

Auditory Characteristics of Children with Autism

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Objectives: The objectives of this study were (1) to describe the auditory characteristics of children with autism relative to those of typically developing children and (2) to describe the test-retest reliability of behavioral auditory test measures with this population of children with autism.

Design: Audiometric data were obtained from 22 children diagnosed with autism and 22 of their typically developing peers. The audiologic test battery consisted of behavioral measures (i.e., visual reinforcement audiometry, tangible reinforcement operant conditioning audiometry, and conditioned play audiometry) and physiological measures (auditory brain stem response audiometry, distortion product otoacoustic emissions, and acoustic reflexes).

Results: Children with autism had physiologic test results equivalent to their typically developing counterparts. That is, no differences in auditory brain stem response audiometry, distortion product otoacoustic emissions, or acoustic reflex results were noted between the children with autism and typically developing children. However, behavioral measures revealed that about half of the children diagnosed with autism presented pure-tone averages outside of normal limits (i.e., >20 dB HL), although their response thresholds to speech were within normal limits. All behavioral test results were within normal limits (i.e., ≤20 dB HL) for the typically developing children. In addition, test-retest variability was typically 15 dB or greater for children with autism as compared with variability of 10 dB or less for most of the typically developing children.

Conclusions: Children with autism demonstrated essentially equivalent results on a battery of physiological auditory tests as those obtained from typically developing children. However, on average, behavioral responses of children with autism were elevated and less reliable relative to those of typically developing children. Furthermore, approximately half of the children with autism demonstrated behavioral pure-tone averages outside of the normal hearing range (i.e., >20 dB HL) despite

having normal to near-normal hearing sensitivity as determined by other audiometric measures.

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INTRODUCTION

Autism is a developmental disorder characterized by a triad of symptoms: qualitative impairments in social interaction, qualitative impairment in communication, and restricted, repetitive, and stereotyped patterns of behaviors, activities, and interests (American Psychological Association [APA], 1994). In recognition of the variability of symptom expression and severity, and the existence of related disorders with overlapping symptomatology (e.g., Rett syndrome), autism was conceptualized as a spectrum disorder in the late 1980s (Allen, 1988). Autism is thought to have an early onset, with symptoms appearing before 30 mo of age in the majority of cases (APA, 1994; Filipek et al., 1999; Stone et al., 1999). However, a definitive diagnosis of autism is often not made until the age of 4 to 4½ yr (Filipek et al., 2000; Siegel, Pliner, Eschler, & Elliot, 1988; Stone & Rosenbaum, 1988) as a result of overlapping conditions and scant information on behavioral characteristics at younger ages. Recently, additional diagnostic tools have become available and may result in a lower average age of identification (Lord et al., 2000; Stone, Coonrod, & Ousley, 2000; Wing, Leekam, Libby, Gould, & Larcombe, 2002).

Prevalence estimates of autism have increased significantly over time from reports of 1 to 5 children per 10,000 in the 1970s (Brask, 1972; Treffert, 1970) to reports of 5 to 60 per 10,000 in the 1990s and this century (Bertrand et al., 2001; Kadesjö, Gillberg, & Hagsberg, 1999; Scott, Baron-Cohen, Bolton, & Brayne, 2002; Yeargin-Allsopp et al., 2003). Whether there has been a true rise in prevalence of autism over time or if the reported changes in prevalence can be explained by changes in diagnostic criteria and increased awareness of the disorder by parents and professionals remains to be seen (Rutter, 2005; Wing & Potter, 2002). Boys are affected with autism more often than girls, at a ratio of 3 to 4:1 (Van Bourgondien, Mesibov, & Dawson, 1987). Seventy percent to 80% of children with autism function intellectually within the range of mental retardation (Freeman, Ritvo, Needleman, & Yokata, 1985; Ghaziuddin, 2000). Autism is presumed to have an

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organic basis, though no single or unique etiological process has been identified (Gillberg, 1990). There is evidence from twin studies that there is a genetic basis in the etiology of over 90% of autism spectrum disorders (Rutter, 2000).

The presence of unusual sensory responses is considered an associated feature of autism, but such features are not required for diagnosis (APA, 1994). However, the literature is replete with clinical and anecdotal reports of abnormal sensory responses in children with autism, and disturbances have been reported for all sensory modalities including the auditory system (Berkell, Malgeri, & Streit, 1996; Novick et al., 1980; Ornitz, 1989; Rapin, 1991; Tang, Kennedy, Koppekin, & Caruso, 2002). In fact, a common characteristic associated with childhood autism is abnormal responses to auditory stimuli. Although it is commonly believed that children with autism exhibit a variety of auditory complications, little empirical evidence exists to support this long-standing premise. A study of home videotapes of first birthday parties revealed a failure of toddlers with autism to orient to their name (Osterling & Dawson, 1994). Other reported auditory problems associated with childhood autism include hypersensitivity to sound, painful hearing, and abnormalities in auditory processing (Berkell, Malgeri, & Streit, 1996; Grandin & Scariano, 1986; Rimland & Edelson, 1992; Rimland & Edelson, 1994).

