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Evaluation of Autism Spectrum Disorder Symptoms in Children with Hearing Loss

İşitme Kayıplı Çocuklarda Otizm Spektrum Bozukluğu Semptomlarının Değerlendirilmesi

Banu BAŞ[a], Gülse AKDEMİR[b], Elif Zehra BİBER[b], Rukiye ÇOLAK SİVRİ[c]

[a]Ankara Yıldırım Beyazıt University Faculty of Health Sciences, Department of Audiology, Ankara, Türkiye
[b]Ankara Yıldırım Beyazıt University Institute of Health Sciences, Department of Audiology, Ankara, Türkiye
[c]Private Physician, Ankara, Türkiye

ABSTRACT Objective: Autism Spectrum Disorder (ASD) and hearing loss can coexist or present with similar findings during early childhood. The aim of this study was to assess ASD symptoms in children with severe to profound hearing loss using the Autism Spectrum Screening Questionnaire (ASSQ). **Material and Methods:** This study included 32 children aged between 6-9 years with severe to profound hearing loss. The Turkish version of the ASSQ (ASSQ-TR) was administered to the children. **Results:** The mean score of all participants was 7.56 (±9.53). The scores of 5 participants exceeded the cutoff score of 16 and showed signs of ASD. The mean score of these children was 26.2 (±9.83). It is noteworthy that the gender of these 5 children is male. The weak negative correlation between the total score of the scale and the duration of the auditory device experience was statistically significant. **Conclusion:** According to the ASSQ-TR scale, the children who scored above the cutoff score had passed the early childhood period but were not assessed for ASD previously. The presence of similar findings in both ASD and hearing loss may delay the diagnosis and intervention of autism in children with hearing loss. Conversely, the overlap of symptoms may also lead to a delayed diagnosis of hearing loss in children with ASD. The overlap of the symptoms of hearing loss and ASD can overshadow the diagnoses, negatively affecting the intervention for individuals with either diagnosis. Assessment methods should be expanded, and professionals should be more attentive to children with either disorder.

Keywords: Autism spectrum disorder; hearing loss; neurodevelopmental disorders; child

ÖZET Amaç: Otizm Spektrum Bozukluğu (OSB) ve işitme kaybı, erken çocukluk döneminde benzer bulgularla ortaya çıkabilir veya birlikte görülebilir. Bu çalışmanın amacı, ileri ve çok ileri düzeyde işitme kaybı olan çocuklarda OSB belirtilerini Otizm Spektrum Tarama Anketinin Türkçe versiyonu (OSTA-TR) ile değerlendirmektir. **Gereç ve Yöntemler:** Çalışmaya, 6-9 yaşları arasında, ileri ve çok ileri düzeyde işitme kaybı olan 32 çocuk dâhil edilmiştir. Katılımcılara OSTA-TR'nin Türkçe versiyonu (OSTA-TR) uygulanmıştır. **Bulgular:** Tüm katılımcıların ortalama puanı 7,56 (±9,53) olarak bulunmuştur. Beş katılımcının puanı 16 olan kesme puanının üzerindedir ve bu çocuklarda OSB belirtileri gözlenmiştir. Bu çocukların ortalama puanı 26,2 (±9,83) olarak hesaplanmıştır. Bu 5 çocuğun tamamının erkek olması dikkat çekicidir. Ölçeğin toplam puanı ile işitsel cihaz kullanım süresi arasında zayıf düzeyde negatif bir korelasyon bulunmuş ve bu ilişki istatistiksel olarak anlamlı çıkmıştır. **Sonuç:** OSTA-TR ölçeğine göre kesme puanının üzerinde puan alan çocuklar, erken çocukluk dönemini geçmiş olmalarına rağmen daha önce OSB açısından değerlendirilmemiştir. OSB ve işitme kaybında benzer bulguların görülmesi, işitme kaybı olan çocuklarda otizm tanı ve müdahalesinin gecikmesine yol açabilir. Aksine, belirtilerin örtüşmesi OSB'li çocuklarda da işitme kaybı tanısının gecikmesine neden olabilir. İşitme kaybı ve OSB belirtilerinin örtüşmesi, tanı süreçlerini gölgede bırakarak her iki duruma yönelik müdahaleleri olumsuz etkileyebilir. Değerlendirme yöntemlerinin çeşitlendirilmesi ve uzmanların her iki durum için de daha dikkatli olmaları gerekmektedir.

