

Original Article

Parents' perception of auditory hypersensitivity in children with clinical signs of risk for Autism Spectrum Disorder¹

Percepção dos pais sobre hipersensibilidade auditiva de crianças com sinais clínicos de risco para o Transtorno do Espectro do Autismo

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Abstract

Objective: To investigate the occurrence and type of auditory hypersensitivity in children with clinical signs of Autism Spectrum Disorder through parents' reports in the context of the COVID-19 pandemic. Method: This is a cross-sectional and descriptive study, composed of parents of 11 children with clinical signs of risk for Autism Spectrum Disorder. The children were of both sexes, with a mean age of 44.8 months. Parents answered a previously validated questionnaire about their children's auditory hypersensitivity behaviors by telephone. A score equal to or greater than eight points in the general score was considered as a sign of hypersensitivity. Results: 63.6% of the children presented results indicative of hypersensitivity and 54.5% obtained maximum scores on questions related to irritability to specific sounds. The sounds cited as generating irritability were: clapping, fireworks, shouting, construction tools, singing and cell phone ringtones. Conclusion: Auditory hypersensitivity was observed, especially related to irritability, which suggests a relationship with the limbic system and, therefore, may refer to misophonia. Thus, it is important for the multidisciplinary team to pay attention to the occurrence and characteristics of hypersensitivity in this population, in order to maximize favorable conditions for rehabilitation.

Keywords: Hyperacusis, Auditory Perceptual Disorders, Autism Spectrum Disorder, Child.

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<u>Resumo</u>

Objetivo: Averiguar a ocorrência e o tipo de hipersensibilidade auditiva em crianças com sinais clínicos de Transtorno do Espectro do Autismo por meio do relato dos pais no contexto da pandemia da COVID-19. Método: Trata-se de um estudo transversal e descritivo, composto por pais de 11 crianças com sinais clínicos de risco para Transtorno do Espectro do Autismo. As crianças eram de ambos os sexos, com média de 44,8 meses de idade. Os pais responderam, por meio de contato telefônico, a um questionário, previamente validado, sobre comportamentos de hipersensibilidade auditiva de seus filhos. Foi considerado como sinalizador de hipersensibilidade a pontuação igual ou superior a oito pontos no escore geral. Resultados: 63,6% das crianças apresentaram resultado indicativo de hipersensibilidade e 54,5% obtiveram pontuação máxima nas questões relacionadas à irritabilidade a sons específicos. Os sons citados como geradores de irritabilidade foram: palmas, fogos, gritos, ferramentas de construção, canto e toque de celular. Conclusão: constatou-se ocorrência de hipersensibilidade auditiva, especialmente relacionada à irritabilidade, o que sugere relação com o sistema límbico e, portanto, pode remeter à misofonia. Assim, torna-se importante a equipe multidisciplinar atentar-se à ocorrência e a características de hipersensibilidade dessa população, a fim de maximizar condições favoráveis à reabilitação.

Palavras-chave: Hiperacusia, Transtornos da Percepção Auditiva, Transtorno do Espectro Autista, Criança.

Introduction

Autism Spectrum Disorder (ASD) is defined, according to the most recent version of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5), as a neurodevelopmental disorder that involves impaired communication and social interaction, in addition to restricted patterns of behavior, the stereotypies (American Psychiatric Association, 2014). Additionally, the referred manual indicates as frequent manifestations the hyper or hyporesponsiveness to sensorial stimuli.

From this perspective, studies have shown that more than 40% of children with ASD have some sensory processing disorder (Ben-Sasson et al., 2009; Schaaf et al., 2014; Tyler et al., 2015), reaching estimates of up to 90% (Leekam et al., 2007); such changes are closely linked to stereotypes and inappropriate behaviors (Boyd et al., 2009; Boyd et al., 2010; Caminha & Lampreia, 2012). Specifically in relation to auditory sensory alterations, previous studies showed wide variability in prevalence, with results from 15% to 100% (Khalfa et al., 2004; Gomes et al., 2008; Bhatara et al., 2013; Danesh et al., 2015).

Thus, auditory hypersensitivity is configured as an abnormal perception of sound, which can impact different everyday situations, such as social and leisure activities (Aazh et al., 2016). However, it is important to emphasize that such a condition can be manifested and defined in different ways. In the literature, there are three main nomenclatures described: misophonia, an irritability to specific sounds; phonophobia, which consists of fear of certain sounds; and hyperacusis, characterized by an abnormal increase in sound sensitivity (Aazh et al., 2014). Furthermore, hypersensitivity may also

be associated with the auditory recruitment phenomenon in individuals with hearing loss (Sanchez et al., 1999).

