respective terms used to designate the DBS, the research instruments, as well as the main results. In this section, the 13 papers will be referenced by the chronological order of publication and not by the authors.

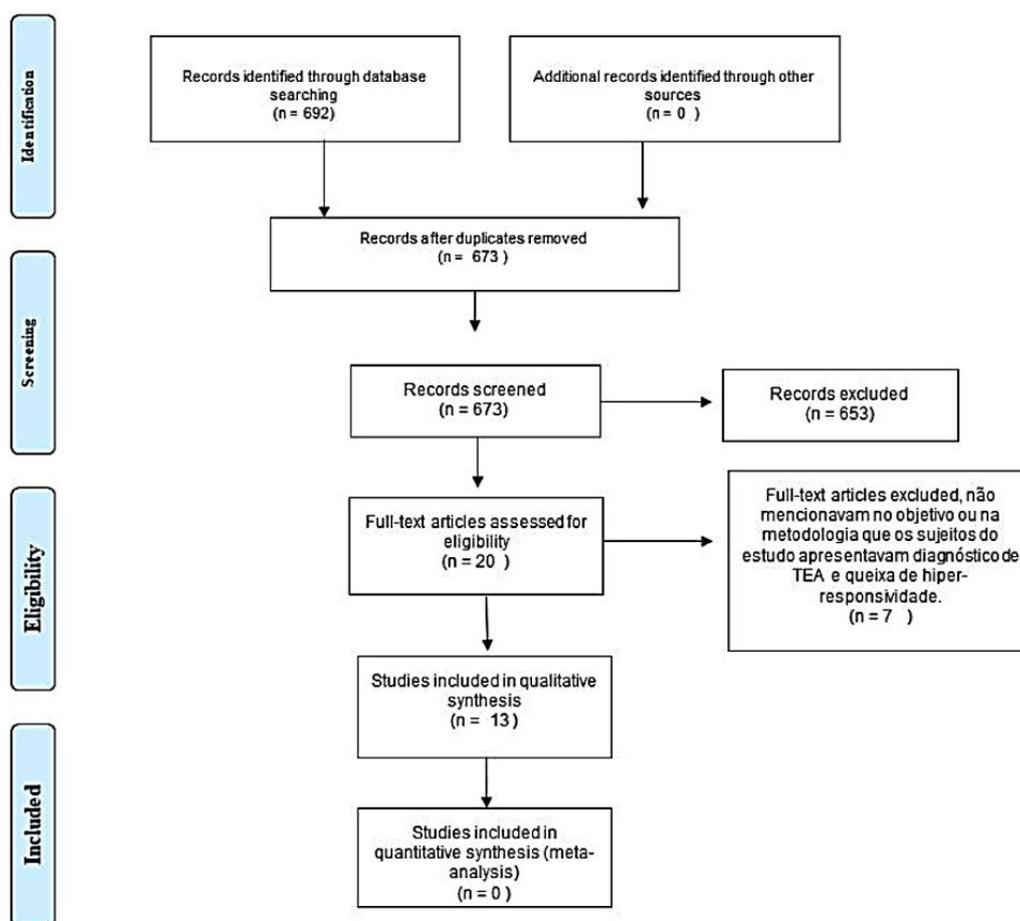


Figure 1. Systematic Review Steps

Table 1. Terminology used, casuistic, symptom assessment instrument, examinations and tests performed, and main results of the selected articles

