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Increased rate of listening difficulties in autistic children

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ABSTRACT

Introduction: Auditory challenges are both common and disruptive for autistic children and evidence suggests that listening difficulties may be linked to academic underachievement (Ashburner, Ziviani & Rodger, 2008). Such deficits may also contribute to issues with attention, behavior, and communication (Ashburner et al., 2008; Riccio, Cohen, Garrison & Smith, 2005). The present study aims to summarize the auditory challenges of autistic children with normal pure-tone hearing thresholds, and perceived listening difficulties, seen at auditory-ASD clinics in the US and Australia.

Methods: Data were compiled on a comprehensive, auditory-focused test battery in a large clinical sample of school-age autistic children with normal pure-tone hearing to date ($N = 71$, 6–14 years). Measures included a parent-reported auditory sensory processing questionnaire and tests of speech recognition in noise, binaural integration, attention, auditory memory and listening comprehension. Individual test performance was compared to normative data from children with no listening difficulties.

Results: Over 40% of patients exhibited significantly reduced speech recognition in noise and abnormal dichotic integration that were not attributed to deficits in attention. The majority of patients (86%) performed abnormally on at least one auditory measure, suggesting that functional auditory issues can exist in autistic patients despite normal pure-tone sensitivity.

Conclusion: Including functional listening measures during audiological evaluations may improve clinicians' ability to detect and manage the auditory challenges impacting this population. **Learner Outcomes:** 1) Readers will be able to describe the auditory difficulties experienced by some autistic patients (ASD). 2) Readers will be able to describe clinical measures potentially useful for detecting listening difficulties in high-functioning autistic children.

1. Introduction

Autism spectrum disorder (ASD) is a neurodevelopmental disorder that often results in sensory processing abnormalities

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(American Psychiatric Association [APA], 2013; Diagnostic Statistical Manual Version V [DSM-V]). Although there is no universal consensus on the use of people-first versus identity-first language, identity-first language is used herein based on the recent cited preferences of the autistic community (Bury, Jellett, Spoor & Hedley, 2020; Gernsbacher, 2017).

Auditory challenges are among the most commonly-reported sensory processing issues affecting autistic children, and there is substantial evidence of these difficulties from subjective questionnaires, clinical assessments and objective tests (Adamson, O'Hare & Graham, 2006; Alcántara, Weisblatt, Moore & Bolton, 2004; Baker, Lane, Angley & Young, 2008; Cui, Wang, Liu & Zhang, 2017; Gopal et al., 2021; Haesen, Boets & Wagemans, 2011; Keary et al., 2009; Kulesza & Mangunay, 2008; Lukose, Brown, Barber & Kulesza, 2013; O'connor, 2012; Rogers, Hepburn & Wehner, 2003; Vlaskamp et al., 2017). Auditory challenges include auditory filtering, speech recognition and comprehension, auditory attention and memory, and binaural integration (Ashburner et al., 2008; Rance, Chisari, Saunders & Rault, 2017, 2014; Schafer et al., 2013, 2016, 2020; Tomcheck & Dunn, 2007). Given the breadth of auditory challenges in autistic children, more comprehensive audiologic assessments may be needed to effectively detect and manage these issues.

1.1. *Sensory processing – auditory filtering*

Auditory filtering refers to the ability to detect, discriminate, and respond to auditory information in the presence of noise, suggesting listening difficulties may be linked to academic underachievement in some autistic children (Ashburner et al., 2008). In a study of caregivers of children with ASD, 75% of respondents identified auditory filtering as their child's most abnormal sensory processing characteristic on the Short Sensory Profile (SSP; Ashburner et al., 2008; McIntosh, Miller, Shyu & Dunn, 1999). Increased auditory filtering abnormalities (93%) have also been reported by others (Lane, Young, Baker & Angley, 2010). These deficits can exist despite normal pure-tone hearing sensitivity and may reflect brain-based differences in speech processing Gopal et al., 2021; Haesen et al., 2011; Russo, Zecker, Trommer, Chen & Kraus, 2009).

1.2. *Speech recognition and listening comprehension*

Autistic children exhibit significantly poorer open-set speech recognition in noise and higher caregiver- and self-reported listening difficulty in noise than neurotypical peers (Alcántara et al., 2004; Rance et al., 2014; 2017; Schafer et al., 2013, 2020). Given the documented speech recognition-in-noise issues, listening comprehension problems may also arise, which have been previously reported in children with neurotypical functioning (Valente, Plevinsky, Franco, Heinrichs-Graham & Lewis, 2012) and autism (Schafer et al., 2020).

1.3. *Auditory attention and memory*

Many autistic children exhibit significant deficits in visual and auditory attention and co-existing diagnoses of attention deficit (ADD) or attention deficit hyperactivity disorder (ADHD) (Corbett & Constantine, 2006; Jang et al., 2013; Sturm, Fernell & Gillberg, 2004). In addition, lower auditory working memory scores have been reported in autistic children relative to neurotypical peers in combination with deficits in binaural integration and speech recognition in noise (Schafer et al., 2020). As a result, difficulties with auditory attention and memory may contribute to the auditory challenges in this population.