Anahtar Kelimeler: Otizm spektrum bozukluğu; işitme kaybı; nörogelişimsel bozukluklar; çocuk

Correspondence: Banu BAŞ
Ankara Yıldırım Beyazıt University Faculty of Health Sciences, Department of Audiology, Ankara, Türkiye
E-mail: fzt_banu@hotmail.com

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Hearing loss, in the absence of sign language usage, leads to a lack of early language access, which can impact the development of children's' language, communication, and social skills. Hearing loss can lead to challenges in cognitive skills such as executive functions and working memory in children.[1] Hearing loss reduces the quality of life in school and social environments and leads to behavioral difficulties in children.[2,3] There is a positive relationship between hearing loss and internalizing behaviors, hyperactivity, and attention deficit.[4] Children with hearing loss may experience emotional and behavioral problems, such as anger and low self-esteem. Difficulties in understanding speech and language skills can complicate children's' learning processes and negatively affect their academic success.

Syndromes, diseases, or neurodevelopmental disorders can accompany hearing loss.[5] Autism Spectrum Disorder (ASD) is one of the diagnoses that can accompany hearing loss.[6] ASD is a neurodevelopmental disorder characterized by limitations in social communication and interaction as well as restrictive, repetitive behaviors.[7] Symptoms vary by chronological age, verbal and intellectual ability, and disorder severity. Specific symptoms include difficulties in understanding and responding appropriately to social communications, an inability to integrate spoken language with nonverbal cues, a lack of skill in using language in social contexts, and deficiencies in social awareness. Repetitive and inflexible behavior patterns, strict adherence to routines, overly ritualized behaviors, repetitive motor movements, and excessive or insufficient responses to sensory stimuli were also observed. These symptoms emerge in early childhood and cause impairments in significant areas of functioning. Individuals with ASD may engage in masking, but the diagnosis remains valid.[8]

Some conditions, such as cytomegalovirus, rubella, chromosomal anomalies, meningitis, and Down Syndrome, can also lead to both ASD and hearing loss.[9] Additionally, a recent study revealed that the MEF2C gene, which is associated with ASD symptoms, reduces neuronal activity in the auditory nerve.[10]

Diagnosing and evaluating one of these disorders in the presence of the other during early childhood is challenging. Similar symptoms in children with both hearing loss and ASD include echolalia, language delays, atypical use of facial expressions and gestures, unique gestures, sensory-seeking behaviors, weak responses to their name, and impaired speech, prosody, and voice volume.[11] There is no standardized assessment for diagnosing ASD in children with hearing loss.[11] Although the similar signs of ASD and hearing loss complicate the diagnosis, professionals specialized in both ASD and hearing loss can differentiate which symptoms are associated with each condition.[11]

Various studies have examined the prevalence of the co-occurrence of hearing loss and ASD from different perspectives. Jure et al. found the rate of ASD in children with hearing loss to be 5.3%.[12] Rosenhall et al. reported the rate of hearing loss in children diagnosed with ASD to be 7.9%.[13] Tas et al. found that 6% of children with ASD had hearing loss.[6] A meta-analysis evaluating the co-occurrence of these 2 disorders found the prevalence rate to be 9%.[14]