Author/ Terminology	Casuistic (n) and age group	Symptom Research Instrument	Exams/tests carried out	Main results
1 Khalifa et al. (2004) ⁽²⁵⁾ HPA	GASD=11 CG=11 9-17 years old	Questioning relatives	Auditory – Psychoacoustics	<ul style="list-style-type: none"> - thresholds for pure tones with no difference between groups - GASD show measures of discomfort thresholds and dynamic field of hearing, smaller than CG - HPA in 63.0% in GASD vs. 27.0% CG - The dynamic field of hearing was lower at 4 and 8 kHz, regardless of the group
2 Danesh and Kaf (2012) ⁽¹⁶⁾ HSA	GASD = 18 CG = 12 6-14 years old X = 8	Not used	<ol style="list-style-type: none"> 1. Evoked Otoacoustic Emissions (EOAE) Sensory response – inner ear 2. Inhibitory effect of emissions otoacoustic 	<ul style="list-style-type: none"> - GASD show lower amplitudes of response than CG - GASD showed lower values of the suppression effect than CG and they were higher for the left ear than to the right - The lower the age the greater the suppression value, without differences between the ages in each of the groups
3 Matsuzaki et al. (2012) ⁽³⁷⁾ HSA	GASD = 18 X = 9 years old GC = 12 X = 10 years old	<i>Infant/Toddler Sensory Profile (Dunn, 2002)</i> ⁽³⁸⁾	Auditory – magnetoencephalography	<ul style="list-style-type: none"> - The score of the questionnaire defined the presence of AHS and subdivided the GASD in with and without AHS - Amplitude measurements of the M50 and M100 components were not different between the groups, regardless of the questionnaire score - Latency of the M50 had a difference only between GASD and AHS when compared with CG - Latency of the M100, there was a difference between the three GASD groups with AHS > GASD without AHS > CG
4 Thabet and Zaghloul (2013) ⁽³⁹⁾ HSA	GASD =14 CG =15 2-8 years X= 3 years ± 1	Informal reporting of relatives	<ol style="list-style-type: none"> 1. ABR Neural response – subcortical 2. Image – Internal ear tomography 	<ul style="list-style-type: none"> - Electrophysiological threshold with no difference between groups - Absolute latencies of components I and III and interpeak latencies I – III, with no differences between groups - Absolute latency of early GASD component V and intervals III – V and I – V with lower values than CG - 29% of GASD showed images compatible with dehiscence of the semicircular canals
5 Bhatara et al. (2013) ⁽⁷⁾ HSA	GASD =17 CG =17 X=10-14 years old	<ol style="list-style-type: none"> 1. <i>Sensory Profile (Dunn, 1999)</i>⁽⁴⁰⁾ 2. <i>Adolescent/adult sensory profile (Dunn, 2002)</i>⁽³⁸⁾ 	Auditory – psychoacoustics	<ul style="list-style-type: none"> - Adolescents with ASD presented higher scores in sensory profile than neurotypical (NT) group - The group with ASD requires a greater ratio signal-noise than control group to understand the words in the speech with noise test - Group with ASD: impaired frequency discrimination
6 Bhatara et al. (2013) ⁽⁴¹⁾ HSA	GASD =33 CG=35 10-19 years old	<ol style="list-style-type: none"> 1. <i>Salk and McGill Musical Inventory</i> 	Not used	<ul style="list-style-type: none"> - Adolescents with ASD (68%) showed auditory hypersensitivity in childhood more frequently than in control group (17%). - Even with hypersensitivity, the interest in music in individuals with ASD was the same as in control group - Verbal performance and IQ do not predict auditory hypersensitivity, as well as chronological age also showed a negative correlation to hypersensitivity.

Caption: AHS= Auditory hypersensitivity; IQ=Intelligence quotient; AHR= Auditory hyper-responsiveness; HPA=hyperacusis; GASD= Group with Autistic Spectrum Disorder; CG= Control group; VEMP= Vestibular evoked myogenic potential; LLAEP=Long latency auditory evoked potential; ASD=Autism spectrum disorder; ABR=Auditory brain response; EOAE=Evoked otoacoustic emissions

Table 1. Continued...