Attempts have been made to examine whether a relationship exists between auditory brain stem dysfunction and autism (Courchesne, Akshoomoff, & Townsend, 1992; Courchesne, Courchesne, Hicks, & Lincoln, 1985; Fein, Skoff, & Mirsky, 1981; Gillberg, Rosenhall, & Johansson, 1983; Klin, 1993; McClelland, Eyre, Watson, Calvert, & Sherrod, 1992; Rosenblum, Arick, Krug, Stubbs, Young, & Pelson, 1980; Rosenhall, Nordin, Brantberg, & Gillberg, 2003; Rumsey, Grimes, Pikus, Duara, & Ismond, 1984; Sohmer & Student, 1978; Tanguay, Edwards, Buchwald, Schwafel, & Allen, 1982; Taylor, Rosenblatt, & Linschoten, 1982; Wong & Wong, 1991). Although several of these studies have reported delayed conduction times in the auditory brain stem evoked potentials (ABRs) of children with autism (Fein et al., 1981; Gillberg et al., 1983; McClelland et al., 1992; Rosenblum et al., 1980; Rosenhall, Nordin, Brantberg, & Gillberg, 2003; Sohmer & Student, 1978; Taylor et al., 1982; Wong & Wong, 1991), other investigators have not identified such distinctions between experimental and control groups (Courchesne et al., 1985; Rumsey et al., 1984). Two studies (Rumsey et al., 1984; Tanguay et al., 1982) have even found shortened conduction times in children with autism. A case report of two young children with autism identified a prevalent

wave I amplitude relative to other waves in their ABRs (Coutinho, Rocha, & Santos, 2002). However, no control subjects were tested for comparison purposes. These ABR studies are not easily comparable because of differences between laboratories in subject selection criteria, stimulus parameters, waveform identification criteria, and definitions of waveform abnormality. For example, because the I-V interpeak interval is dependent on peripheral hearing status, different audiometric configurations will have different effects on the conduction time. Not all studies included information on how, or even if, audiometric or tympanometric data were obtained before obtaining ABRs. In addition, a lack of control in gender of subject populations could explain, at least in part, the noted ABR differences in previous studies (McClelland & McCrea, 1979; Mochizuki, Ohkubo, Tataru, & Motomura, 1982; O'Donovan, Beagley, & Shaw, 1980). In summary, the ABR data currently available do not provide clear evidence for brain stem dysfunction in individuals with autism.

Several studies using retrospective parental reports have provided evidence for abnormal auditory responses in some young children with autism. Relative to typically developing children, children with autism have been described more often as being preoccupied with or agitated by noises (Monville & Nelson, 1994; Ornitz, Guthrie, & Farley, 1978). Percentages of parents reporting these symptoms ranged from 21 to 39% for preoccupation and from 42 to 53% for agitation (Ornitz et al., 1978; Volkmar, Cohen, & Paul, 1986). Children with autism have been reported to demonstrate more abnormal responses to noises (e.g., demonstrations of fascination or distress) relative to children with mental retardation, typically developing children, and children with expressive aphasia (Dahlgren & Gillberg, 1989; Wing, 1969). Hypersensitivity to noises has been reported by 32 to 81% of parents of children with autism (Hoshino et al., 1982; Ohta, Nagai, Hara, & Sasaki, 1987; Ornitz et al., 1978; Veale, 1994; Volkmar et al., 1986). Hypersensitivity is reported more frequently for children with autism than for children with mental retardation and typically developing children (Dahlgren & Gillberg, 1989; Hoshino et al., 1982; Ohta et al., 1987).

Finally, Jure, Rapin, & Tuchman (1991) examined the records of 46 children diagnosed with hearing impairment and autism. The children were identified from a total population of 1150 children with hearing impairment. The severity of autistic behavior was not found to be related to the severity of hearing loss. In almost one half of the children, there was inappropriate educational management because of inaccurate diagnosis; either autism was not identified once hearing loss was diagnosed or hear-

ing loss was not identified once autism was diagnosed.

Clearly, a need exists to investigate systematically the auditory characteristics of children with autism. Specifically, a distinction between what auditory behaviors may reflect peripheral auditory sensitivity and what involves the perception of sound has not been made in children with autism. Previous studies comparing the ABR of children with autism with typically developing children have failed to (1) institute appropriate matching criteria, such as age and gender, between groups, (2) document hearing or middle ear status of the children, and/or (3) ensure that the children were in sufficiently quiet states to obtain valid test results. This last point is of particular importance, given the likelihood that children with autism would be in an agitated state during testing if not sedated. Furthermore, behavioral measures of auditory sensitivity in children with autism have not previously been examined for validity and test-retest reliability. Nonetheless, behavioral auditory tests have been used to provide a rationale for some treatments to determine treatment protocols and as an indicator of treatment effectiveness in children with autism by some investigators (e.g., auditory integration training; Berard, 1993; Rimland & Edelson, 1994). As such, we sought to quantify objectively the auditory characteristics of children with autism by using a variety of physiological test procedures. In addition, we examined the validity and repeatability of behavioral audiologic measures commonly used with this population. Specifically, we tested two hypotheses:

(1) Children with the diagnosis of autism demonstrate physiological auditory test results equivalent to those of typically developing children of similar chronological ages;

(2) Traditional behavioral hearing test methods have poor test-retest reliability and are in poor agreement with other physiological measures when used with a population of children with autism.

MATERIALS AND METHODS

Subjects

Twenty-two children (19 boys and 3 girls) with autism were enrolled in the experimental group. The average age of the experimental subjects was 5:7 yr (range, 3:2 to 10:3 yr). Eligibility determination was made by a licensed psychologist and included a diagnosis of autism consistent with the classification of the Diagnostic and Statistical Manual (DSM) IV (APA, 1994) and the Childhood Autism Rating Scale (Schopler, Reichler, & Renner, 1988). All children received cognitive evaluations performed by a licensed psychologist. However, level

of cognitive functioning did not serve as a selection criterion for the experimental group. The decision to include children with a range of cognitive function within the experimental group was based on practical and scientific considerations. That is, given that 70 to 80% of children with autism function outside the range of normal cognition (Freeman, Ritvo, Needleman, & Yokata, 1985; Ghaziuddin, 2000), eliminating children with autism who had cognitive deficits would pose an extreme restriction on our ability to locate subjects. In addition, given the large number of children with autism who have cognitive deficits, eliminating them from our subject pool would restrict the generalizability of our findings. Finally, matching groups on age rather than cognitive level was preferable for between-group comparisons of the physiologic tests included in the test battery.