1.4. *Binaural integration*

Another auditory challenge in individuals with ASD is binaural integration, or the ability to combine different auditory stimuli simultaneously presented to the two ears (i.e., dichotic stimuli). On dichotic tests examining binaural integration, pronounced deficits have been observed in autistic individuals (Kozou, Azouz, Abdou & Shaltout, 2018; Schafer et al., 2020). As stated previously, these binaural integration deficits have been identified in combination with other auditory challenges including degraded auditory filtering, speech recognition in noise, listening comprehension, and working memory (Schafer et al., 2020).

1.5. *Study aim and rationale*

Despite the evidence of multiple auditory challenges in autistic children, audiological assessments of these patients generally focus on standard audiometric measures (e.g., pure-tone threshold detection). These basic assessments are insensitive to common patient- and parent-reported auditory challenges (e.g., speech recognition in noise, auditory filtering). In a multisite, international effort, audiologists in the United States and Australia have been comprehensively assessing and managing the auditory challenges of autistic children in a coordinated fashion. The present study aims to summarize the auditory challenges of autistic children on a comprehensive test battery delivered at these dedicated ASD-audiology clinics.

Retrospective analyses were used to report auditory function in a large group of autistic patients who were seen clinically due to listening concerns. Descriptive statistics were used to define the characteristics of this sample, and statistical analyses were performed on functional and clinical measures to document auditory challenges. These results extend findings from previous studies and highlight the need for more holistic assessment of autistic patients.

2. Methods

2.1. Research design and ethics

The present study used a single-group, retrospective design. De-identified data were mined from three clinics that served school-age autistic children. Two were located in the United States (Oklahoma, Texas) and one was in Australia (Melbourne). Given the retrospective nature of this study, it qualified for an exemption from the University of North Texas Institutional Review Board (UNT IRB); Hearts for Hearing (Oklahoma, Texas) was included as a test site on the UNT IRB application. Ethical approval for analyzing data from the Melbourne, Australia site was obtained through The University of Melbourne Ethics Research Committee (ID: 2,056,646.1).

2.2. Patients

Clinical data were analyzed from seventy-one patients (17 females) between 6 and 14 years of age (mean age at initial visit = 9.3, SD = 2.1 years) with a positive history of ASD and normal pure-tone hearing sensitivity bilaterally ($\leq$ 20 dB HL at octave frequencies from 250 to 8000 Hz). Tympanometry ($n = 50$) revealed normal middle ear function in 92.0% of patients, with the remaining presenting with reduced compliance (2.0%) or negative middle ear pressure (6.0%). All patients had a positive history of self- or caregiver-reported auditory difficulties as documented on case history forms. Data were excluded from children with known intellectual impairment, those lacking adequate verbal language skills (i.e., unable to repeat test stimuli), and non-native English speakers.

Documentation of ASD diagnoses were determined from external reports provided by parents or caregivers. Independent corroboration of previously established ASD diagnoses was beyond the scope of the current study. According to outside reports, ASD diagnoses were established based on DSM-IV or DSM-V criteria using one or more of the following tools: Autism Diagnosis Interview – Revised (ADI-R) (Tadevosyan-Leyfer et al., 2003), Autism Diagnostic Observation Schedule – Second edition (ADOS-2) (Lord et al., 2012), Childhood Autism Rating Scale – Second edition (CARS-2) (Schopler, Van Bourgondien, Wellman & Love, 2010), Behavior Assessment System for Children- 3rd Edition (BASC-3) Parent Rating Scale and Teacher Rating Scales (Reynolds & Kamphaus, 2015), Connor's 3 Teacher and Parent Reports (Conners, 2008) and the Vineland Adaptive Behavior Scales- 3rd Edition (Vineland- 3) Teacher Report Form (Sparrow, Cicchetti & Saulnier, 2016). Additional tools to support the ASD diagnoses included parent interview, health and developmental history, patient observations, and screening questionnaires for both home and educational settings. Diagnoses were established by a primary care provider such as a developmental pediatrician with input from other health professionals including psychologists, speech-language pathologists, and occupational therapists. Median age at time of ASD diagnosis was 5.6 years (SD = 2.4) and ranged from 2.0 to 12.0 years.

2.3. Facilities and equipment

Clinical testing was completed in double-walled sound booths with calibrated equipment. Tympanometry was performed using GSI 39 (Texas site), GSI TympStar Pro (Oklahoma Site) and GSI Tympstar (Melbourne site) machines. Separate-ear, pure-tone testing was completed using insert earphones (ER-3A) (US sites), supra-aural headphones (TDH-50) (US sites), or Telephonics TDH-39P audiometric headphones (Australian site) and clinical audiometers (GSI 61, US sites; Interacoustics Affinity 2, Australian site). Other recorded stimuli were presented via the earphones specified above, Sennheiser HD 215 earphones (Australian site) or loudspeakers (Grayson-Stadler Standard; Sony CFD-ZW755). Recorded stimuli were played from clinical audiometers or CD players (Sony 5-CD Changer, Sony CD-Radio-Cassette-Corder CFD-ZW755) (US sites) (Buddee CD player BD903208) (Australian site).

2.4. Tests and procedures

The clinical measures used in this study were selected based on the published literature describing auditory performance in autistic children with normal sound detection thresholds and tests used to document benefit from evidence-based auditory interventions (Rance et al., 2014, 2017; Schafer et al., 2013, 2016, 2019, 2020).