Author/ Terminology	Casuistic (n) and age group	Symptom Research Instrument	Exams/tests carried out	Main results
7 Thabet (2014) ⁽⁴²⁾ HSA	GASD =14 CG =15 2-4 years old	Not applicable	Not used	- Study Group: VEMP amplitude increase in five subjects - VEMP showed diagnostic capability in the differentiation of hypersensitivity caused by semicircular canal dehiscence syndrome or by atypical cortical development.
8 Matsuzaki et al. (2014) ⁽⁴³⁾ HSA	GASD =21 CG =15 X= 9 years old ± 1	1. <i>Infant/Toddler Sensory Profile</i> (Dunn, 2002) ⁽³⁸⁾	Magnetoencephalography	- Group with ASD and auditory hypersensitivity showed more prolonged responses with latencies (M50/M100) than other groups; - Latency was shown to be significantly related to the severity of auditory hypersensitivity
9 Donkers et al. (2015) ⁽⁴⁴⁾ AHR	GASD =29 CG =30 4-12 years old	1. <i>The Sensory the Tactile Defensiveness</i> 2. <i>Discrimination Test-Revised</i> 3. <i>Sensory Processing Assessment for Young</i> 4. <i>Sensory Experiences Questionnaire</i> 5. <i>Sensory Profile</i>	LLAEP Neural responses -subcortical and cortical	- It was observed for standard sounds, the relation between wave P1 and N2: P1 responses greater and N2 attenuated, auditory more severe hyper-responsiveness. - P1 attenuated and N2 attenuated, - lighter hyper-responsiveness.
10 Danesh et al. (2015) ⁽²⁹⁾ AHR	GASD =55 4-42 years old X=17 years old	1. <i>Hyperacusis Questionnaire</i> (Khalfa et al., 2002) ⁽⁴⁵⁾	Not used	- 69% of the subjects reported hyperacusis - 35% reported buzzing sound - Higher prevalence of hyperacusis and buzzing sound in the population with ASD than in general population.
11 Green et al. (2015) ⁽⁴⁶⁾ AHR	GASD =19 TG = 19 9-17 years old X = 14	1. <i>Short Sensory Profile</i> . 2. <i>Sensory Over-Responsivity Scales (SORS)</i>	Functional resonance	- There was no difference in activity activation between the groups for auditory stimulation, but for tactile and for concomitant stimulation, GASD showed a higher response than CG. - There was a positive correlation between SORS score and activation of cortical areas. - There was no difference between groups for habituation onset, but once the process began, it was slower in GASD and in GASD with SOR, it was slower for time when compared to CG GASD
12 Dunlop et al. (2016) ⁽⁸⁾ AHR	GASD =16 20-52 years old CG =31 19 years old	1. <i>Auditory Attention and Distress Questionnaire</i>	Auditory – psychoacoustics	- There was a statistically significant difference between the discomfort threshold of CG and GASD with without hypersensitivity, demonstrating that the latter has lower values of auditory discomfort thresholds
13 Wilson et al. (2017) ⁽¹⁷⁾ HPA	GASD =18 CG=14 10-16 years old	1. <i>Hyperacusis Questionnaire</i> (Khalfa et al., 2002) ⁽⁴⁵⁾	Auditory – electroacoustic	- Positive correlation between severity of hyperacusis and suppression of EOAE. - The suppression effect is higher in all frequencies of GASD with severe HPA when compared to GASD without severe HPA and CG.

Caption: AHS= Auditory hypersensitivity; IQ=Intelligence quotient; AHR= Auditory hyper-responsiveness; HPA=hyperacusis; GASD= Group with Autistic Spectrum Disorder; CG= Control group; VEMP=Vestibular evoked myogenic potential; LLAEP=Long latency auditory evoked potential; ASD=Autism spectrum disorder; ABR=Auditory brain response; EOAE=Evoked otoacoustic emissions

Instruments and terminologies

For the investigation and DBS characterization of were used standardized questionnaires in 69.2% (9/13) of the papers (studies 3, 5, 6, 8, 9, 10, 11, 12 and 13). The most frequent questionnaire was the Sensory Profile (study 5), when its three versions were counted, Short Sensory Profile (study 11), Infant/Toddler Sensory Profile (Studies 3 and 8), Adolescent/adult Sensory Profile (study 5). Another repeated questionnaire was the Hyperacusis Questionnaire (Studies 10 and 13). The other 30.8% (4/13) did not use structured questionnaires and in three of them, the DBS identification was done by means of family reports (studies 1, 4 and 7).

Of the nine studies using questionnaires, only 33.3% (3/9) (studies 6, 10 and 11) used them as an exclusive tool, while the remaining 66.7% (6/9) associated their use to some auditory function test.

Of the 13 studies, 76.9% (10/13) investigated hearing by psychoacoustic and/or objective measures, with different tests: discomfort threshold (studies 1 and 12); evoked otoacoustic emission (EOAE) with and without suppression effect (studies 2 and 13); magnetoencephalography (MEG) (studies 3 and 8); behavioral assessment of auditory processing (studies 5 and 12); short- and long-latency electrophysiological evaluation, and Vestibular Evoked Myogenic Potentials (VEMP) (studies 4, 9 and 7, respectively).

The AHS terminology was used in 61.5% (8/13), followed by HPA 23.1% (4/13) and, lastly, the term AHR in 15.4% (2/13) (Table 1).

Thus, it could not be observed a relationship between the use of terminology and the instruments designated to identify the magnitude of disproportional behavior to sound. For example, AHS was identified with structured questionnaires (studies 3, 5, 6, 8 and 12), because of the relative complaint (study 2) and by means of psychoacoustic and/or electrophysiological tests (studies 3, 7 and 8). The same occurred as a result of the use of HPA, which was studied with the psychoacoustic test of the discomfort threshold (study 1), by a specific questionnaire (study 10) and electroacoustic (study 13).