Twenty-two typically developing children matched on the variables of chronological age (+6 or -6 mo) and gender to the experimental subjects served as a control group. The average age of the control subjects was 5:3 yr (range, 3:2 to 9:9 yr). Each child had normal cognitive function, based on performance on the Peabody Picture Vocabulary Test (Dunn & Dunn, 1997; +1 or -1 standard deviation of standard score) and, for children younger than 5 yr of age, the Child Development Inventory was also conducted as a determinant of age appropriate development (Ireton & Glascoe, 1995). See Table 1 for a summary of demographic data on all enrolled subjects.

Children in either group were excluded from the study if they had ever undergone auditory integration training, or if they had a history of myringotomy tubes. An additional 18 children (10 experimental and 8 control subjects) were initially enrolled but were unable to complete the study as a result of repeated abnormal tympanometry (8 children), enrollment in auditory integration training (3 children), change in their diagnosis of autism (3 children), and scheduling conflicts (4 children). The three children who received a change in diagnosis were reclassified as still on the autism spectrum but not meeting DSM-IV criteria for autism.

Subject recruitment procedures were approved by the Vanderbilt University Institutional Review Board. Children with autism were recruited from diagnostic clinics and treatment programs designed for children with autism within the university and the community at large. Parental consent was obtained for each child participant at the onset of the investigation. Children were compensated for their time.

Equipment and Procedures

Testing occurred across two to five sessions, depending on the level of cooperation of the child. The

TABLE 1. Demographic data and test results for children with autism

	Behavioral					Test type	ABR			CA	Cognitive level	Gender	
	Speech	0.5 kHz	1 kHz	2 kHz	4 kHz		Tones	Click	OAE				AR
1	0	5	5	5	5	TROCA	WNL	WNL	Present	Present	7:8	Normal	Male
2	10	25	20	25	5	TROCA	-	-	Present	Present	6:2	Low average	Female
3	10	50	20	30	30*	VRA	WNL	WNL	-	Present	6:9	Severe	Male
4	0	15	20	10	15	TROCA	-	WNL	Present	-	10:3	Severe	Male
5	20	25	20	25	35*	VRA	WNL†	WNL	Present	Present	3:7	Mild	Male
6	25	25	25	50	25*	VRA	-	-	-	-	6:7	Moderate	Male
7	30	50	20	10	20*	VRA	WNL‡	-	Present	Present	3:10	Mild	Male
8	20	NR	50	50	60*	VRA	WNL§	WNL	-	Present	3:6	Mild	Male
9	20	NR	NR	NR	NR*	VRA	-	-	Present	Present	3:2	Moderate	Female
10	5	25	15	5	10	CPA	WNL	WNL	Present	Present	5:10	Moderate	Female
11	50	NR	30	30	NR*	VRA	WNL	WNL	Present	-	3:11	Moderate	Male
12	15	10	20	15	10	TROCA	-	-	Present	Present	6:9	Mild	Male
13	0	10	0	10	10	TROCA	WNL	WNL	-	-	7:6	Mild	Male
14	5	20	15	15	15	TROCA	WNL	WNL	-	-	8:9	Mild	Male
15	5	15	20	10	10	TROCA	-	-	Present	Present	5:4	Low average	Male
16	10	5	5	5	5	TROCA	WNL	WNL	Present	Present	8:5	Moderate	Male
17	10	10	20	20	10	TROCA	WNL	WNL	-	-	6:8	Mild	Male
18	20	NR	10	20	10	VRA	WNL	WNL	-	Present	3:4	Moderate	Male
19	10	25	20	20	20*	VRA	-	-	-	-	4:4	Severe	Male
20	20	NR	25	35	35*	VRA	WNL	WNL	-	Present	6:5	Mild	Male
21	10	10	20	10	20*	VRA	WNL	WNL	-	Present	3:10	Moderate	Male
22	10	10	10	10	0*	TROCA	WNL	WNL	Present	Present	5:2	Mild	Male

ABR, auditory brain stem response; OAE, otoacoustic emission; AR, acoustic reflex; CA, chronological age; NR, no response; WNL, within normal limits; VRA, visual reinforcement audiometry; TROCA, tangible reinforcement operant conditioning audiometry; CPA, conditioned play audiometry.

- Signifies could not test, no result.

*Sound field testing required.

†ABR WNL at 2 and 4 k.

‡ABR WNL at 0.5 k.

§ABR WNL at 1, 2, and 4 k.

22 children with autism and their typically developing counterparts received a comprehensive audiological battery of tests.

The intent of the behavioral testing was two-fold: (1) to provide an indication of subject responsivity to auditory stimuli as compared with his or her physiological responses to sound and (2) to determine the test-retest reliability of the behavioral measures. Furthermore, the behavioral test battery was developed with two primary concerns in mind: clinical applicability and test time. The protocols were designed to represent those that are commonly used clinically. In addition, the protocols were necessarily flexible to accommodate individual child differences and preferences. This was of particular importance in evaluating children with autism who, as noted earlier, may be difficult to test and often demonstrate abnormal responses to sound. Therefore, some clinician discretion could be used in making slight modifications to the protocol during the behavioral test sessions. These options are noted below in the review of the test battery. All behavioral testing was conducted by one examiner, who has more than 20 yr of pediatric testing experience.

