



## Clinical Profile of Impaired Hearing in Children with Autism Spectrum Disorder (ASD) at UGM Academic Hospital

\*Ade Febrina Lestari<sup>1,2</sup>, Mei Neni Sitaresmi<sup>1,3</sup>, Anton Sony Wibowo<sup>1,4</sup>, Shinta Kusumalarna Sari<sup>5</sup>, and Firda Ridhayanti<sup>5</sup>

<sup>1</sup>Faculty of Medicine, Public Health and Nursing, Universitas Gadjah Mada, Yogyakarta, Indonesia

<sup>2</sup>Department of Child Health, Academic Hospital Universitas Gadjah Mada, Yogyakarta, Indonesia

<sup>3</sup>Department of Child Health, Dr. Sardjito Hospital, Yogyakarta, Indonesia

<sup>4</sup>Department of Otolaryngology, Academic Hospital Universitas Gadjah Mada, Yogyakarta, Indonesia

<sup>5</sup>Academic Hospital Universitas Gadjah Mada, Yogyakarta, Indonesia

\*Correspondence: [adefebriana@ugm.ac.id](mailto:adefebriana@ugm.ac.id)

Publish: September 2024

### Abstract

**Background:** Hearing tolerance is impaired in children with Autism Spectrum Disorder (ASD), which can affect social and academic functioning. This study aims to describe the clinical profile of hearing impairments in children with ASD and determine the prevalence of such impairments.

**Method:** A cross-sectional study was conducted at Academic Hospital UGM on pediatric ASD patients aged 18 months to 12 years who underwent hearing exams like BERA (brain-evoked response auditory) and/or audiometry. ASD severity was assessed using the Childhood Autism Rating Scale (CARS) and a questionnaire on demographics and comorbidities, followed by BERA tests.

**Results:** A total of 41 ASD children were obtained, with 31 children (76%) being boys and an average diagnosis age of 3.3 years ( $\pm 1.3$  SD). Hearing loss was suffered by 4 children (10%) and 37 children (90%) had normal hearing. Sound sensitivity and degree of ASD correlated with head banging ( $p=0.01$ ), and male gender correlated with hearing loss ( $P=0.006$ ). CARS score was statistically associated with sound sensitivity ( $p=0.041$ ) and degree of ASD ( $<0.001$ ).

**Conclusion:** Children diagnosed with ASD face an increased susceptibility to communication impairments which can be attributed to hearing impairments. This issue warrants particular attention in the context of ASD, highlighting the necessity for thorough screening of hearing capabilities, especially in ASD children who are highly suspicious of having hearing loss.

**Keywords:** Autism Spectrum Disorder (ASD), hearing loss, clinical profile, CARS (childhood autism rating scale)

### 1. INTRODUCTION

Autism Spectrum Disorder (ASD), as delineated in the Diagnostic and Statistical Manual of Mental Disorders 5th edition (DSM-5), is characterized by persistent social interaction deficits in both social reciprocal relationships, verbal and nonverbal communicative behavior, and skills in developing, maintaining, and understanding relationships, accompanied by restricted and repetitive patterns of behavior, interests, and activities 1,2. The World Health Organization predicts that 1 in 160 children worldwide are affected by autism spectrum

disorder. Indonesia projecting an annual rise of 500 people suffering from ASD. Notably, in the year 2020-2021, there were 5,530 cases of developmental disorders in children, including ASD, receiving services at community health centers 3. Several factors contribute to heightened susceptibility to developing ASD in children, including environmental and genetic factors and the interaction of the two 4. The risk determinants encompass heritability, genetic changes, gender, immune dysfunction, prenatal and perinatal conditions, socioeconomic status, as well as exposure to pharmaceuticals and

harmful substances, with gender emerging as the primary factor influencing ASD prevalence 5.