Occurrence of CDS

The occurrence of disproportional behavior for acoustic stimuli, independently of the terminology, ranged from 47.4 to 69.0% (studies 6, 10 and 11). The percentage of 47.4% appeared in study 11 and refers to the auditory and visual hyper-responsiveness at the same time. The frequency identified in studies 6 and 10 was similar, 68 and 69%, respectively.

Outcome

The HPA was specifically investigated in only 15.4% (2/13) of the papers (studies 1 and 12) and both reported significant differences in the discomfort threshold and/or dynamic field of hearing between individuals with and without ASD, and the values of the first group were smaller compared to the second. The HPA complaint was investigated by means of a specific questionnaire in two studies, 10 and 13.

In Table 1 are listed the results of the studies of auditory and vestibular tests and their correspondences regarding the type of test in relation to the type of response and mechanisms evaluated (sensory vs. neural) with their corresponding structures.

Newcastle-Ottawa

Regarding the Newcastle-Ottawa criteria for case-control studies, of the 13 works, 11 of them (84.6%) reached a total score higher than six, which cover the selection criteria for groups constitution, comparability among them and chances of exposure to the studied condition (studies 1, 2, 3, 5, 6, 8, 9, 10, 11, 12, 13). Among all the criteria, one of the issues was the least fulfilled and was the absence of the evaluation to exclude the ASD diagnosis in the control groups (studies 2, 3, 9, 10, 11).

DISCUSSION

This systematic literature review aimed to identify a possible relationship between the terminologies to study DBS in ASD and the tools used in their search. Other objectives established were to identify the occurrence of DBS and the outcome in relation to physiological mechanisms involved in it.

Regarding the instruments of DBS evaluation, it could be noted that most of the studies used questionnaires, but there were a variety of them^(7,8,17,37-46). One of them has different versions according to the age of the individual^(38,40) and most of them were answered by parents, teachers and/or caregivers^(7,37,43,44,46).

That diversity may suggest the absence of a gold standard tool in the investigation of DBS in ASD, which can be justified by two aspects. The first aspect is the heterogeneity of ASD manifestations, which vary from the absence of communicative and social interaction to its presence in a functional manner; thus, a single instrument may be more indicated to a group of individuals, but not to others. The second aspect relates to chronological age because questionnaires, even from guardians answering them, should be adapted to contemplate characteristics of each age group.

In the studies analyzed, the occurrence of DBS in individuals with ASD confirms what literature reports for this population, although it is extremely variable, from 18 to 90.0%^(25,29,41,46-50). Interestingly, two of the studies, from different groups, presented a similar frequency, 68.0 and 69.0%^(25,29). Another study reported the occurrence of disorder in 47.4%, but visual and auditory hyper-responsiveness were computed concomitantly⁽⁴⁶⁾. If we assume the similarity of DBS with HPA description, the percentages of DBS reported above are higher than HPA in pediatric population without ASD and/or hearing loss, which is 3.2 to 17.1%^(19,26,51,52).

The HPA was identified in only two of the studies^(8,25), as recommended by specialists in the area^(19,26,31). The results showed that children with ASD studied presented acoustic discomfort thresholds at lower intensities than usual^(8,25).

The other studies that used auditory tests for investigating DBS in ASD assumed that the symptom is more frequent in ASD and had as objective evaluating the involvement or not of the central auditory nervous system (CANS) mechanisms^(8,16,39,42).

Two of the studies showed that children with ASD had altered neural synchrony of auditory pathways, both at subcortical and cortical levels^(39,44). Another study⁽⁷⁾ reported a worse score in behavioral speech in noise test, a low redundancy monotic listening mechanism that makes up the auditory processing^(10,53). Imaging studies identify noise attenuation to the detriment of verbal message as a mechanism that starts in cochlear nuclei (low trunk) and extends to inferior colliculus (upper trunk) and cortical areas⁽⁵⁴⁾.

Although the results mentioned above describe the afferent auditory pathway, there is a hypothesis that HPA, regardless of population, is modulated by auditory efferent system, mainly by olivocochlear medial bundle^(17,54). This was the hypothesis studied by two of the studies with ASD, Danesh and Kaf⁽¹⁶⁾ and Wilson et al.⁽¹⁷⁾, who reported that children with ASD show response values for the cochlear inhibitory effect (suppression of otoacoustic emissions) lower than the group without ASD, which evidences a failure in sensory-neural modulation.