All behavioral testing was conducted in a sound-attenuating double-walled test suite meeting ANSI

S3.1 standards (ANSI, 1999). The behavioral assessment options consisted of visual reinforcement audiometry (VRA), tangible reinforcement operant conditioning audiometry (TROCA), or conditioned play audiometry (CPA). A Grason Stadler 16, two-channel audiometer calibrated to ANSI (1996) standards was used for all VRA and CPA. A Maico MA40 portable audiometer, also calibrated to ANSI (1996) standards, was used for testing via TROCA (Gordon N. Stowe and Associates, Inc.). Etymotic Research ER3A insert ear phones were used when possible and were fitted with small-size, disposable EarLink foam tips; otherwise, TDH-39 phones or loudspeakers were used. In addition to meeting ANSI S3.6 (1996) calibration standards, biological calibrations were conducted on every day of data collection.

The behavioral test selected was determined as the procedure that the child was willing or able to do. The examiner attempted to use the test procedure that required the child's highest functioning capability. This occasionally resulted in having to change to an easier task if conditioning on a higher level task could not be achieved. VRA is typically required for children between developmental ages of about 6 mo and 2 yr of age, TROCA is typically used

for those between developmental ages of about 2 and 4 yr, and CPA for those with developmental ages above approximately 2.5 yr (Diefendorf, 1988). Certainly, it would have been optimal for comparative purposes to use the same behavioral test procedure for all participants. That was not feasible, given the considerable differences in behavioral characteristics of our study participants. However, it should be noted that minimum response levels (MRLs) obtained from VRA and CPA have been shown to be in good agreement across a variety of degrees and configurations of hearing (Diefendorf, 1988; Talbott, 1987). In addition, to minimize the potential effects of different test procedures, the same step size was used for all three procedures. For all behavioral test procedures, acquisition of thresholds for .5, 1.0, 2.0, and 4.0 kHz and speech stimuli was attempted. The protocols for each of the behavioral procedures were as follows:

- **Visual reinforcement audiometry:** Whenever possible, insert earphones were used to obtain ear-specific information. If the child would not tolerate insert earphones, circumaural phones were used or, as a last resort, testing was conducted in sound field. The test protocol was the same whether testing with earphones or through loudspeakers. The first examiner sat in the test booth and manipulated quiet toys to engage the child's attention at midline and initiated trials when the child was considered to be in a ready state. The second examiner was in the control booth operating the audiometer, viewing the child through a one-way glass and activating a reinforcer when indicated. The two experimenters communicated via microphone, earphones, and a hand-held switch under the first examiner's control that activated a light emitting diode (LED) in the control room. To avoid any inadvertent cuing of the child, the first examiner was masked with noise through earphones providing approximately 70 dB of attenuation if testing was conducted via loudspeakers. Therefore, the first examiner was generally unaware if a signal or control trial was initiated unless the signal exceeded the masking amount.

The first examiner signaled the second examiner when a trial interval should begin, based on participant readiness, by activating the LED. The second examiner then initiated a signal or control trial. If the first examiner indicated that a head turn occurred (by activating the LED) and, in fact, a signal trial occurred, the second examiner activated the reinforcer on the appropriate side. If a head turn occurred during a control trial, the reinforcer was withheld.

VRA thresholds, or MRLs, were determined using an adaptive one-up, one-down tracking procedure for rapid convergence on MRL. Step size was down 10 dB and up 5 dB.* If a child became obviously distracted during an observation interval, the trial was repeated. Similarly, if a child became obviously bored with the procedure, the second examiner could switch the type of stimulus or switch the speaker or earphone in an attempt to regain, or obtain, the child's interest. The stop criterion for MRL was the lowest level at which the child responded to two of three ascending runs.

- **Tangible Reinforcement Operant Conditioning Audiometry and Conditioned Play Audiometry:** All TROCA and CPA testing was attempted with earphones. Speech reception thresholds (SRTs) were obtained (monitored live voice) bilaterally for all children who participated in TROCA and CPA testing. Pure-tone thresholds were determined by using the same adaptive one-up, one-down bracketing procedure as described above for VRA. Children received positive verbal reinforcement on responding appropriately for the tasks. In addition, children participating in the TROCA task received a small piece of cereal as the tangible reinforcement.

Finally, a parent of each of the participants was asked (1) Have you ever suspected that your child could not hear? (2) Are there certain sounds that your child does not hear or does not seem to hear? and (3) Does your child seem to regard certain sounds as painful or distressing? These queries were made to determine if behavioral test results reflected the observations of the participants' parents.

Physiological Measures.

- **Immittance:** All immittance testing was conducted with the Welch Allyn Microtympanometer® or Grason Stadler middle ear analyzers (GSI 33 and GSI 1723) using a 226 Hz probe tone. Tympanograms were obtained in both ears on all participants on every day of data collection. Any tympanogram for which tympanometric width could not be calculated (i.e., no measurable peak) resulted in a rescheduling of the participant for testing at a later date.

* The 10 dB down and 5 dB up step size reflects a slight deviation from the protocol recommended by Tharpe and Ashmead (1993). This change to a smaller step size was considered necessary because a 5 dB difference in thresholds between adjacent frequencies on an audiogram is considered reflective of hypersensitivity by proponents of Auditory Integration Training (AIT; Bernard, 1993). AIT is a proposed and controversial treatment for auditory disorders such as those believed by some to accompany autism.

Ipsilateral acoustic reflex thresholds were attempted at 0.5, 1.0, 2.0, and 4 kHz from the right ear of all subjects. It was reasoned that because of the potential of tactile and acoustic sensitivities of many individuals with autism, obtaining reflexes from just one ear was likely to reduce temper outbursts.