Clinical assessments were comprised of two sessions, each lasting approximately 1.5 h. Breaks were offered as needed throughout the sessions. Sample sizes vary across measures due to appointment time constraints (i.e., insufficient time to complete all measures) and patient factors (i.e., task too difficult or mastery could not be demonstrated on practice items). Caregiver forms, including the history form and Short Sensory Profile 2, were completed while patients were performing other measures. In visit one, tests were completed in the following order: tympanometry, hearing screening, speech recognition-in-noise, and dichotic testing. In visit 2, measures of short-term and working memory, listening comprehension, attention and cognitive ability were administered. Attention and cognitive tasks were reserved for the end of the appointment because they were time-consuming tests and many participants had external test results for these measures.

2.4.1. History form

Patient health history and demographic information were obtained via a case history questionnaire that was completed by caregivers during the initial appointment. Questions included, but were not limited to, general medical and audiological health history information (e.g., diagnoses, history of middle ear problems), growth and development (e.g., regular therapies), and educational status (e.g., school placement, special education services). Information regarding cognitive ability and ASD diagnoses were obtained from external clinical reports submitted by caregivers.

2.4.2. Short sensory profile 2

The Short Sensory Profile 2 (SSP2; Dunn, 2014) is a standardized survey evaluating children's sensory processing in various environments including home, school and community along four domains (i.e., seeking, avoiding, sensitivity and registration). Although caregivers were instructed to complete the full survey, responses were exclusively analyzed on the "Sensitivity/Sensor" domain because it contained the most auditory relevant questions that best reflected auditory filtering ability (e.g., "is distracted when there is a lot of noise around"). The total raw score for the "Sensitivity/Sensor" quadrant was calculated for each patient and compared with normative data in a sample of children without disabilities ($n = 697$) (Dunn, 2014). Based on a bell curve normed distribution, the raw score total for each quadrant can be classified as "much less than others" (lower 2%), "less than others" (between 1 SD and 2 SD below the mean, accounting for 14% of the normative sample), "just like the majority of others" (± 1 SD from the mean and accounting for 68% of the normative sample), "more than others" (between 1 SD and 2 SD above the mean), and "much more than others" (upper 2%). Patient scores falling two or more standard deviations above the mean for the "Sensitivity/Sensor" domain represented a "definite difference" from average (Simpson, Adams, Alston-Knox, Heussler & Keen, 2019) and were classified as "outside normal limits."

2.4.3. Speech recognition in noise

The Bamford-Kowal-Bench Speech-in-Noise test (BKB-SIN; Etymotic Research, 2005) is a recorded (CD), open-set sentence recognition test that was presented under headphones at a level of 60 dBA (i.e., 47 dB HL). Target sentences are produced by a male talker and embedded in multitalker babble at pre-recorded signal-to-noise (SNR) ratios varying in 3-dB steps from +21 to −6 dB SNR. Patients were instructed to repeat each target sentence. Scores were compared to age-based normative data in children with normal sound detection thresholds (Etymotic Research, 2005), and as per user guide data, patient scores falling one or more standard deviations below the mean were regarded as "outside normal limits".

The Listening in Spatialized Noise – Sentences (LiSN-S; Cameron & Dillon, 2007) is an open-set test, computerized sentence recognition test producing a three-dimensional auditory environment under specialized circumaural headphones (Sennheiser HD 215). Target sentences are produced by a female talker and are adaptively varied in signal level while distractor speech, comprised of stories spoken by three female talkers, is presented at a fixed level of 55 dB HL. This test yields a 50% correct speech-in-noise threshold for target words. The 'high-cue' condition of this test was administered in this study as it best represents a real-world listening task where spatial separation is expected between competing talkers. This condition spatially separates the target speaker from different-sounding background voices by 90°. Patients were instructed to repeat each target sentence, and patient scores of two or more standard deviations below the mean adjusted for age, were regarded as "outside normal limits", as proposed by (Cameron and Dillon, 2007).

2.4.4. Dichotic digits test (DDT)

The Dichotic Digits Test (DDT; Musiek, 1983) assesses the dichotic listening skill of binaural integration, which reflects an individual's ability to perceive multiple auditory inputs (i.e., two different digits presented in sequence) presented simultaneously at each ear separately. Target digits spoken by a male talker are presented at 55 dB HL, and patients are instructed to repeat back all digits in a free recall manner (i.e., order not scored). Patient scores falling two or more standard deviations below the mean for one or both ears were regarded as "outside normal limits" (Tomlin, Dillon & Kelly, 2014). Significant interaural asymmetry was individually defined as a left and right ear score difference of greater than 20% as proposed by Tomlin et al. (2014).

2.4.5. Integrated visual and auditory quick screen continuous performance task (IVA-QS)

The Integrated Visual and Auditory Quick Screen Continuous Performance Task (IVA-QS CPT; Sandford & Anton, 2014) is a computer-based test designed to assess auditory and visual attention and impulse control. The IVA-QS presents a series of 1′s and 2′s in a pseudo-random combination of visual and auditory stimuli. Patients are instructed to click a mouse when a "1″ is seen or heard, but not respond to "2″. Auditory stimuli were presented under headphones (Sennheiser HD215) at a volume adjusted to user comfort prior to the start of the test. Full-scale attention scores, comprised of auditory and visual subscales, were analyzed from the IVA-QS test and compared with age-matched normative data (Sandford & Anton, 2014). Standard scores of 70 and below, two or more standard deviations below the mean, were classified as "extremely impaired" by the test developers and were regarded as "outside normal limits".