Two studies were included in the systematic review that did not use auditory tests, the studies of Thabet and Zaghoul⁽³⁹⁾ e Thabet⁽⁴²⁾. The studies started to assume that the individuals with ASD show DBS, which they call HSA, and this auditory symptom is commonly found in individuals with semicircular canal dehiscence (posterior labyrinth of the inner ear)⁽³⁹⁾. Based on this assumption, the authors investigated the VEMP in the group with ASD, accompanied by an imaging exam to identify the dehiscence of the vestibular canal. They identified its presence in the group with ASD, in six children of a total of 14, but five out of those six, had HSA⁽⁴²⁾. Due to the anatomical and physiological characteristics of inner ear, between the anterior and posterior labyrinth, the authors judged as pertinent the permanence of these two papers.

As initially described, the different methodologies used in the studies made it difficult to compare them, especially in relation to the results of auditory tests. There are indications that DBS in ASD, regardless of the terminology, there is the involvement of auditory subcortical and cortical neural afferent pathways, as well as the efferent pathway.

The fact that there was no minimum date limit for the research in the database, contributed to the fact that, only 13 studies out of 692, were in accordance with the established criteria, which were, in a certain way, rigid, but necessary for the objectives of the research. Even so, of the 13 selected, only nine of them used auditory tests and six investigated DBS with two different tools, the questionnaire and at least one auditory test. These results highlight the need for further studies to understand better the relationship of hearing test results in individuals with ASD with DBS.

CONCLUSION

The term AHS was the most used to name DBS in ASD, followed by HPA and AHR, but no relation was found between the terms and the respective investigative tool. The use of questionnaires was the most used instrument to study the occurrence of DBS, which ranged from 42.1 to 69.0%. The auditory tests, when performed, showed the involvement of afferent and efferent

auditory neural pathways in the response of DBS in ASD. The better understanding of DBS in ASD may contribute to specific therapeutic interventions in the population with ASD.

REFERENCES

1. Kanner L. Autistic disturbances of affective contact. *Acta Paedopsychiatr.* 1969;35(4):100-36. PMID:4880460.
2. Frith U, Happé F. Autism spectrum disorder. *Curr Biol.* 2005;19(19):786-90. <http://dx.doi.org/10.1016/j.cub.2005.09.033>. PMID:16213805.
3. Rogers SJ, Hepburn S, Wehner E. Parent reports of sensory symptoms in toddlers with autism and those with other developmental disorders. *J Autism Dev Disord.* 2003;33(6):631-42. <http://dx.doi.org/10.1023/B:JADD.0000006000.38991.a7>. PMID:14714932.
4. O'Riordan M, Passetti F. Discrimination in autism within different sensory modalities. *J Autism Dev Disord.* 2006;5(5):665-75. <http://dx.doi.org/10.1007/s10803-006-0106-1>. PMID:16639532.
5. Ben-Sasson A, Hen L, Fluss R, Cermak SA, Engel-Yeger B, Gal E. A meta-analysis of sensory modulation symptoms in individuals with autism spectrum disorders. *J Autism Dev Disord.* 2009;39(1):1-1. <http://dx.doi.org/10.1007/s10803-008-0593-3>. PMID:18512135.
6. McCormick C, Hepburn S, Young GS, Rogers SJ. Sensory symptoms in children with autism spectrum disorder, other developmental disorders and typical development: a longitudinal study. *Autism.* 2016;5(5):572-9. <http://dx.doi.org/10.1177/1362361315599755>. PMID:26395236.
7. Bhatara A, Babikian T, Laugeson E, Tachdjian R, Sininger Y. Impaired timing and frequency discrimination in high-functioning autism spectrum disorders. *J Autism Dev Disord.* 2013;43(10):2312-28. <http://dx.doi.org/10.1007/s10803-013-1778-y>. PMID:23386117.
8. Dunlop WA, Enticott PG, Rajan R. Speech discrimination difficulties in high-functioning autism spectrum disorder are likely independent of auditory hypersensitivity. *Front Hum Neurosci.* 2016;401(10):401. <http://dx.doi.org/10.3389/fnhum.2016.00401>. PMID:27555814.
9. Baranek GT, Boyd BA, Poe MD, David FJ, Watson LR. Hyperresponsive sensory patterns in young children with autism, developmental delay, and typical development. *Am J Ment Retard.* 2007;12(4):233-45. [http://dx.doi.org/10.1352/0895-8017\(2007\)112\[233:HSPIYC\]2.0.CO;2](http://dx.doi.org/10.1352/0895-8017(2007)112[233:HSPIYC]2.0.CO;2). PMID:17559291.
10. American Academy of Audiology. American Academy of Audiology Clinical Practice Guidelines: 2010. Diagnosis, treatment and management of children and adults with central auditory processing disorder. Reston: American Academy of Audiology [citado em 2016 Dez 16]. Disponível em: <http://www.audiology.org/resources/documentlibrary/documents/capd>
11. Dawes PL, Bishop DV, Sirimanna T, Bamiou DE. Profile and etiology of children diagnosed with auditory processing disorder (APD). *Int J Pediatr Otorhinolaryngol.* 2008;72(4):483-9. <http://dx.doi.org/10.1016/j.ijporl.2007.12.007>. PMID:18262288.
12. Iliadou V, Bamiou DE, Kaprinis S, Kandylis D, Kaprinis G. Auditory processing disorders in children suspected or learning disabilities a need for screening? *Int J Pediatr Otorhinolaryngol.* 2009;73(7):1029-34. <http://dx.doi.org/10.1016/j.ijporl.2009.04.004>. PMID:19427040.
13. Gravel JS, Dunn M, Lee WW, Ellis MA. Peripheral audition of children on the autistic spectrum. *Ear Hear.* 2006;27(3):299-312. <http://dx.doi.org/10.1097/01.aud.0000215979.65645.22>. PMID:16672798.
14. Rosenhall U, Nordin V, Sandström M, Ahlsén G, Gillberg C. Autism and hearing loss. *J Autism Dev Disord.* 1999;29(5):349-57. <http://dx.doi.org/10.1023/A:1023022709710>. PMID:10587881.
15. Tharpe AM, Bess FH, Sladen DP, Schissel H, Couch S, Schery T. Auditory characteristics of children with autism. *Ear Hear.* 2006;27(4):430-41. <http://dx.doi.org/10.1097/01.aud.0000224981.60575.d8>. PMID:16825892.
16. Danesh AA, Kaf WA. DPOAEs and contralateral acoustic stimulation and their link to sound hypersensitivity in children with autism. *Int J*