Research findings indicate that the incidence of hearing impairment among children with ASD ranges from 4-7% 6,7. Typically, hearing loss in children with ASD is identified only after 48 months of age, predominantly falling within the 50-66 month bracket 8. Manifestations of hearing loss in children with ASD include difficulty with hearing abilities, tinnitus, auditory processing disorders, and hypersensitivity to loud sounds. Several studies report that the prevalence of hearing loss in ASD children is higher than in non-ASD children, varying from 18% to 37% 9. Hyperacusis, a form of hearing loss, induces extreme sensitivity or insensitivity to sound, potentially leading to ear discomfort and pain. Children with hearing loss generally have difficulty tolerating sounds that are not loud to others but are a nuisance to the sufferer, for example, the sound of a running water tap, the sound of a vehicle horn, a dishwasher, or folding paper 10. This heightened auditory sensitivity frequently serves as a pivotal indicator for an ASD diagnosis, exacerbating symptoms and eliciting adverse reactions like covering the ears, crying, or seeking to escape. Such reactions can detrimentally affect the child's social and academic performance 11-13. There has not been much research related to hearing loss in children with ASD. Most children

with ASD undergo hearing screening examinations due to communication disorders. It is hoped that this research will provide good data for the management of ASD.

## 2. MATERIALS AND METHODS

This research is a cross-sectional study. The population is all ASD children aged 18 months to 17 years who are patients at the Academic Hospital of the University of Gadjah Mada from March 2023 to November 2023. Inclusion criteria encompass; (1) a patient diagnosed with ASD by a specialist in growth and development at the UGM Academic Hospital; (2) age range of 18 months to 17 years; (3) children with ASD who undergo BERA and/or audiometry examinations; (4) parents are willing to take part in the research by filling out informed consent. Exclusion Criteria pertain to patients who failed to undergo Brain Evoked Response Auditory (BERA). Diagnosis of ASD was carried out by a pediatrician or growth and development consultant employing DSM-5 as a diagnostic instrument. This study has successfully undergone ethical evaluation by the Medical and Health Research Ethics Committee (MHRCE) Faculty of Medicine, Public Health and Nursing Universitas Gadjah Mada – Dr. Sardjito General Hospital with an ethical number No. KE/FK/1079/EC/2023.

## 3. RESULT

**Table 1.** Mother and child characteristics (n=41)

Variables	n (%)
Age (mean, SD)	4.8 (1.9)
Gender (n, %)	
Male	31 (76)
Female	10 (24)
Age of diagnosis (mean, SD)	3.3 (1.3)
Duration of therapy (mean, SD)	19.1 (21.7)
Mother's education (n, %)	
Low educational status	16 (39)
High educational status	25 (61)
Father's education (n, %)	
Low educational status	11 (27)
High educational status	30 (73)
Income (n, %)	
Low	4 (10)
Moderate	20 (49)
High	17 (41)
Hearing impairment (n, %)	
No	37 (90)
Yes	4 (10)
Sensitive to sound (n, %)	
No	27 (70)
Yes	14 (30)
Hit the head (n, %)	

No	32 (78)
Yes	9 (22)
Degree of ASD (n, %)	
Mild (n, %)	19 (46)
Moderate-severe (n, %)	22 (54)
CARS Score (mean, SD)	39.06 (3.71)

ASD = Autism Spectrum Disorder; CARS = Childhood Autism Rating Scale

The study included 41 children diagnosed with ASD, of which 31 (76%) were boys, with an average age of diagnosis of 3.3 years ( $\pm 1.3$  SD). Hearing loss was identified in 4 children (10%), while the remaining 37 children (90%) did not experience hearing loss.

Sound sensitivity was observed in 27 children (70%), while 14 children (30%) showed sound insensitivity. Repetitive head hitting occurred in 9 children (22%) without clear triggers. Among the participants, 22 children (54%) had moderate to severe ASD, and 19 children (46%) had mild ASD.