2.4.6. Auditory short-term and working memory

Two subtests from the Test of Auditory Processing Skills (TAPS-4; Martin, Brownell & Hamaguchi, 2018), number memory forward and number memory reverse, were used to assess patient's short-term and working memory, respectively. Patients were instructed to repeat back numbers presented in a monitored live voice, open-set format. Task difficulty is titrated by length of number string. Scores were compared to age-related normative data ($N = 2023$) (Martin et al., 2018), and scaled scores of seven or lower, one or more standard deviations below the mean, were defined as "outside normal limits".

2.4.7. Listening comprehension

Two subtests from the Test of Auditory Processing Skills (4th edition) (TAPS-4; Martin et al., 2018), Processing Oral Directions and Auditory Figure-Ground, were used to assess patient's auditory comprehension ability in quiet and in the presence of noise, respectively. These subtests were selected because they have different acoustic backgrounds (i.e., quiet and noise), and previous research documented deficits in both (Schafer et al., 2020). Auditory stimuli were presented from a CD player through speakers at a volume adjusted to user comfort prior to the commencement of the test. Each child was seated near the examiner and responded verbally to each presented stimulus. Scores were compared with age-related normative data ($N = 2023$) (Martin et al., 2018), and scaled scores of seven and below, one or more standard deviations below the mean, were defined as “outside normal limits”.

2.4.8. *Cognitive evaluation*

Caregivers denied any known history of cognitive impairment in their children. Cognitive functioning was confirmed to be in the average or above-average range in 38 patients based on outside reports that used various tests including the Wechsler Intelligence Scale for Children (4th and 5th editions) (WISC-IV; Wechsler, 2003 WISC-V; Wechsler, 2014) ($n = 17$), Wechsler Preschool & Primary Scale of Intelligence (4th edition) (WPPSI-IV; Wechsler, 2012) ($n = 18$), or an unspecified intelligence scale ($n = 3$). Cognitive functioning was assessed in an additional three patients during their second clinical appointment using the Test of Nonverbal Intelligence, fourth edition (TONI-4) (Brown, Shecbenou & Johnsen, 2010). Standard scores of 70 or below, two or more standard deviations below the mean, were regarded as “outside normal limits”. In 30 patients, formal cognitive evaluation reports were not provided, but all these patients showed the ability to communicate in a conversational manner, comprehend instructions and reliably respond during testing. Additional analyses were conducted to determine if this missing cognitive data could have influenced the analyses of other data obtained from the participating patients.

2.5. *Data analysis*

De-identified clinical data were mined from each site, entered into a spreadsheet, and stored on a password-protected computer. Data were analyzed using descriptive statistics and appropriate parametric tests using R software (R core team, 2021).

Individual patient scores were compared to published normative data from children without reported listening difficulties and the proportion of fails (i.e., frequency of performance outside normal limits over total observations) was determined for each test. Pearson correlations were performed on tests to evaluate the relationship between measures and agreement was categorically classified according to Landis and Koch (1977). McNemar’s chi-square test was used to determine the degree of agreement (i.e., fail rates) across measures.

To test the potential influence of missing intelligence quotient (IQ) scores on auditory test performance, patients were subdivided into groups, “IQ normal” versus “IQ unknown”. Potential differences between groups were assessed over the battery of tests using Welch’s two-sample t-tests for continuous variables and Pearson chi-squared tests for categorical variables. The goal was to confirm that patients missing the IQ variable performed similarly on the battery of tests to the group with confirmed normal IQ scores.

A conservative significance criterion of 0.01 was applied for all tests to control for the probability of false positive (Type I) errors.

Table 1
Observed scores from autistic patients and expected scores from neurotypical peers.

Name of test:	Sample size (n)	Mean score +/- SD	Score range (min, max)	Minimum score required to score WNL	Observed proportion scores ONL	Expected proportion scores ONL	Observed vs. expected score (p value)
SSP2 SS Raw score	43	35.58 (6.94)	14.0–46.0	31.0 (<2SD above mean)	0.79	0.025	<0.001
BKB-SIN Z score	63	−1.19 (1.68)	−6.8–1.5	−0.9 (<1SD below mean)	0.54	0.159	<0.001
LiSN-S High Cue Z score	64	−1.84 (1.43)	−5.7–1.3	−1.9 (<2SD below mean)	0.42	0.025	<0.001
DDT	27	−2.27 (1.60)	(−7.1–0.3)	−1.9 (<2SD below mean)	0.41	0.025	<0.001
IVA-QS FS Att Standard score	53	80.31 (25.52)	19.0–119.0	71.0 (<2SD below mean)	0.34	0.025	<0.001
List Comp Q Scaled score	45	9.85 (3.41)	1.0–19.0	8.0 (<1SD below mean)	0.14	0.159	0.737
List Comp N Scaled score	45	9.60 (3.07)	5.0–19.0	8.0 (<1SD below mean)	0.13	0.159	0.838
Mem F Scaled score	67	11.16 (2.73)	5.0–16.0	8.0 (<1SD below mean)	0.03	0.159	0.001
Mem B Scaled score	67	11.37 (2.84)	6.0–19.0	8.0 (<1SD below mean)	0.02	0.159	<0.001

Note. BKB-SIN = Bamford-Kowal-Bench Speech-in-Noise test; DDT= Dichotic Digits test; IVA-QS FS Att = Integrated Visual and Auditory Quick Screen-Full scale Attention subscore; LiSN-*S* = Listening in Spatialized Noise-Sentences test; List Comp *Q* = Listening Comprehension-in-quiet test; List Comp *N* = Listening Comprehension-in-noise test; Mem *F* = Number Memory Forward test; Mem *B* = Number Memory Backward test; SSP2 SS = Short Sensory Profile 2 Sensitivity/Sensor subscale.