- Audiol. 2012;5(4):345-52. <http://dx.doi.org/10.3109/14992027.2011.626202>. PMID:22299666.
17. Wilson US, Sadler KM, Hancock KE, Guinan JJ Jr, Lichtenhan JT. Efferent inhibition strength is a physiological correlate of hyperacusis in children with autism spectrum disorder. *J Neurophysiol.* 2017;118(2):1164-72. <http://dx.doi.org/10.1152/jn.00142.2017>. PMID:28592687.
 18. Stiegler LN, Davis R. Understanding sound sensitivity in individuals with autism spectrum disorders. *Focus on Autism and Other Developmental Disabilities.* 2010;25(2):67-75. <https://doi.org/10.1177/1088357610364530>.
 19. Tyler RS, Pienkowski M, Roncancio ER, Jun HJ, Brozoski T, Dauman N, et al. A review of hyperacusis and future directions: part I. Definitions and manifestations. *Am J Audiol.* 2014;23(4):402-19. http://dx.doi.org/10.1044/2014_AJA-14-0010. PMID:25104073.
 20. Northen JL, Dows MP. *Hearing in children.* 3rd ed. Filadélfia: Lippincott Williams & Wilkin; 1984. p. 89.
 21. International Bureau for Audiophonology. BIAP recommendation 02/1: audiometric classification of hearing impairments [citado em 2017 Abr 9]. Disponível em: <https://www.biap.org/en/recommendation/recommendations-pdf/ct-02-classification-des-deficiences-auditives-1/55-02-1-audiometric-classification-of-hearing-impairments>
 22. OMS: Organização Mundial de Saúde. [Internet]. Hearing loss grades and the international classification of functioning, disability and health [citado em 2014 Abr 10]. Disponível em: http://www.who.int/pbd/deafness/hearing_impairment_grades/en/
 23. ISO: International Organization for Standardization [Internet]. ISO 8253-1:2010. Acoustics - Audiometric test methods - Part 1: Pure-tone air and bone conduction audiometry. Geneva: ISO.
 24. American National Standard Institute. American National Standard specification for audiometers (ANSI S3.6-1996). New York: ANSI; 1996.
 25. Khalfa S, Bruneau N, Rogé B, Georgieff N, Veuillet E, Adrien JL, et al. Increased perception of loudness in autism. *Hear Res.* 2004;198(1-2):87-92. <http://dx.doi.org/10.1016/j.heares.2004.07.006>. PMID:15617227.
 26. Coelho CB, Sanchez TG, Tyler RS. Hyperacusis, sound annoyance, and loudness hypersensitivity in children. *Prog Brain Res.* 2007;166:169-78. [http://dx.doi.org/10.1016/S0079-6123\(07\)66015-4](http://dx.doi.org/10.1016/S0079-6123(07)66015-4). PMID:17956781.
 27. Dauman R, Bouscau-Faure F. Assessment and amelioration of hyperacusis in tinnitus patients. *Acta Otolaryngol.* 2005;5(5):503-9. <http://dx.doi.org/10.1080/00016480510027565>. PMID:16092541.
 28. Leekam SR, Nieto C, Libby SJ, Wing L, Gould J. Describing the sensory abnormalities of children and adults with autism. *J Autism Dev Disord.* 2007;37(5):894-910. <http://dx.doi.org/10.1007/s10803-006-0218-7>. PMID:17016677.
 29. Danesh AA, Lang D, Kaf W, Andreassen WD, Scott J, Eshraghi AA. Tinnitus and hyperacusis in autism spectrum disorders with emphasis on high functioning individuals diagnosed with Asperger's Syndrome. *Int J Pediatr Otorhinolaryngol.* 2015;79(10):1683-8. <http://dx.doi.org/10.1016/j.ijporl.2015.07.024>. PMID:26243502.
 30. Paulin J, Andersson L, Nordin S. Characteristics of hyperacusis in the general population. *Noise Health.* 2016;18(83):178-84. <http://dx.doi.org/10.4103/1463-1741.189244>. PMID:27569405.
 31. Pienkowski M, Tyler RS, Roncancio ER, Jun HJ, Brozoski T, Dauman N, et al. A review of Hyperacusis and future directions: part II. Measurement, mechanisms, and treatment. *Am J Audiol.* 2014;23(4):420-36. http://dx.doi.org/10.1044/2014_AJA-13-0037. PMID:25478787.
 32. Desai NS, Rutherford LC, Turrigiano GG. Plasticity in the intrinsic excitability of cortical pyramidal neurons. *Nat Neurosci.* 1999;2(6):515-20. <http://dx.doi.org/10.1038/9165>. PMID:10448215.