Children with hearing loss are diagnosed with ASD at a later age than their hearing peers.[15] As a result of the improved newborn hearing screening programs, hearing loss is diagnosed at an earlier stage. Autism symptoms may resemble the behavioral consequences of hearing loss. Meinzen Derr et al. reported that the average age of ASD diagnosis in children with hearing loss is approximately 66.5 months (around 5-6 years).[16] In children with normal hearing, the average age of ASD diagnosis is approximately 38 months.[17] Therefore, it is essential for a professional to screen and assess the early signs of ASD in children with hearing loss. Early intervention in children with ASD may facilitate a faster compensation for challenges. In contrast, a late diagnosis of autism can lead to delays in compensating for existing challenges and delays in all developmental stages. Identifying ASD symptoms accompanying hearing loss is crucial for the success of auditory rehabilitation in individuals with hearing loss.

The aim of this study was to assess ASD symptoms in children with severe to profound hearing loss using the Autism Spectrum Screening Questionnaire (ASSQ-TR).

MATERIAL AND METHODS

The Ethics Committee of Ankara Yıldırım Beyazit University (date: April 22, 2024; no: 696) approved this study. The study was conducted in accordance with the ethical standards of the institutional and national research committee and with the 1964 Declaration of Helsinki and its later amendments or comparable ethical standards. Each child's' parent signed an informed consent form indicating that participation was voluntary and that they knew they could withdraw at any time.

PARTICIPANTS

A total of 32 individuals participated in this study online via Google Forms (Google LLC, USA) in May 2024. The study included children aged 6-9 years with severe-to- profound prelingual hearing loss in at least 1 ear, as determined by pure tone audiometry conducted within the last 6 months, and who had no other diagnosed disabilities. None of the children or their parents who participated in the study used sign language. The parents of the children with hearing loss had normal hearing. The participants consisted of children who either used auditory amplification/auditory implants or did not use any auditory devices. The children's' age, gender, the type of auditory device for each ear, and whether they had any additional diagnosed disabilities were collected. Additionally, the age at which the children started using hearing devices was asked.

AUTISM SPECTRUM SCREENING QUESTIONNAIRE

ASSQ-TR, which has been validated and tested for reliability in Turkish by Köse et al. was used in this study.[18] The ASSQ is a 27-item screening tool developed by Ehlers and Gillberg.[19] The scale consists of 11 items assessing social interaction, 6 items evaluating communication problems, 5 items addressing restricted and repetitive behaviors, and 5 items focusing on motor functions and motor-vocal tics. The ASSQ-TR is a 3-point Likert-type scale scored as "No" (0 points=normal), "Somewhat" (1 point=some abnormality), or "Yes" (2 points=definite abnormality). The scale was designed for children aged 6 to 18 years. In the Turkish version, a cutoff score of 16 was determined to be optimal for Pervasive Developmental Disorders (PDD). Under the Diagnostic and Statistical Manual of Mental Disorders–IV (DSM-IV) criteria, the umbrella term PDD includes autism disorder, asperger's disorder, childhood disintegrative disorder, Rett syndrome, and PDD not otherwise specified. However, in the DSM-5, all of these disorders are consolidated under the name ASD.

For the cutoff score of 16, the sensitivity was 94.1%, specificity was 89.0%, and diagnostic accuracy was 90.7% (14). Permission to use the scale was obtained from the corresponding author of the article. The ASSQ-TR is completed by the parents. Hattori et al. categorized the ASSQ into 3 domains: restricted and repetitive behaviors (items 2, 3, 9, 10, 18, 20, 21, 22, 23, 24, 27), social interaction (items 1, 12, 14, 15, 16, 17, 19, 25, 26), and communication problems (items 4, 5, 6, 7, 8, 11, 13).[20] In this study, ASD symptoms in children with hearing loss were screened using the ASSQ-TR and classified according to Hattori et al.[20]

STATISTICAL ANALYSIS

Statistical analyses were performed using SPSS version 25.0 (IBM Corporation, Armonk, NY). The distribution of the numerical data was examined using the Shapiro-Wilk test. Descriptive statistics are presented as mean±standard deviation or median (1st quartile to 3rd quartile). The Mann-Whitney U test was used for comparisons between the 2 independent groups. Qualitative variables are presented as frequencies and percentages. The Spearman's rho test was used in the correlation analysis. Categorical variables are presented as frequencies and percentages. Spearman's rho test was used for the correlation analysis. Additionally, p values less than 0.05 were considered statistically significant.