Therefore, psychoacoustic tests are considered the gold standard for identifying auditory hypersensitivity (Tyler et al., 2014). However, due to daily living in different family contexts, it is the parents who first suspect an alteration and, generally, their suspicions are accurate and true. In this sense, interviews with parents of children with ASD are important sources of data (Coonrod & Stone, 2004).

A recent systematic review (Stefanelli et al., 2020), a scoping review (Potgieter et al., 2020) and a systematic review with meta-analysis (Williams et al., 2021) revealed that questionnaires, whether or not associated with structured interviews, have been the most common instruments in the investigation of the frequency of auditory hypersensitivity. Among such instruments, the questionnaire proposed by Coelho et al. (2007), used by different studies, both in its original form and in adapted versions, stands out.

Using the aforementioned questionnaire in the investigation of children between four and seven years of age with typical development, hyperacusis was diagnosed in 13.8% of the sample and the results indicated the use of shorter sentences by children with hyperacusis, in addition to a possible effect of this condition in lexical access (Ralli et al., 2018). When considering children at risk for ASD, the possibility of the influence of hyperacusis can be extrapolated to the entire diagnostic process and multidisciplinary intervention.

In challenging scenarios, such as the COVID-19 pandemic context, children with ASD have several particularities to face changes in safe and comfortable routines (Houting, 2020), and it is important to adopt intervention measures and strategies (Fernandes et al., 2021). On the other hand, such a context may be opportune for parents to observe their children's behavior, given that the period of social isolation and suspension of elective clinical care enables longer coexistence.

Although ASD has been studied over the years, as well as its perceptual changes, no evidence in the literature was found to determine the type of hypersensitivity present in this population. Such data is important to understand how this change occurs, as well as to provide parents and caregivers with intervention strategies and better quality of life. Therefore, having as references studies that showed changes in the limbic system, related to emotions, in children with ASD (Haznedar et al., 2000; Stigler et al., 2011), the hypothesis listed is that parents perceive behaviors of Misophonia-type auditory hypersensitivity, with irritability to specific sounds and consequent impact on daily life situations. Additionally, we hypothesized that, during the period of social isolation resulting from the COVID-19 pandemic, such behaviors could be minimized, since children possibly stayed longer in controlled environments and had less exposure to outdoor environments.

Therefore, considering that hearing is one of the sensory inputs of the human body, essential for the development of oral language, this study aimed to investigate the occurrence and type of auditory hypersensitivity in children with clinical signs of ASD through the report of the parents in the context of the COVID-19 pandemic.

Method

This is an observational and cross-sectional study carried out with the approval of the Research Ethics Committee of a Federal University, opinion No. 4,378,709. The cross-sectional design is most relevant when there is interest in frequency of occurrence measures, focusing on the number of existing events (Kesmodel, 2018), without interventions. Participants signed the Free and Informed Consent Term (ICF).

The sample consisted of parents of 11 children, of both sexes, between 26 and 62 months of age (mean of 44.8 and standard deviation of 12.4 months). Being parents or guardians of children under the age of six years, with previous hearing assessment indicating absence of hearing loss and clinical signs of ASD, confirmed by multidisciplinary assessment at a referral center in the state, was adopted as eligibility criteria. As exclusion criteria, the presence of visual impairment or associated syndromes was considered.

The children included in the study were in the process of being evaluated by a multidisciplinary team when the social distancing measures imposed by COVID-19 were initiated and they showed clinical signs of ASD; however, due to the pandemic context, no intervention was initiated.

For the present study, a previously validated questionnaire on auditory hypersensitivity was applied, consisting of seven questions, with possible answers "yes", "no" or "I don't know"; if affirmative, citing examples (Coelho et al., 2007). We chose to use this questionnaire because it includes questions related to all types of hypersensitivity, from fear to irritability to sounds. In addition, the instrument is easy to apply and to understand by the respondents, and includes an answer option "I don't know", which minimizes the chance of parents answering inaccurately in cases of doubts.

The application took place through telephone contact and the questions were asked to the parents or guardians by a single speech therapist, in order to reduce the probability of biases regarding the application.

The criterion used to establish the score and determine the presence of hypersensitivity was the same used in a previous study (Ralli et al., 2018). Scores are assigned to the first four questions and hypersensitivity was considered by obtaining a sum equal to or greater than eight points. For affirmative reports, that is, which corresponded to "yes", four points were assigned; for negatives, zero points were assigned, and, in case of doubt, two points were assigned to the question.

For questions 5 to 7, which refer to historical information regarding the use of medications, surgeries and episodes of otitis, similarly to previous studies, scores were not assigned, with a descriptive analysis of the responses obtained.

It is worth mentioning that, due to the COVID-19 pandemic scenario experienced at the time of the study, in addition to the questionnaire, an additional question was asked to the participants: "During the period of social isolation, was there any change in your child's hearing behavior? If so, please describe".