 33. McDermott HJ, Lech M, Kornblum MS, Irvine DR. Loudness perception and frequency discrimination in subjects with steeply sloping hearing loss: possible correlates of neural plasticity. *J Acoust Soc Am.* 1998;104(4):2314-25. <http://dx.doi.org/10.1121/1.423744>. PMID:10491696.
 34. Moore BCJ, Vinay SN. Enhanced discrimination of low-frequency sounds for subjects with high-frequency dead regions. *Brain.* 2009;132(Pt 2):132524-36. PMID:19036764.
 35. Gothelf D, Farber N, Raveh E, Apter A, Attias J. Hyperacusis in Williams syndrome: characteristics and associated neuroaudiologic abnormalities. *Neurology.* 2006;66(3):390-5. <http://dx.doi.org/10.1212/01.wnl.0000196643.35395.5f>. PMID:16476938.
 36. Wells GA, Shea B, O'Connell, D, Peterson J, Welch V, Losos M, Tugwell P. *The Newcastle-Ottawa Scale (NOS) for assessing the quality of nonrandomized studies in meta-analyses.* Ottawa: Ottawa Hospital Research Institute.
 37. Matsuzaki J, Kagitani-Shimono K, Goto T, Sanefuji W, Yamamoto T, Sakai S, et al. Differential responses of primary auditory cortex in autistic spectrum disorder with auditory hypersensitivity. *Neuroreport.* 2012;23(2):113-8. <http://dx.doi.org/10.1097/WNR.0b013e32834ebf44>. PMID:22146579.
 38. Dunn W. *Infant/toddler sensory profile.* San Antonio, TX: The Psychological Corporation; 2002.
 39. Thabet EM, Zaghoul HS. Auditory profile and high resolution CT scan in autism spectrum disorders children with auditory hypersensitivity. *Eur Arch Otorhinolaryngol.* 2013;270(8):2353-8. <http://dx.doi.org/10.1007/s00405-013-2482-4>. PMID:23580033.
 40. Dunn W. *Sensory profile.* San Antonio, TX: The Psychological Corporation; 1999.
 41. Bhatara A, Quintin EM, Fombonne E, Levitin DJ. Early sensitivity to sound and musical preferences and enjoyment in adolescents with autism spectrum disorders. *Psychomusicology.* 2013;32(2):100-8. <http://dx.doi.org/10.1037/a0033754>.
 42. Thabet EM. Ocular vestibular evoked myogenic potentials n10 response in autism spectrum disorders children with auditory hypersensitivity: an indicator of semicircular canal dehiscence. *Eur Arch Otorhinolaryngol.* 2014;271(5):1283-8. <http://dx.doi.org/10.1007/s00405-013-2736-1>. PMID:24100882.
 43. Matsuzaki J, Kagitani-Shimono K, Sugata H, Hirata M, Hanaie R, Nagatani F, et al. Progressively increased M50 responses to repeated sounds in autism spectrum disorder with auditory hypersensitivity: a magnetoencephalographic study. *PLoS One.* 2014;9(7):e102599. <http://dx.doi.org/10.1371/journal.pone.0102599>. PMID:25054201.
 44. Donkers FC, Schipul SE, Baranek GT, Cleary KM, Willoughby MT, Evans AM, et al. Attenuated auditory event-related potentials and associations with atypical sensory response patterns in children with autism. *J Autism Dev Disord.* 2015;45(2):506-23. <http://dx.doi.org/10.1007/s10803-013-1948-y>. PMID:24072639.
 45. Khalfa S, Dubal S, Veuillet E, Perez-Diaz F, Jouvent R, Collet L. Psychometric normalization of a hyperacusis questionnaire. *ORL J Otorhinolaryngol Relat Spec.* 2002;64(6):436-42. <http://dx.doi.org/10.1159/000067570>. PMID:12499770.
 46. Green SA, Hernandez L, Tottenham N, Krasileva K, Bookheimer SY, Dapretto M. Neurobiology of sensory overresponsivity in youth with autism spectrum disorders. *JAMA Psychiatry.* 2015;72(8):778-86. <http://dx.doi.org/10.1001/jamapsychiatry.2015.0737>. PMID:26061819.
 47. Tomchek SD, Dunn W. Sensory processing in children with and without autism: a comparative study using the short sensory profile. *Am J Occup Ther.* 2007;61(2):190-200. <http://dx.doi.org/10.5014/ajot.61.2.190>. PMID:17436841.
 48. Schoen SA, Miller LJ, Brett-Green B, Hepburn SL. Psychophysiology of children with autism spectrum disorder. *Res Autism Spectr Disord.* 2008;3(3):417-29. <http://dx.doi.org/10.1016/j.rasd.2007.09.002>.
 49. Baranek GT, Berkson G. Tactile defensiveness in children with developmental disabilities: responsiveness and habituation. *J Autism Dev Disord.* 1994;24(4):457-71. <http://dx.doi.org/10.1007/BF02172128>. PMID:7961330.