RESULTS

A total of 32 children with profound and severe hearing loss participated in our study. The mean age of the children was 7.72 (±1.19) years. 59.4% (n=19) of the participants used a bilateral auditory device, and 12.5% (n=13) used a unilateral auditory device. The mean duration of experience with the auditory devices was 5.76 (±1.85) years. The children's average age of starting to use hearing devices was 1.95

(±1.31) years. Demographic information and details regarding auditory device use are presented in Table 1 and Table 2.

The ASSQ-TR was completed by the parents of all participants. The mean score of all participants was 7.56 (±9.53), with the subscore for restricted and repetitive behaviors being 2.31 (±4.19), the subscore for social interaction being 2.66 (±3.57), and the subscore for communication problems being 2.59 (±2.66). However, 5 participants scored above the designated cutoff score of 16, indicating the characteristics of ASD (Figure 1). The mean score for these five children was 26.2 (±9.83). Notably, the mean age of these 5 children was 7.20 (±1.30) years, and all of them were male.

When participants were grouped by gender, no statistically significant differences were found in the total scores or subscale scores (p=0.584, Z=0.558). However, it is noteworthy that all 5 participants with higher scores were male. Analysis based on the direction of auditory device use (bilateral vs. unilateral) also revealed no significant difference in the total scores between the groups (p=0.623, Z=0.520). The 5 participants who exceeded the cutoff score included three unilateral auditory device users and 2 bilateral auditory device users.

Correlation analyses showed a statistically non-significant, weak negative correlation between age and total score of the participants (p=0.282 r_s=-0.196). There was a statistically significant weak negative correlation between the total score and duration of auditory device use (p=0.040, r_s=-0.370).

When assessing individual scale items, more "Yes" responses were noted for questions such as "Understands the literal and concrete use of language rather than its metaphorical and vague use", "Lacks empathy skills", and "Lacks common sense", compared to other questions in children with hearing loss.

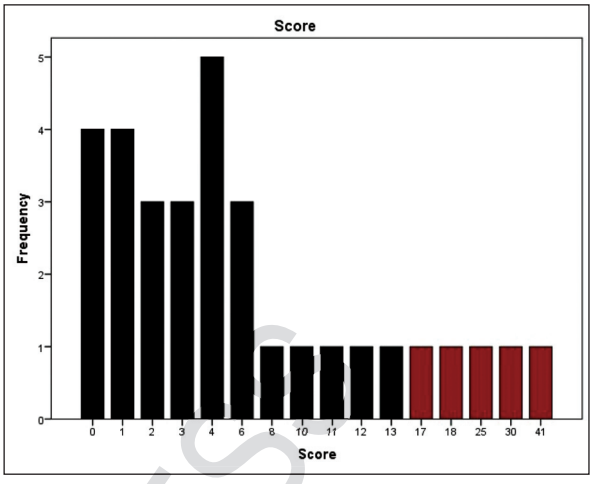

FIGURE 1: Frequency distribution of children's ASSQ-TR Scores
The red bars represent the five children who exceeded the cutoff score.

TABLE 1: Demographic characteristics of participants

	Gender	
	Female	Male
n (%)	11 (34.4%)	21 (65.6%)
Age	8.09 (±1.04)	7.52 (±1.25)

Data are summarized as mean±standard deviation.