**Table 2.** CARS Score relationship based on variable

Variable	Sum	Df	Mean square	F	P value
Age	80.291	18	4.461	1.407	0.221
Mother's education	4.556	18	.253	1.071	0.434
Father's education	3.582	18	.199	.980	0.512
Family income	6.261	18	.348	.721	0.758
Age of diagnosed	30.071	18	1.671	.944	0.544
Duration of therapy	6912.839	18	384.047	.710	0.768
<b>Sensitive to sound</b>	5.920	18	.329	2.192	<b>0.041*</b>
Hit the head	3.558	18	.198	1.254	0.304
<b>Degree of ASD</b>	8.995	18	.500	9.162	<b>&lt;0.001*</b>
Comorbid	.480	18	.027	.255	0.998
Hearing impairment	1.526	18	.085	.896	0.590

P value is significant if  $<0.05$

Analysis with one-way ANOVA

Table 2 shows no significant association between hearing impairment and the mean CARS score ( $p=0.590$ ). However, sound sensitivity was significantly linked to the CARS score ( $p=0.041$ ) and the severity of ASD ( $p<0.001$ ).

#### 4. DISCUSSION

##### a. Mother and child characteristics

The majority of ASD children identified in the study are male, totaling 31 children (76%), while females account for 10 children (24%). There was no statistical difference between gender and the severity of ASD ( $p=0.175$ ). The average age of children with ASD was 4.8 years (1.9 SD), with the mean age of ASD diagnosis was 3.3 years (1.3 SD). The average duration of therapy, whether in the form of medical drug therapy or medical rehabilitation measures such as occupational therapy, speech therapy, sensory integration, and behavioral therapy by psychologists, was 19.1 months (21.7 SD) and there was no statistical difference between the therapy duration and the severity of ASD or the CARS score ( $P=0.519$ ).

Medical rehabilitation therapy and behavioral therapy by psychologists are not differentiated based on the type of therapy. A majority of mothers and fathers have attained higher education levels, exceeding the completion of high school, while the majority of parents fall within the income bracket of 2-5 million (49%).

Out of the 41 ASD children under study, only 4 individuals (10%) were identified as experiencing hearing impairments, specifically hyperacusis and Sensorineural Hearing Loss (SNHL). Challenges were encountered in assessing hyperacusis in ASD children, particularly those with moderate to severe severity and those under the age of 6. Consequently, the hearing loss assessment proceeded with BERA (Brain Evoked Response Audiometry). Sensitivity to sound was reported in 27 children (70%), with 14 children (30%) exhibiting

insensitivity to sound. Sensitivity to various sounds was noted, including a sister crying, a blender operating, a hair dryer functioning, a duck quacking, and thunder rumbling. Additionally, 9 children (22%) displayed repetitive head-hitting behavior without a discernible trigger. Of the total ASD participants, 22 children (54%) were classified as having moderate to severe severity, while 19 children (46%) exhibited mild severity. The mean score on the Childhood Autism Rating Scale (CARS) was determined to be 39.06 (3.71 SD).

The relationship between the degree of ASD severity based on the average CARS score and the variables studied can be seen in Table 2 below.

#### **b. CARS Score relationship based on variable**

The results presented in Table 2 of this investigation indicate the absence of a statistically significant association between hearing impairment and the mean CARS score ( $p=0.590$ ). What is interesting is that the presence of sensitivity to sound turns out to be statistically significant for the CARS score ( $p=0.041$ ). Apart from that, the CARS score was also significantly associated with the degree of ASD ( $p<0.001$ ). The CARS score serves as an evaluative instrument for categorizing the severity of ASD into mild, moderate, and severe classifications. Within this study, the severity classification of ASD was condensed into mild and moderate-severe categories. Through the application of correlation analysis, a statistically significant link was also established between sensitivity to sound from head impacts ( $p=0.01$ ) and hearing loss among male individuals ( $p=0.006$ ).