- **Auditory Brain Stem Response Audiometry:** ABR testing was conducted to provide both an objective estimate of auditory sensitivity and an indication of auditory brain stem pathway integrity. Testing was performed with the child in a natural or sedation-induced state of sleep,[†] whichever was required for a quiet test. Ethical considerations prevented the sedation of the typically developing control group of children as the reliability of their audiometric test results was not in question. Previous work has indicated that the data from two ears of the same subject are highly correlated (Gorga, Reiland, Beauchaine, Worthington, & Jesteadt, 1987). Therefore, as a result of the difficulty of keeping children with autism sedated for long periods of time, only the data from the right ear of each subject were obtained.

All subjects were tested by using the Nicolet Spirit Evoked Potential Unit with IBM compatible 80486 SX (25 MHz) processor and 120 Mbyte hard drive with insert earphones (Etymotic Research ER3A). Testing included threshold estimation, using tone bursts with center frequencies of 0.5, 1.0, 2.0, and 4.0 kHz with linear rise times equaling 2 cycles, plateau times of 1 cycle, and linear fall time of 2 cycles. Neurological assessment used click stimuli (100 μ sec) with alternating polarity at a rate of 21.1 per second. Analysis time was 15 msec. The EEG was filtered between 30 and 3000 Hz (12 dB/octave slope). The electrode montage consisted of vertex to left earlobe, vertex to right earlobe, one-channel recordings. An electrode placed at the forehead served as the ground electrode. Electrode impedances were <5 kOhms and interelectrode impedances were within 1.5 kOhms. Standard artifact rejection was used to eliminate any sweep in which the voltage exceeded the maximum range of the A/D converter. In addition, a manual pause mechanism could be used at any time according to the examiner's discretion.

Click stimuli were presented at 80 dB nHL. The click was averaged across 2000 sweeps and was replicated. Wave I, III, and V latencies were iden-

tified for the 80 dB nHL click-evoked waveforms. In addition, wave V thresholds for click and tonal stimuli were also identified. Identification of wave forms and threshold levels for all subjects was made by a single examiner after the test session to maintain consistency. This examiner was blind to the behavioral test results, and to the subject group (i.e., autistic versus typically developing group). ABR threshold was determined as the lowest stimulus intensity level where the presence of a wave V was observed. In all cases, the examiner presented stimuli below this level to verify the absence of a waveform.

- **Distortion Product Otoacoustic Emissions:** Stimuli for DPOAE measurements were generated by the Otodynamics, Ltd., V5 ILO Otoacoustic Emissions System (ILO92). DPOAEs were ascertained by using the iso—(f_2/f_1) paradigm, or the "DPOAE audiogram" (Smurzynski et al., 1993). Two stimuli of different intensities (65/55 dB SPL) with a frequency ratio of approximately 1.2 were used. Measurements included the distortion product level, noise floor, and the signal-to-noise ratio (DP level minus the noise floor). A response was considered present when the DP level was equal to or greater than 3 dB, defined relative to the mean noise level plus two standard deviations, and a minimum of 0 dB SPL.

RESULTS

For purposes of interpreting the significance of the behavioral test findings, it is important to note that although every child with autism did not cooperate for every test procedure (for reasons delineated in the following discussion), every child was determined to demonstrate normal to near-normal hearing sensitivity in at least one ear. In most cases, this conclusion was based on the results of a combination of at least two test measures including behavioral test results, DPOAEs, or frequency-specific ABR.[‡] Table 1 provides a summary of all audiometric test results for each subject with autism.

Behavioral Auditory Assessment

All children in the autistic and typically developing groups participated in this portion of the study ($N = 22$ for each group). As evident in Table 1, the most frequently used test with the children in the autistic group was VRA (50%). In contrast, the majority of the

[†] Children requiring sedation were administered 40 to 60 mg of chloral hydrate per kilo of body weight if given orally and 60 to 70 mg per kilo of body weight if given rectally. If that dosage was not satisfactory in inducing sleep, they were administered a second half-dosage. All sedated children were monitored continuously by a registered nurse and/or a pediatrician until they awoke.

[‡] For two subjects who could not be sedated for physiologic measures, normal hearing was confirmed after the completion of this study by the presence of a normal frequency-specific ABR (subject 19) or subsequent behavioral tests indicating normal hearing sensitivity (subject 6).

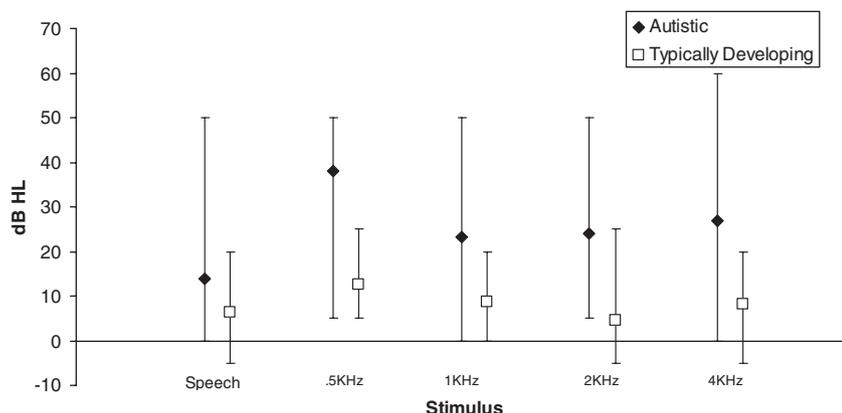


Fig. 1. Mean behavioral response thresholds (bars represent range) in dB HL for children with autism ($N = 22$) and typically developing children ($N = 22$). This figure excludes five thresholds for 0.5 kHz, one threshold for 1.0 kHz, one threshold at 2 kHz, and two thresholds at 4.0 kHz for children with autism who did not respond at those frequencies.

typically developing children were able to participate for CPA (77%). None of the children in the control group required testing with VRA. There did not appear to be any association between cognitive status of the children with autism and which behavioral task they required. That is, of those three children with normal to low average cognitive status and the nine with mild cognitive impairment, four required testing with VRA and eight participated for TROCA. The one child with autism who participated for CPA had a diagnosis of moderate cognitive impairment. Conversely, of the three children with autism and diagnoses of severe cognitive impairment, two required the VRA task but one participated for TROCA.