The data obtained were tabulated in Microsoft Excel 2016 and the analysis was performed using descriptive statistics.

Results and discussion

The sample of respondents of the questionnaire was entirely made up of mothers and grandmothers. When considering the first four questions, three children did not obtain any scores, with negative responses to all questions. This data demonstrates that, although hypersensitivity may be frequent in children with ASD, there are children who will not present such reactions. In addition, it was evident that the absence of complaints does not seem to be related to age, identified in children aged 26, 40 and 62 months, or to sex (Table 1).

Table 1. Characterization of the children in terms of age and sex and synthesis of the answers given by the guardians to four questions of the hyperacusis questionnaire.

			Q1	Q2	Q3	Q4	
Р	Age (Months)	Sex	Is your child too sensitive to everyday sounds?	your child does	Are there any sounds that your child finds painful?	Is there any sound that makes your child upset?	Total score
1	32	М	Yes	No	No	Yes	8*
2	46	М	Yes	Yes	No	Yes	12*
3	45	М	No	No	No	Yes	4
4	36	М	Yes	No	No	Yes	8*
5	43	М	Yes	Yes	Yes	Yes	16*
6	62	М	No	Yes	Yes	No	8*
7	39	F	Yes	Yes	No	No	8*
8	40	М	No	No	No	No	0
9	26	F	No	No	No	No	0
10	62	М	Yes	Yes	No	Yes	12*
11	62	М	No	No	No	No	0

Caption: Q= question; P= participant; *indicative score of auditory hypersensitivity.

In common with the different studies aimed at individuals with ASD, in the sample, there was a greater representation of males (81.8%) (Table 1). Among the eight participants who scored, the minimum score obtained was four points and the maximum score of the questionnaire, which is equivalent to 16 points, was obtained by one of the children (Table 1).

In the literature, evidence of abnormal responses to sounds in individuals with ASD throughout life is found (Williams et al., 2021). However, there is no characterization regarding the most frequent type of hypersensitivity, nor the sounds that commonly trigger such reactions. In addition, there is a divergence in the nomenclature (Baranek et al., 2007; Stiegler & Davis, 2010; Stefanelli et al., 2020) and there is no concrete scientific evidence about the pathophysiological mechanism involved, which also makes it difficult to know about the type of manifestation of this change. Some hypotheses range from a failure in neural encoding (Pienkowski et al., 2014) to peripheral areas, such as the efferent fibers of the olivocochlear system that reach the outer hair cells (Gothelf et al., 2006).

When considering the adopted criterion of a score equivalent to or greater than eight points, in our study, 63.6% of the children presented results indicative of

hypersensitivity. Among the six children who obtained the maximum score on question 1, five (83.3%) also scored on question 4 (Table 1). The sounds cited as irritating generators are shown in Figure 1.

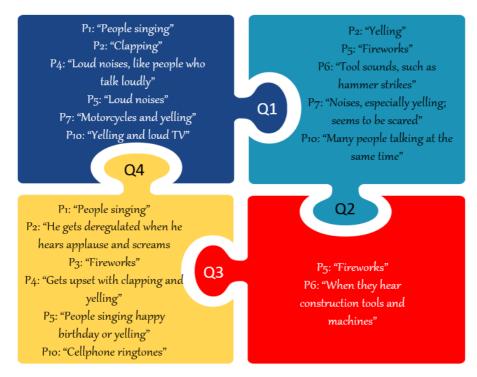


Figure 1. Description of sounds cited as irritant generators.

Previous research that applied a questionnaire with parents of children with ASD to estimate the occurrence of auditory hypersensitivity found wide variability, which can be attributed to the methodological differences used and in the characterization of the participants (Khalfa et al., 2004; Gomes et al., 2008; Bhatara et al., 2013; Danesh et al., 2015; Ralli et al., 2020). The percentage of 63.6% obtained in the present study reinforces the frequent perception of hypersensitivity in this population by parents and guardians and, therefore, emphasizes the importance of professionals involved in paying attention to the matter both in the evaluation and intervention process themselves and for the interview and guidance to family members.

When analyzed under the teachers' perception, the occurrence of auditory hypersensitivity in this population was estimated at about 30% and, when analyzed in a combined way between parents or guardians and teachers/therapists, the occurrence was similar, with 23.9% (Gomes et al., 2008). Although parents, teachers and therapists can perceive reactions that refer to hypersensitivity, the prevalence can still be underestimated and some reactions of discomfort, fear or irritability are not noticed, considering that, in the evaluation through psychoacoustic tests, 63% of the children could not tolerate sounds at an intensity higher than 80 decibel hearing level (dBHL) (Gomes et al., 2008), a percentage similar to that obtained in the parental perception of

the present study. Another detail that deserves attention in our results and that reiterates the observation of children's auditory behavior by their guardians is that no "I don't know" answer was obtained for any of the questions.