3. Results

3.1. Descriptive statistics on patient sample

The available clinical data were analyzed in 71 patients (males $n = 54$, 76.1%) between 6 and 14 years of age (Mean = 9.3; SD = 2.1) with a mean age of ASD diagnosis of 5.6 years (SD = 2.4; Range = 2.0–12.0). According to the case history forms ($n = 55$), patients were educated in mainstream classrooms ($n = 49$, 89.1%), special schools ($n = 4$, 7.3%), inclusive educational settings ($n = 1$, 1.8%) or home schooled ($n = 1$, 1.8%). At the time of the initial assessment, many patients were regularly engaging in therapies from other allied health disciplines including occupational therapy ($n = 41$, 74.5%), speech and language therapy ($n = 37$, 67.3%), psychology ($n = 27$, 49.1%) and social skills training ($n = 11$, 20.4%).

The most frequently reported co-occurring conditions were aversion to noise or abnormal sensitivity to loud sounds ($n = 56$, 73.2%); attention issues (i.e., attention deficit hyperactivity disorder [ADHD], attention deficit disorder [ADD]) ($n = 59$, 49.2%); and recurrent otitis media ($n = 52$, 34.6%).

3.2. Statistical analyses of test results

Table 1 and Fig. 1 summarize group scores and results from binomial tests used to determine the statistical significance of deviations from expected theoretical distributions for each test. Auditory performance on each test was compared to published normative data from neurotypical children without reported listening issues, and the proportion of abnormal scores was computed. Autistic patients with reported listening issues performed significantly worse than expected compared to a theoretical neurotypical population on five of the nine measures and significantly better on the memory measures ($p < 0.01$, Fig. 1).

3.2.1. Sensory processing (SSP2)

Binomial testing showed that 79.1% of patients had scores outside the normal range on the “Sensitivity Sensor” subscale of the SSP2, more than thirty-one times the predicted rate in a theoretical population ($n = 43$, observed proportion of failures = 0.79, $p < 0.001$).

3.2.1.1. Speech recognition in noise. Speech-in noise scores were below normal limits in a significant proportion of patients on both the BKB-SIN and LiSN-S high-cue assessments. Abnormal scores on speech-in-noise tasks were observed at approximately three times the 16% expected failure rate for BKB-SIN ($n = 63$, observed proportion of failures = 0.54, $p < 0.001$) and fourteen times the 2.5%

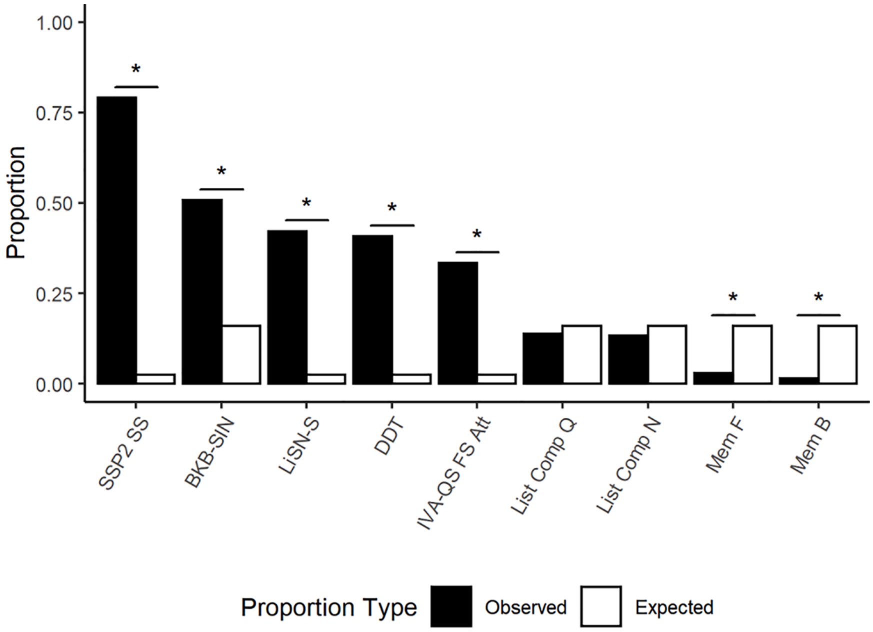

Fig. 1. Proportion of observed abnormal performance scores in autistic patients relative to expected proportions in an age-matched, neurotypical population. * = $p < 0.001$

Note. BKB-SIN = Bamford-Kowal-Bench speech-in-noise test; DDT = Dichotic digits test; IVA-QS FS Att = Integrated visual and auditory quick screen-full scale attention subscore; LiSN-S = Listening in spatialized noise-sentences test; List Comp Q = Listening comprehension-in-quiet test; List comp N = Listening comprehension-in-noise test; Mem F = Number memory forward test; Mem B = Number memory backward test; SSP2 SS = Short sensory profile 2 sensitivity/sensor subscale.

expected failure rate for LiSN-S ($n = 64$, observed proportion of failures = 0.42, $p < 0.001$) in theoretical populations.