Response Thresholds • Recall that behavioral testing was conducted twice on two separate visits. For 93% of all participants, the second behavioral test occurred within 6 weeks of the first test. Delays in testing for the remaining 7% were the result of scheduling problems. Initial analyses of response levels were made from the data of the first behavioral test (VRA, TROCA, or CPA) for all children. Thresholds were established for all of the typically developing children for all test stimuli. Although all of the children with autism were able to provide a response threshold for speech stimuli, five children (23%) did not respond at all to one or more of the tones in the first test session. Therefore, for children who did not respond to a tone at the maximum output of the audiometer, a threshold of 120 dB was entered for initial analyses. Mean response thresholds and ranges for all subjects, regardless of test type, can be viewed in Figure 1. As seen in that figure, on average, response thresholds were higher for children with autism than for those of their typically developing peers. Note that all behavioral thresholds for the typically developing children were within normal limits bilaterally (i.e., <20 dB HL) for all test stimuli. However, the mean response thresholds for the children with autism were within normal limits for speech stimuli but were >20 dB HL for tones. To assess effects of group and stimulus,

mean response thresholds from the right ear of all the children were subjected to a mixed model analysis of variance (ANOVA). In cases for whom only sound field results were obtained (11 subjects in the experimental group, none in the control group), sound field response thresholds were used in the analyses. Results indicated significant main effects of group [$F(1, 42) = 12.97, p < 0.01$] and stimulus [$F(4, 168) = 6.07, p < 0.01$] but no significant stimulus x group interaction.

To follow up on the significant effect of stimulus, an ANOVA was conducted for each group, comparing the response thresholds for speech and tones (collapsed across frequency). For the children with autism, response thresholds for the tones were significantly higher than their response thresholds for speech (29.7 dB and 13.9 dB, respectively) [$F(1, 21) = 8.53, p < 0.01$]. For the typically developing children, there was not a significant difference between their average response threshold for tones and speech (8.8 dB and 6.6 dB, respectively). Because the difference in response thresholds for the tones and speech stimuli in the autistic group could have been driven solely by those children who did not respond to one or more of the tones, the data for those children ($N = 5$) were removed, and analysis was repeated. Response thresholds for tones were still significantly higher than for those of speech (16.5 dB and 10.8 dB, respectively) [$F(1, 17) = 10.75, p < 0.01$].

In the autistic group, one child was tested behaviorally by using CPA and 10 were tested by using TROCA. In the control group, 17 children were tested by using CPA and five were tested by using TROCA. The mean values and standard deviations of the thresholds for speech and tones for both groups are presented in Table 2. All children, typically developing, and those with autism capable of performing these play audiometric procedures provided thresholds within normal limits for speech and tones (i.e., ≤ 20 dB HL).

In contrast, children with autism requiring the

TABLE 2. Mean audiometric thresholds in dB (standard deviation) for children participating in play audiometric tasks (TROCA and CPA)

Speech*	Children with autism (N = 11)				Speech*	Typically developing children (N = 22)			
	0.5 Hz	1.0 Hz	2.0 Hz	4.0 Hz		0.5 Hz	1.0 Hz	2.0 Hz	4.0 Hz
6.3 (6.6)	13.6 (7.7)	13.6 (8.4)	11.3 (8.7)	8.6 (6.8)	6.3 (5.9)	12.5 (5.5)	8.6 (6.1)	4.5 (7.3)	8.2 (7.5)

*Speech Awareness Thresholds (SAT) or Speech Reception Thresholds (SRTs) were obtained depending on the developmental level of the child.

VRA test procedure, on average, exhibited response thresholds to speech that were within normal limits but response thresholds that were elevated for tones. On average, VRA response thresholds to tones for the children with autism were about 15 dB higher (worse) than response thresholds to speech resulting in a significant effect of stimulus [$F(4,40) = 3.48, p < 0.01$]. Furthermore, the average response threshold for speech was significantly lower than the average response threshold for tones (collapsed across frequency [$F(1,10) = 10.49, p < 0.01$]. In planned analytical comparisons, results indicated that the speech threshold was significantly lower than all tonal thresholds except 4.0 kHz (i.e., 0.5, 1.0, and 2.0 kHz) [$F(1,17) = 11.10, p < 0.01$; $F(1,19) = 8.51, p < 0.01$; $F(1,20) = 4.91; p < 0.05$, respectively].

Test-Retest Reliability • Both behavioral tests were always of the same type. That is, if a subject was tested with VRA during the first evaluation, the second evaluation also used VRA. We chose to consider test-retest reliability in the context of change in response thresholds between the first and second behavioral test. Figure 2 presents the test/re-test differences (in dB) as a function of percentage of all thresholds for each study group. Six children with autism did not respond to one or more frequencies for either the first or second behavioral test. Three of

these six subjects did not respond to one or more frequencies for the first test but did on the second test; one responded to all frequencies for the first test but not the second test; and two subjects did not respond to one or more frequencies for both tests.