Individuals with ASD may present a variety of complaints related to both misophonia and hyperacusis and phonophobia (Williams et al., 2021). Among the children participating in this study, 54.5% scored the maximum score on question 4, which provides information about annoyance or irritability for some sound (Table 1). Such data refers to a negative emotional reaction and, therefore, may suggest a relationship with misophonia. Although most of these children also had a maximum score in relation to the sensitivity of everyday sounds (Table 1), it is possible to observe a group of very specific sounds for questions 1 and 4 (Figure 1).

Although there is a limited number of studies that have investigated misophonia in this population, there is evidence that a proportion of children with ASD may also have some diagnostic criteria for misophonia (Williams et al., 2021). On the other hand, only two informants presented reports of demonstration of painful sensations for some auditory stimulus (Table 1), which indicates that despite the possibility of the occurrence of characteristic behaviors of phonophobia in children with ASD, these were less frequent in the series studied.

Data from a literature review showed that, regardless of the assessment using behavioral tests, questionnaires or structured interviews, the results tend to support the hypothesis that this population often perceives everyday sounds abnormally and, similarly to our findings, pointed out that some individuals have anxiety associated with specific sounds (phonophobia) that may be concomitant with hyperacusis (Williams et al., 2021).

Regarding questions 5 to 7 of the questionnaire, according to those responsible, none of the participants in this study used medication (Q5), had ear surgery (Q6) or had episodes of otitis in the last 12 months (Q7), conditions that could have some influence on the occurrence of complaints related to auditory hypersensitivity. Additionally, there was no perception of change in auditory behavior during the period of the COVID-19 pandemic for any of the children, whether it was evidence of atypical reactions or minimization of common responses. Such data may indicate that the sounds that trigger negative reactions in children occur even in more controlled environments, given that, during the period of isolation, children were limited in exposure to outdoor environments.

An auditory symptom associated with stress that became more noticeable in the context of social isolation was tinnitus (Beukes et al., 2020). Although the literature is variable, there is a consensus that, in addition to hyperacusis, tinnitus is relatively common in childhood, with a prevalence that varies considerably according to the study design and population, as well as the research question, with a prevalence of 4.7% to 74.9% (Rosing et al., 2016). The impact of tinnitus appears to be smaller in children than in adults and, particularly in individuals with ASD, it can be difficult to identify.

A study evaluated the prevalence of tinnitus and hyperacusis in individuals with Asperger's Syndrome and identified the occurrence of combined symptoms in 31% of its sample. Despite including a broad age group, from four to 42 years, in the analysis by age group, it was found that both isolated and combined symptoms were identified in children (Danesh et al., 2015). Thus, additional studies, with larger samples, are important.

In view of the results obtained in the present study, it is emphasized that auditory hypersensitivity is frequent in this population. Obtaining parental information about the type of hypersensitivity and what are the triggering sound stimuli, even during the diagnostic process, may favor care in different areas, whether in clinical care or in sensitization activities, as suggested for challenging scenarios such as of the COVID-19 pandemic (Fernandes et al., 2021).

The perception of those responsible should be considered not only for possible complaints of hearing loss, but also for other auditory symptoms prevalent in this population. Such care can help, from performing a complete audiological assessment, providing information to guide the best conditioning strategy for behavioral tests, to avoiding sounds that may trigger discomfort or irritability throughout the assessment process and intervention of global development.

Conclusion

When investigating the occurrence of auditory hypersensitivity in children with clinical signs of ASD, it was found that, according to the parents' perception, it was present in 63.6% of the children. The most common manifestation is related to irritability, which suggests a relationship with the limbic system and may, therefore, refer to the type of auditory hypersensitivity called misophonia.

The study had limitations regarding the restricted sample and, as it was a crosssectional study carried out during the diagnostic intervention process, it did not guarantee that all children would complete the diagnosis. However, the data obtained highlight for the multidisciplinary team the occurrence and characteristics of hypersensitivity in this population with clinical signs of risk for ASD. Knowledge of such information can maximize rehabilitation and minimize possible negative impacts in daily life situations, in the school context, in external environments and even in more restricted situations such as social isolation. When considering the prevalence of ASD, the importance of including individuals with this condition in different sectors of society and the need for health education actions, the data obtained makes an important contribution to society in general.

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Author's Ccontributions

Krisia Thayná Lima da Costa: participated in the conception and design of the study, as well as in the interpretation of the results and the writing of the article; Vanessa Giacchini e Ana Manhani Cáceres-Assenço: contributed to the stages of design, interpretation of results and review of the article; Eliene Silva Araújo: participated in the conception and design of the study, in the analysis and interpretation of results and in the review of the article. All authors approved the final version of the text.

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