3.2.1.2. Dichotic listening (DDT). Dichotic listening performance on the DDT was outside normal limits in 40.7% of patients compared to an estimated 2.5% failure rate in a theoretical population ($n = 27$, observed proportion of failures = 0.41, $p < 0.001$). Of the 11 patients who scored outside normal limits on the DDT task, three (27.3%) showed an atypical right-side dominance pattern, two (18.2%) showed a general weakness on both sides, and six (54.5%) showed a left-side dominance reversal pattern.

3.2.1.3. Attention (IVA-QS). Approximately one-third of patients had full-scale attention subscale scores outside the normal range on the IVA-QS test compared to an estimated 2.5% expected failure rate in a theoretical neurotypical population ($n = 53$, observed proportion of failures = 0.34, $p < 0.001$).

3.2.1.4. Auditory short-term and working memory. Only a small percentage of patients performed outside normal limits on digit-span tasks involving short-term and working-memory. In fact, patients scored significantly better than expected for a theoretical neurotypical population on short-term ($n = 67$, observed proportion of fails = 0.03, $p = 0.001$) and working memory ($n = 67$, observed proportion of fails = 0.02, $p < 0.001$).

3.2.1.5. Listening comprehension. The rate of abnormal performance on listening comprehension did not significantly differ from the expected fail rate in an age-matched, theoretical population in quiet ($n = 65$, observed proportion of failures = 0.14, $p = 0.74$) or noise ($n = 45$, observed proportion of failures = 0.13, $p = 0.84$).

3.2.2. Correlation analysis

Cross -test comparisons were performed exclusively on measures showing a greater than expected fail rate (i.e., SSP2, BKB-SIN, LiSN-S, DDT, IVA-QS) relative to theoretical neurotypical peers with no reported listening difficulties. Each of these five measures produced abnormal scores in more than 30% of patients, compared to less than 15% on each of the remaining four tests.

On the two speech recognition-in-noise measures (i.e., BKB-SIN and LiSN-S), the analyses indicated a significant positive correlation between performance ($r = 0.60$ $p < 0.001$) and moderate agreement between classification of performance as “normal” or “outside normal” (kappa = 0.48, 74% agreement). Also, no significant difference was found in classification of normal versus outside normal performance for BKB and LiSN-S ($X^2 = 1.56$, $p = 0.21$).

Additionally, DDT scores were significantly correlated with performance on the BKB-SIN ($r = 0.63$; $t = 3.87$, $p < 0.001$) and LiSN-S ($r = 0.56$; $t = 3.22$, $p = 0.004$) speech recognition tests. There was moderate agreement between classification of performance as “normal” or “outside normal” on DDT and BKB-SIN (kappa = 0.3953, 69% agreement) and DDT and LiSN-S (kappa = 0.37, 69% agreement). There was no significant difference in score classifications for BKB-SIN and DDT ($X^2 = 1.13$, $p = 0.29$) or LiSN-S and DDT ($X^2 = 0.13$, $p = 0.72$). This suggests that patients who performed abnormally on speech-in-noise tests were also likely to exhibit abnormal performance on dichotic integration tests, and vice versa.

There was no significant correlation ($p > 0.05$) between patient IVA-QS full scale attention score and performance on any of the other measures (SSP2: $r = 0.29$, $p = 0.14$; BKB-SIN, $r = 0.12$, $p = 0.43$; LiSN-S: $r = 0.17$, $p = 0.28$; DDT: $r = 0.08$, $p = 0.75$), suggesting that attention does not contribute to degraded performance on those measures. Furthermore, there was no significant correlation ($p > 0.05$) between SSP sensitivity/sensor scores and performance on the BKB-SIN ($r = -0.22$, $p = 0.90$), LiSN-S ($r = -0.004$, $p = 0.98$), or DDT ($r = 0.01$, $p = 0.96$).

3.2.3. Relationship between IQ and auditory performance

To ensure that there were no performance differences between patients with normal IQ scores ($n = 41$) and unknown IQ scores ($n = 30$), a Welch’s two-sample *t*-test was performed between groups with normal and unknown IQ scores on each test listed in Table 2. No

Table 2
Comparison of auditory performance in patients with normal versus unknown IQ scores.

Auditory test (Continuous)	Sample known/ Unknown IQ (n)	Mean for missing IQ group	Mean for normal IQ group	*t*-statistic	Degrees of freedom	*p*-value
SSP2 SS	27/16	35.50	35.63	−0.06	31.28	0.95
BKB-SIN	39/24	−1.00	−1.30	0.72	59.14	0.47
LiSN-S High cue	38/26	−2.02	−1.72	−0.81	52.76	0.42
IVA-QS	27/18	82.83	78.63	0.56	41.84	0.58
Memory F	41/26	11.35	11.05	0.42	48.12	0.68
Memory B	40/27	11.37	11.38	−0.01	52.85	1.00
List Comp Q	38/27	10.26	9.55	0.78	44.58	0.44
List Comp N	29/16	10.25	9.24	1.16	39.60	0.25
DDT	17/9	−2.12	−2.35	−0.36	17.58	0.72

Note. BKB-SIN = Bamford-Kowal-Bench Speech-in-Noise test; IVA-QS FS Att = Integrated Visual and Auditory Quick Screen-Full scale Attention subscore; LiSN-*S* = Listening in Spatialized Noise-Sentences test; List Comp *Q* = Listening Comprehension-in-quiet test; List Comp *N* = Listening Comprehension-in-noise test; Mem *F* = Number Memory Forward test; Mem *B* = Number Memory Backward test; SSP2 SS = Short Sensory Profile 2 Sensitivity/Sensor subscale.

significant differences were found on any of the test scores by group.