Ninety-seven percent of the response thresholds of the typically developing children varied by 10 dB or less between tests and none of their thresholds for the two test sessions varied by more than 15 dB. However, 64% of the response thresholds of the children with autism varied by 15 dB or more between tests.

Parent Observations • Recall that parent(s) of the participants were asked three questions about their child’s responses to sounds. In response to the question “Have you ever suspected that your child could not hear?”, the parents of eight of the children with autism answered “yes,” whereas parents of only two of the typically developing children responded “yes.” Of those 10 children suspected at some time of not hearing by their parents, three did not respond to one or more of the pure tone stimuli during behavioral testing. Parents of two of the children with autism responded that there were certain sounds that their child could not or did not seem to hear. None of the parents of the typically developing children responded affirmatively to that question. Finally, in response to the question “Does your child seem to regard certain sounds as painful or distressing?”, parents of 17 of the children with autism responded “yes,” whereas parents of only 6 of the typically developing children responded affirmatively. Therefore, although the majority of the parents of the children with autism reported some apparent hypersensitivity to certain sounds, the behavioral thresholds for these children were higher overall.

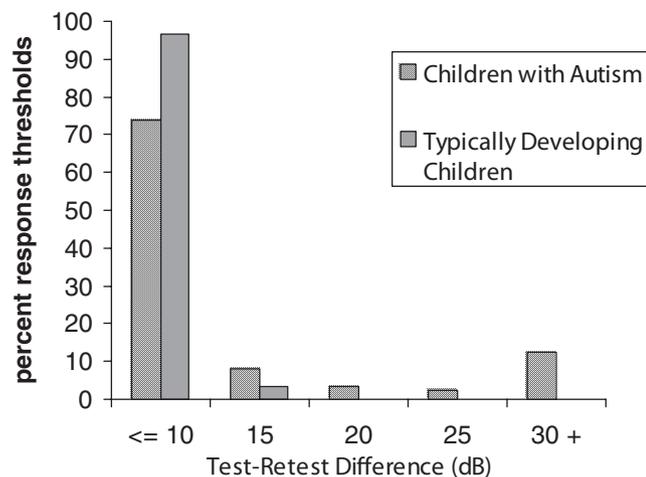


Fig. 2. Percentage of total response thresholds from the first behavioral test within 10, 15, 20, 25 dB or ≥30 dB of the second behavioral test for both the typically developing children (N = 22) and the children with autism (N = 22).

Acoustic Reflexes

Ipsilateral acoustic reflexes were obtained for 0.5, 1, 2, and 4 kHz tones for the right ear of 15 children with autism and 21 typically developing children. The children who were not tested exhibited resistive behavior that resulted in uninterpretable results. No significant between-group differences or a significant group x frequency interaction was revealed. To ensure against bias with our uneven sample sizes, an ANOVA was conducted on 15 children in each

TABLE 3. Mean auditory brain stem response wave V threshold values (SD) in nHL for clicks and 0.5, 1.0, 2.0, and 4.0 kHz tone bursts for the right ears of children with autism and typically developing children

Group	Click	0.5 kHz	1.0 kHz	2.0 kHz	4.0 kHz
Typically developing	8.4 (6.6) (N = 21)	21.0 (8.9) (N = 21)	10.7 (6.9) (N = 19)	5.7 (8.4) (N = 18)	6.5 (9.9) (N = 20)
Autistic	12.6 (8.1) (N = 15)	26.2 (6.6) (N = 13)	17.5 (9.9) (N = 14)	9.7 (6.1) (N = 14)	11.4 (6.6) (N = 14)

All values were within normative values for this clinic.

group who were matched for age. Again, no significant between-group differences or group x frequency interaction were revealed.

Auditory Brain Stem Response

Sixteen of the 22 children with autism and 18 typically developing children participated in this component of the study. Recall that for ethical reasons, the typically developing children were not sedated for this procedure because the reliability of their auditory thresholds was not in question. Additionally, not all children with autism could fall asleep or stay asleep even with sedation. Therefore, we were unable to obtain data for all five stimuli for all participating subjects because of limitations imposed by subject state (see Table 1).

ABR Thresholds • Wave V threshold values (M and SD in dB nHL) for click- and tonal stimuli for both groups of subjects can be viewed in Table 3. There was a significant main effect of stimulus [$F(4,112) = 11.49, p < 0.01$], with thresholds tending to be higher for 0.5 and 1.0 kHz tones than for 2.0 and 4.0 kHz tones and clicks. There was no significant effect of group and no significant stimulus x group interaction.

ABR Latencies • Mean wave I, III, and V absolute latencies and I-III, III-V, and I-V latency intervals for children in both the experimental and control groups are listed in Table 4. These data were obtained in response to a click at 80 dB nHL. No significant between-group differences for absolute or interwave latencies were observed.

Despite the lack of between-group differences in ABR thresholds and latencies, it was of concern that a number of children, particularly in the experimental group, could not participate for this testing.

Therefore, we reanalyzed the ABR threshold and latency data, using only age-matched pairs ($N = 12$) of subjects. Again, there were no significant between-group differences in ABR thresholds or latencies (absolute or interwave).

Otoacoustic Emissions

Level • Average DPOAE levels and noise levels for the four f_2 frequencies for both subject groups are provided in Table 5. Twelve children with autism and 13 typically developing children participated in this part of the study. For purposes of these analyses, mean DPOAE levels for f_2 frequencies of 1.5, 2.0, 3.0, and 4.0 kHz for both groups were subjected to an ANOVA. A significant effect of frequency was observed [$F(3,69) = 5.47, p < 0.01$], with higher DPOAE levels associated with lower f_2 frequencies but no significant effect of group or frequency x group interaction was observed.