TABLE 2: Participant information regarding device usage

Auditory device lateralityn (%)		Auditory device type n (%)	
Bilateral	19 (59.4%)	None	13 (20.3%)
		Hearing aid	13 (20.3%)
		Cochlear implant	37 (57.8%)
Unilateral	13 (12.5%)	Brainstem implant	1 (1.6%)
Total	32 (100%)	Total	64 (100%)

DISCUSSION

The effects of hearing loss vary depending on its severity. Therefore, the study group consists solely of children with severe to profound hearing loss. Due to the similarity between certain findings observed in children with hearing loss and those associated with ASD, the diagnosis of ASD is often delayed, typically around the ages of 5-6.[16] This study aimed to assess ASD-related findings in children with hearing loss. The study group consists of a specific population aged 6-9 years, which limits the number of participants. This study serves as a preliminary investigation for larger-scale research and provides sufficient findings to evaluate the methodology used.

In our study, a decrease in ASSQ-TR scores was observed as the duration of auditory stimulation with the auditory device increased for the participating children. Early use of auditory devices in children

with hearing loss may reduce the risk of exhibiting autism-related symptoms.

Out of the 32 children who participated in the study, 5 exceeded the cutoff score on the ASSQ-TR scale. Research conducted on a large scale has revealed that the prevalence of autism is 2-5 times higher in boys than in girls.[21] In this study, the 5 children with hearing loss who exceeded the threshold score were all male. This situation is explained by the "Female Protective Effect" model, which suggests that girls have a lower genetic risk of autism.[22] Additionally, females may present autism in a different phenotype compared with males[23] Diagnostic and screening tools have been developed based on the autism criteria established primarily from a male-dominated population.[23] Therefore, diagnostic and screening tools may be limited in identifying the subtle differences in the autism phenotype between females and males.[23]

Studies have reported that the diagnosis of ASD in children with hearing loss is delayed by 3 years compared to their typically hearing peers.[16,17] Typically, hearing children are diagnosed with ASD around the age of 3, while children with hearing loss receive their diagnosis around the age of 5-6.[16,17] The reason for this is the shared symptoms observed in both children with autism and children with hearing loss, such as reduced eye contact, diminished interaction, difficulty with joint attention, lack of response to their name, abnormal prosody usage, difficulty in understanding and using gestures and facial expressions, delayed language acquisition, and poor coordination skills.[11] The mean age of the 5 children showing ASD-related symptoms in our study was 7.20 (±1.30). The fact that these children have not been suspected of having autism until now may be due to the behavioral outcomes caused by hearing loss masking the signs of autism.

Abnormal findings in the auditory system of individuals with autism are revealed in electrophysiological studies in the literature.[24] Gillberg et al. assessed auditory brainstem responses (ABR) in children with autism and found abnormal ABR findings in 13.3% of the autistic children.[25] Klin found abnormal auditory function of varying types and degrees in 33-46% of individuals with autism.[26] According to the ABR results in individuals with autism, the prolongation of I-V interpeak latencies is the most common finding.[27] Cortical studies revealed that the P300 amplitude is abnormally reduced in individuals with autism compared to controls, which is associated with a deficiency in allocating attention to new stimuli.[24] Mismatch negativity studies showed that shorter latencies and increased amplitudes in individuals with autism indicate a faster detection of changes in auditory inputs. This may explain the heightened sensitivity of individuals with autism to acoustic changes.[24] The abnormalities observed in the Auditory Evoked Potentials, P300, and Mismatch Negativity tests in individuals with autism may result from myelination defects in the central nervous system, extending from the brainstem to the cortical-subcortical structures.[24,28]

In addition to the statistical analysis, the questions of the ASSQ-TR scale were evaluated individually. Based on the responses from the scale we used, we observed that children with hearing loss experience difficulties in understanding common sense and the figurative use of language. The literature includes studies showing that children with hearing loss have lower levels of empathy skills and understanding of figurative language compared to typically hearing children.[29-31] The limitations in these skills may lead to delays in social interaction abilities, restricted emotional connections, and communication difficulties in children with hearing loss. Therefore, the key characteristics of autism, such as lack of empathy, limitations in social interaction and communication, and the use of figurative language, may be overshadowed by hearing loss. If autism is diagnosed first, the signs of hearing loss may be overshadowed by autism. This may delay the diagnosis of hearing loss and negatively impact the individual's' developmental areas. Therefore, it is crucial to establish an appropriate early intervention program, considering that children with both diagnoses may exhibit overlapping symptoms.