4. Discussion

4.1. Summary of novel findings

The current retrospective study examined clinical data from three ASD-audiology clinics offering coordinated comprehensive auditory assessments that extend beyond the pure-tone audiogram. Data summarized in a large sample of autistic patients with perceived listening difficulties showed significant deficits on measures of auditory filtering, speech recognition in noise, binaural integration and attention. Fail rates (i.e., abnormal scores) on the aforementioned tests were significantly higher for the ASD patient group than a theoretical neurotypical population with no reported listening difficulties. In fact, the majority of patients in this study (86%) performed abnormally on at least one measure, despite enhanced performance on short-term and working memory tasks.

Further analyses evaluating relationships between performance on tests showed a positive correlation between measures of speech recognition in noise (LiSN-S and BKB-SIN), as well as between both tests of speech recognition in noise and binaural integration (DDT). The relationship between speech recognition-in-noise and binaural integration skills suggests that those with speech-in-noise difficulty also display poor dichotic listening ability and abnormal ear asymmetry (e.g., left-side dominance). The latter finding may reflect a reduced priority for speech and speech-like auditory signals (Lindell & Hudry, 2013) and brain-based differences in speech processing (Gopal et al., 2020; Haesen et al., 2011; Russo et al., 2009).

No relationship was observed between attention and patient performance on any auditory tests (i.e., BKB-SIN, LiSN-S, DDT), suggesting that attention does not contribute to the observed deficits. Furthermore, scores on the SSP2, reflecting caregiver perception of sensory deficits, were not correlated with performance on any of the other tests. Because the sensitivity/sensor subscale of the SSP2 is not exclusively related to auditory filtering and distractibility (i.e., 6 of 10 questions were auditory in nature) scores may be more reflective of global sensory processing issues than auditory deficits. Alternatively, the lack of correlation may imply that caregivers overestimate sensory issues experienced by their children, or clinical measures do not accurately depict real-world functioning.

Overall, findings show significant auditory deficits on auditory filtering, speech recognition in noise, binaural integration and attention in a population of autistic patients with normal pure-tone hearing thresholds and reported listening difficulties. These auditory challenges would have been missed if relying only on traditional audiological tests measuring peripheral hearing sensitivity.

4.2. Comparisons to published research

The types of parent-reported auditory issues identified in the present study are similar to those reported in previous investigations, particularly those related to aversion to noise, attention, and a history of otitis media (Table 3 (Adams et al., 2016; Antshel & Russo, 2019; Myne & Kennedy, 2018; Williams, Suzman & Woynaroski, 2021)). The frequency of co-occurring conditions other than ASD was computed from history forms and compared to published reports in autistic children and neurotypical peers (Table 3). Differences between published and observed rates of comorbid conditions may reflect targeted study recruitment of patients with presenting auditory concerns. In the present study, almost half (49%) of the patients had a comorbid diagnosis of ADHD, and over a third (33%) failed the full-scale attention subscale of the IVA-QS assessment with similar proportions in the auditory and visual subscales. These findings are consistent with previous research showing high proportions of attention deficits in children with a diagnosis of ASD (Corbett & Constantine, 2006; Jang et al., 2013; Sturm et al., 2004). Although poor attention may influence performance on auditory tasks, in the present study, no relationship was observed between attention and patient performance on any auditory tests (i.e., BKB-SIN, LiSN-S, DDT), suggesting that attention did not significantly influence results. There are a number of factors, including attention, that can contribute to listening difficulties in children, especially those with ASD (Dawes & Bishop 2009; Dawes, Bishop, Sirimanna & Bamiou, 2008; Dillon & Cameron, 2021). However, current study findings reiterate that attention and listening issues are not necessarily always related. Listening difficulties can be present without attention difficulties, and vice-versa.

Results of the current study, showing that the majority of autistic patients with reported listening difficulties (86%) performed abnormally on at least one measure, align with previous research showing auditory deficits across multiple test measures in autistic children with normal hearing sensitivity (Rance et al., 2014; Schafer et al., 2013, 2020). This may not reflect the listening abilities of the broader population of autistic children because our sample included only those with reported listening concerns. In the current study, 40% of patients had significantly reduced scores on measures of speech recognition in noise (BKB-SIN, LiSN-S), which has also been reported in previous studies with similar populations and methodologies (Rance et al., 2014, 2017; Schafer et al., 2013, 2016,

Table 3
Prevalence of common caregiver-reported conditions compared to published data in school-aged autistic children and neurotypical peers.

Co-occurring condition	ASD sample (%)	Published ASD (%)	Published non-ASD (%)
Aversion to Noise	73.2	41 – 61 (Williams et al., 2021)	3 – 17 (Rosing, Schmidt, Wedderkopp & Baguley, 2016)
Attention/ADHD/ADD	49.2	40 – 70 (Antshel & Russo, 2019)	8–11 (Goodsell et al., 2017)
Recurrent otitis media	34.6	Increased, unspecified (Adams et al., 2016)	1 – 12 (Daly, 1997)

Schafer et al., 2020). Additionally, our study patients who demonstrated poor speech recognition-in-noise skills were also likely to display poor dichotic listening ability and ear asymmetry (e.g., left-side dominance). As a reminder, performance on functional auditory and sensory measures could not be explained by hearing sensitivity or attention. Fourteen percent of patients showed no deficits on any test. In these patients, it is possible that the parent-reported auditory concerns prompting the clinical appointment resemble listening issues but arise from other underlying deficits (e.g., language), or that the tests used herein did not capture parent-reported listening issues.