50. Gomes E, Pedroso FS, Wagner MB. Auditory hypersensitivity in the autistic spectrum disorder. *Pro Fono*. 2008;20(4):279-84. <http://dx.doi.org/10.1590/S0104-56872008000400013>. PMID:19142473.
51. Rosing SN, Schmidt JH, Wedderkopp N, Baguley DM. Prevalence of tinnitus and hyperacusis in children and adolescents: a systematic review. *BMJ Open*. 2016;6(6):e010596. <http://dx.doi.org/10.1136/bmjopen-2015-010596>. PMID:27259524.
52. Aazh H, McFerran D, Salvi R, Prasher D, Jastreboff M, Jastreboff P. Insights from the first international conference on hyperacusis: causes, evaluation, diagnosis and treatment. *Noise Health*. 2014;16(69):123-6. <http://dx.doi.org/10.4103/1463-1741.132100>. PMID:24804717.
53. Moore DR, Rosen S, Bamiou DE, Campbell NG, Sirimanna T. Evolving concepts of developmental auditory processing disorder (APD): a British Society of Audiology APD special interest group 'white paper'. *Int J Audiol*. 2013;1(1):3-13. <http://dx.doi.org/10.3109/14992027.2012.723143>. PMID:23039930.
54. Basta D, Tzschentke B, Ernst A. Noise-induced cell death in the mouse medial geniculate body and primary auditory cortex. *Neurosci Lett*. 2005;38(1-2):199-204. <http://dx.doi.org/10.1016/j.neulet.2005.02.034>. PMID:15882817.

Authors' contributions

ACGFS participated in the study design, data collection and manuscript preparation; SZ participated in the study design, supervision of data collection and contributed to the preparation of the manuscript and final correction; EFF, coordinator of the study, participated in the design and final correction of the manuscript.