Children with hearing loss and those diagnosed with ASD share common sensory sensitivities.[32,33] Therefore, a multidisciplinary approach is recommended in the assessment and therapy processes for

children diagnosed with both hearing loss and ASD. The auditory rehabilitation process for children with hearing loss and ASD can be more challenging compared with that for children without ASD.[34] Due to their tactile and auditory sensitivities, as well as difficulties adapting to unfamiliar environments, these children may refuse to undergo hearing tests or wear hearing aids.[35] Collaboration among the family, audiologist, occupational therapist and psychiatrist is essential during both the hearing assessment and the hearing aid fitting process.[15] Children with hearing loss and ASD should be given time to become familiar with the environment and the devices, and a hurried approach should be avoided. If behavioral test results are considered unreliable, hearing aid settings should be based on objective test findings.[34] Due to their auditory sensitivities, the maximum output level of the hearing aid must be adjusted carefully.[34] Therefore, it is important that the audiologist conducting the application is knowledgeable and aware of ASD-related considerations.

Although there are more studies investigating auditory functions in individuals with ASD, the limited number of studies assessing ASD symptoms in individuals with hearing loss is a strength of our study. This situation can be explained by the overlap of symptoms between hearing loss and ASD, the lack of sufficient knowledge among clinicians regarding both disorders, and the limitations of the assessment methods.[15] The small sample size is a limitation of the study. This may limit the generalizability and affect the overall validity of the results. Another limitation was the absence of objective hearing tests performed on the participants. It is recommended that highly sensitive ASD screening scales specific to individuals with hearing loss be developed in the future.

CONCLUSION

This study assessed ASD symptoms in children aged 6-9 years with severe to profound hearing loss. Among the 32 children who participated in the study, 5 exhibited autism-related symptoms according to the ASSQ-TR scale. It is noteworthy that these children, who had passed early childhood, were not previously evaluated for autism. A high score on the ASSQ-TR does not necessarily indicate that a child has autism, and further evaluation is needed for an ASD diagnosis. The presence of similar findings in both ASD and hearing loss may delay the diagnosis and intervention of autism in children with hearing loss. Conversely, the overlap of symptoms may also lead to a delayed diagnosis of hearing loss in children with ASD. The overlap of the symptoms of hearing loss and ASD can overshadow the diagnoses, negatively affecting the intervention for individuals with either diagnosis. Assessment methods should be expanded, and professionals should be more attentive to children with either disorder.

Source of Finance

During this study, no financial or spiritual support was received neither from any pharmaceutical company that has a direct connection with the research subject, nor from a company that provides or produces medical instruments and materials which may negatively affect the evaluation process of this study.

Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: *Banu Baş, Gülse Akdemir, Elif Zehra Biber, Rukiye Çolak Sivri;* ***Design:*** *Banu Baş, Gülse Akdemir, Elif Zehra Biber;* ***Control/Supervision:*** *Banu Baş, Elif Zehra Biber;* ***Data Collection and/or Processing:*** *Gülse Akdemir, Elif Zehra Biber;* ***Analysis and/or Interpretation:*** *Gülse Akdemir, Elif Zehra Biber;* ***Literature Review:*** *Gülse Akdemir, Elif Zehra Biber;* ***Writing the Article:*** *Gülse Akdemir, Elif Zehra Biber;* ***Critical Review:*** *Banu Baş, Rukiye Çolak Sivri;* ***References and Fundings:*** *Banu Baş, Rukiye Çolak Sivri;* ***Materials:*** *Banu Baş, Gülse Akdemir, Elif Zehra Biber, Rukiye Çolak Sivri.*

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