4.3. Clinical implications and future directions

To date, this is the first study to document auditory challenges in a large clinical population of autistic children with normal sound detection thresholds and perceived listening difficulties. The majority of patients exhibited abnormal auditory performance that could not have been identified via traditional audiological testing (i.e., pure-tone audiogram). Findings provide a compelling justification for expanding the traditional audiological battery when assessing autistic children to include sensitive assessments beyond the pure-tone audiogram, which may detect a broader range of functional auditory challenges.

By creating a pathway for detecting listening difficulties not associated with pure-tone hearing loss, hearing healthcare professionals can potentially offer interventions to support autistic children by improving auditory functioning. There is a growing body of literature supporting the efficacy of auditory training and personal and soundfield remote microphone technology for autistic children with normal sound detection thresholds (Rance et al., 2014, 2017; Schafer et al., 2013, 2016, 2019; Wilson et al., 2021). In these studies, remote microphone technology use has been shown to not only improve speech understanding in noise and over distance, but also to be associated with broader improvements in functioning such as reduced listening effort and improved parent- and teacher-reported behavior. As a result, management options should be considered for autistic children who have listening difficulties given their potential to improve behavior in home, school and other real-world environments (Rance et al., 2017; Schafer et al., 2016).

Future research to develop or identify an existing auditory-specific questionnaire assessing the listening difficulties of autistic children may be useful, particularly for patients who are lower functioning in whom behavioral auditory assessments cannot be completed. While the SSP2 appears to be sensitive and efficient, it is a multisensory questionnaire lacking a specific auditory subscale. Because 35% of our study patients reported recurrent otitis media, future research should explore the impact of otitis media on performance on this and other test measures used in the current study. Previous research has reported an association between otitis media and auditory hypersensitivity, speech and language delay, deficits in social communication, and listening difficulties in autistic individuals (Yu & Wang, 2021). Furthermore, in neurotypical children, otitis media and conductive hearing loss has been linked to poorer speech perception in background noise years after sound detection returned to normal (Graydon, Rance, Dowell & Van Dun, 2017; Tomlin & Rance, 2014).

4.4. Limitations

The primary limitations of this study relate to missing data, the wide range of tools accepted to support ASD diagnoses, and potential issues generalizing findings to the broader ASD population. Because this was a retrospective study on a clinical population, results were not available in all patients on all tests due to cooperation, time, ability, etc. Test order may have impacted results for some tests because several patients were less willing or able to complete testing later in the test order. Also, results may not apply to autistic children who are lower functioning or to those without perceived listening difficulties. The current study only included high-functioning autistic children who presented to listening clinics due to listening difficulties. Another limitation of the current study is the lack of information regarding cognitive, speech, language, and other areas of auditory processing in some patients, which could impact performance on auditory tests (Dawes & Bishop 2009; Dawes et al., 2008; Dillon & Cameron, 2021).

Finally, as evidenced by the variety of tools used by outside professionals to diagnose ASD, there is no universally-accepted approach to diagnosis. This study would have benefited from a validated screening tool such as the Social Communication Questionnaire (SCQ; Rutter, Bailey & Lord, 2003) to support ASD diagnoses made in the absence of ASD diagnostic tools.

5. Conclusion

This study evaluated auditory functioning in a large, multicenter clinical sample of autistic children with normal pure-tone hearing and perceived listening difficulties. Eighty-six percent of autistic patients showed deficits on one or more of the tests. Significantly increased fail rates were documented in autistic children relative to normative data in neurotypical children with no reported listening difficulties on tests of speech recognition in noise, binaural integration, and attention. Positive correlations were calculated between speech recognition in noise (LiSN-S and BKB-SIN) and binaural integration (DDT) measures, but neither were related to parent-reported auditory sensory scores (SSP2). Attention and cognitive ability did not influence performance on any of the tests. Given the large proportion of autistic children who presented clinically with deficits on study test measures, incorporating measures beyond the pure-tone audiogram will position hearing healthcare providers to identify and potentially mitigate listening difficulties in autistic children.

CRediT authorship contribution statement

Philippa James: Conceptualization, Methodology, Investigation, Data curation, Writing – original draft, Writing – review &

editing. **Erin Schafer:** Conceptualization, Methodology, Investigation, Data curation, Project administration, Writing – original draft, Writing – review & editing. **Jace Wolfe:** Conceptualization, Methodology, Writing – review & editing, Project administration. **Lauren Matthews:** Methodology, Investigation. **Stephanie Browning:** Methodology, Investigation. **Jacob Oleson:** Methodology, Software, Formal analysis, Data curation. **Eldon Sorensen:** Software, Formal analysis, Data curation. **Gary Rance:** Writing – review & editing, Supervision. **Lucy Shiels:** Investigation. **Andrea Dunn:** Conceptualization, Methodology, Formal analysis, Resources, Data curation, Writing – original draft, Writing – review & editing, Supervision, Project administration.

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