In this study, findings indicate a higher prevalence of Autism Spectrum Disorder (ASD) in males compared to females. This observation aligns with a report by the CDC (Centers for Disease Control and Prevention) which notes that males are four times more likely to be affected by ASD than females<sup>14,15</sup>. The age range for first diagnosis in this study was 3.3 years (1.3 SD). A meta-analysis conducted by van Hof et al. (2021) revealed that the mean age at diagnosis for ASD was 43.18 months (ranging from 30.90 to 74.70 months) among children under the age of 10<sup>16</sup>. The primary reason for delayed diagnosis was attributed to the challenge in clearly establishing the diagnosis of ASD subtype or identifying potential comorbidities. Notably, our research discovered that 4 children (10%) exhibited hearing impairment, while the remaining 37 children (90%) did not manifest any hearing issues. Zeglam and Al-Ksaik (2020) documented that out of 71 autistic

children at Tripoli University Hospital, 26 encountered Sensorineural Hearing Loss, advocating against conducting BERA tests in ASD children without parental suspicion of hearing loss, particularly in resource-limited settings<sup>17</sup>. This recommendation is given to developing countries, which can reduce unnecessary inspection costs.

Several studies have highlighted a higher incidence of hearing impairment in ASD children when compared to their non-ASD counterparts, with rates ranging from 18% to 37%<sup>9</sup> and even reaching 69%<sup>11</sup>. The forms of hearing loss in this study encompassed hyperacusis and SNHL. Consistent with the findings of Danesh et al. (2020), hearing anomalies such as tinnitus and hyperacusis were observed in children diagnosed with Asperger's syndrome, now categorized within the broad autism spectrum<sup>11</sup>. Challenges were encountered in conducting hyperacusis assessments in our study due to the majority of participants being ASD children with moderate to severe symptomatology, especially those under the age of six, who often display uncooperative behavior. Furthermore, the absence of a validated questionnaire in Indonesian prompted the study to solely utilize audiometry without specific inquiries regarding tinnitus or hyperacusis. The manifestation of hyperacusis and tinnitus may lead to difficulties in social interactions, with affected children displaying restlessness, covering of ears, outbursts, and head-banging<sup>9</sup>. Through Correlation analysis, a significant association was established between sound sensitivity triggering head-banging ( $p=0.01$ ) and the presence of gender and hearing impairment in ASD children ( $P=0.006$ ). Conversely, no significant correlation was detected between the CARS score and hearing loss, possibly influenced by the limited number of subjects experiencing hearing difficulties (10%).

The limitation inherent in this study is primarily attributed to the modest sample size. Furthermore, the diagnosis of hearing loss in children solely relies on audiometry and BERA, lacking supplementary questionnaires for tinnitus and hyperacusis assessment, potentially introducing bias to the findings concerning the presence of hearing impairment. The advantage of this research is that it can be the basis for subsequent research with a larger sample size, creating further research with validated tinnitus and hyperacusis questionnaires for children with ASD so that a simpler and cheaper method can reduce the cost of BERA examinations, especially in minimal health facilities.

## 5. CONCLUSIONS

A correlation exists between sensitivity to sound and the degree of ASD. Males are correlated with experiencing hearing loss. No statistically significant correlation was found between the CARS score and the hearing impairment. Children diagnosed with ASD face an increased susceptibility to communication impairments which can be attributed to hearing impairments. The low prevalence of hearing loss in children with ASD highlights the necessity for thorough screening of hearing capabilities, especially in ASD children who are highly suspicious of having hearing loss.

## REFERENCES

- World Health Organization. Autism [Internet]. 2022. Available from: <https://www.who.int/news-room/fact-sheets/detail/autism-spectrum-disorders>
- Tsai LY. The Impact of DSM-5 on Autism Spectrum Disorder. *J Exp Psychopathol.* 2015;2(1):3–16.
- Indonesian Ministry of Health. Autism A-Z Webinar Commemorating World Autism Awareness Day 2022 [Internet]. Indonesian Ministry of Health, Directorate General of Public Health. 2022. Available from: <https://kesmas.kemkes.go.id/konten/133/0/autisme-a-z-webinar-peringatan-hari-peduli-autisme-sedunia-2022#>.
- Chaste P, Leboyer M. Autism risk factors: Genes, environment, and gene-environment interactions. *Dialogues Clin Neurosci.* 2012;14(3):281–92.
- Salari N, Rasoulpoor S, Rasoulpoor S, Shohaimi S, Jafarpour S, Abdoli N, et al. The global prevalence of autism spectrum disorder: a comprehensive systematic review and meta-analysis. *Ital J Pediatr [Internet]. BioMed Central;* 2022;48(1). Available from: <https://doi.org/10.1186/s13052-022-01310-w>
- Augustyn M. Autism spectrum disorder (ASD) in children and adolescents: Terminology, epidemiology, and pathogenesis [Internet]. UpToDate. 2020. Available from: <https://www.uptodate.com/contents/autism-spectrum-disorder-asd-in-children-and-adolescents-terminology-epidemiology-and-pathogenesis#H18>
- Bougard C, Picarel-Blanchot F, Schmid R, Campbell R, Buitelaar J. Prevalence of Autism Spectrum Disorder and Co-morbidities in Children and Adolescents: A Systematic Literature Review. *Front Psychiatry.* 2021;12(October):1–16.
- Weissman L. Autism spectrum disorder in children and adolescents: Screening tools [Internet]. UpToDate. 2020. Available from: <https://www.uptodate.com/contents/autism-spectrum-disorder-in-children-and-adolescents-screening-tools>
- Potgieter I, Fackrell K, Kennedy V, Crunkhorn R, Hoare DJ. Hyperacusis in children: A scoping review. *BMC Pediatr. BMC Pediatrics;* 2020;20(1):1–13.
- Jastreboff PJ, Jastreboff MM. Decreased sound tolerance: Hyperacusis, misophonia, diplacusis, and polyacusis [Internet]. 1st ed. *Handbook of Clinical Neurology.* Elsevier B.V.; 2015. 375–387 p. Available from: <http://dx.doi.org/10.1016/B978-0-444-62630-1.00021-4>
- Danesh AA, Howery S, Aazh H, Kaf W, Eshraghi AA. Hyperacusis in Autism Spectrum Disorders. *Audiol Res.* 2021;11(4):547–56.
- Brignell A, Morgan AT, Woolfenden S, Klopper F, May T, Sarkozy V, et al. A systematic review and meta-analysis of the prognosis of language outcomes for individuals with autism spectrum disorder. *Autism Dev Lang Impair.* 2018;3.
- Di Renzo M, di Castelbianco FB, Alberto V, Antonio DV, Giovanni C, Vanadia E, et al. Prognostic factors and predictors of outcome in children with autism spectrum disorder: the role of the pediatrician. *Ital J Pediatr. Italian Journal of Pediatrics;* 2021;47(1):1–12.
- Centers for Disease Control and Prevention. Autism Spectrum Disorder (ASD) [Internet]. 2022. Available from: <https://www.cdc.gov/autism/about/index.html>
- Beggiato A, Peyre H, Maruani A, Scheid I, Rastam M, Amsellem F, et al. Gender differences in autism spectrum disorders: Divergence among specific core symptoms. *Autism Res.* 2017;10(4):680–9.
- van 't Hof M, Tisseur C, van Berckeleer-Onnes I, van Nieuwenhuyzen A, Daniels AM, Deen M, et al. Age at autism spectrum disorder diagnosis: A systematic review and meta-analysis from 2012 to 2019. *Autism.* 2021;25(4):862–73.
- Zeglam A, Al-Ksaik S. Hearing testing in autistic spectrum disorder: Is it unnecessary in low and middle-income countries? *East Mediterr Heal J.* 2020;26(2):176–81.