Noise Floor • There was an obvious trend for the noise levels to decrease as the frequency increased. There was a significant effect of frequency [$F(3,69) = 37.7, p < 0.01$] but no significant effect of group or group x frequency interaction.

DISCUSSION

To our knowledge, this report represents the first attempt to describe objectively the auditory characteristics of children with autism. Our interest in this population was largely driven by the expansion of our intervention program for children with autism spectrum disorders over the last decade. Because children with autism frequently demonstrate an apparent hypersensitivity to sound or abnormal auditory processing (Berkell et al., 1996; Grandin & Scariano,

TABLE 4. Mean auditory brain stem response latency values (SD) in milliseconds for waves I, III, and V, and I-III, and I-V interwave latency for children with autism and typically developing children

Group	Wave I	Wave III	Wave V	I-III	III-V	I-V
Typically developing (N = 21)	1.64 (0.10)	3.92 (0.20)	5.78 (0.24)	2.28 (0.18)	1.86 (0.14)	4.13 (0.21)
Autistic (N = 15)	1.65 (0.09)	3.99 (0.12)	5.89 (0.18)	2.34 (0.11)	1.90 (0.16)	4.23 (0.17)

All latencies were within normative values for this clinic.

TABLE 5. Mean DPOAE levels and noise levels in dB (standard deviation) for the right ears of children with autism and the typically developing children

Group	f_2 Frequency			
	1.5 kHz	2.0 kHz	3.0 kHz	4.0 kHz
Typically developing (<i>N</i> = 13)				
Level	14.3 (5.2)	12.8 (6.5)	11.2 (5.4)	9.7 (2.6)
Noise level	2.2 (3.7)	1.6 (4.6)	-2.6 (2.6)	-5.7 (3.1)
Autistic (<i>N</i> = 12)				
Level	11.8 (4.2)	11.6 (4.9)	9.3 (3.7)	8.9 (3.4)
Noise level	6.1 (9.2)	2.4 (6.3)	-2.7 (5.3)	-4.2 (2.7)

1986; Rimland & Edelson, 1994; Rosenhall, Nordin, Sandstrom, Ahlsen, & Gillberg, 1999), proponents of a highly controversial treatment, auditory integration training (AIT), have suggested that children with autism may be good candidates for this intervention. Furthermore, because the founder of one of the more popular versions of AIT asserted that many children with autism have auditory "difficulty" that can be effectively treated by retraining the auditory system via AIT (Berard, 1993, p. 53), it was of interest to us to determine if, in fact, children with autism have hearing that is measurably distinct from that of typically developing children. The results of this study bring into question some of the assumptions underlying AIT.

Specifically, children with autism in this study demonstrated essentially equivalent results as those of typically developing children on physiologic measures of auditory function. Data from ABR, DPOAE, and acoustic reflex measures yielded no differences between children with autism and their typically developing peers. However, behavioral measures suggested that many children with autism presented with elevated response thresholds relative to other physiologic measures of hearing that did not require the child's active involvement. That is, when the hearing of children with autism was assessed in a manner requiring them to indicate behaviorally when they heard a sound, 41% responded in such a way as to indicate that they did not hear normally for at least one test stimulus when, in fact, other measures verified normal to near-normal hearing sensitivity.

Furthermore, the test-retest reliability of behavioral responses in children with autism was poorer than that of a typically developing control group. Although a 10 dB variation in response thresholds between tests is generally considered by clinicians to represent normal variability, the majority of children with autism in this study demonstrated a 15 dB or greater difference in response thresholds between tests. This certainly suggests that comparison

of pre- and post-treatment audiograms, as is often recommended with AIT, may not be a valid indication of treatment effectiveness.

Some caution should be maintained when considering these findings. This study included only 22 children with autism, and not all tests could be conducted on all of these children. However, given the similarity in test results across participants, specifically for the physiologic measures, it is unlikely that a larger population would yield contradictory information. Another cautionary point in the interpretation of these data is that the physiological measures used targeted the auditory system only through the early cortical projection areas (early ABR). It is possible that physiologic measures of auditory function beyond this area (e.g., middle or late latency response, event-related potentials) may yield differences between children with autism and their typically developing peers.

Another problem inherent in the behavioral testing of children with varying functional capabilities, as was the case in this study, is the necessity of utilizing different test procedures (e.g., VRA, CPA, and so forth). However, as noted previously, thresholds obtained from VRA and CPA have been shown to be in good agreement across a variety of degrees and configurations of hearing (Diefendorf, 1988; Talbott, 1987).

The results of this study also offer some important implications for clinicians, teachers, and parents. First, it is not reasonable to assume that a traditional behavioral test battery is appropriate for children with autism. Although a diverse test battery approach is often recommended and used when assessing the hearing of infants and very young children, it is not uncommon when testing older children, such as those included in the current study, to use only behavioral audiometrics. The results of the current study add support to the recommendation that children who cannot be conditioned to age-appropriate behavioral test procedures and who present response thresholds outside of the normal range be tested with additional physiological measures before assuming the presence of a hearing loss. In addition, parents and teachers should be advised of the inconsistencies between behavioral responses to auditory stimuli and true hearing sensitivity of children with autism.

It is natural to ask at this point why some children with autism demonstrate an apparent hyposensitivity to auditory stimuli despite the fact that their peripheral hearing appears to be normal based on physiological measures. Some investigators have suggested that the deficit in responsivity or orienting to sound has more to do with attentional than sensory processes (Ceponiene et al., 2003; Dawson, Meltzoff, Osterling, Rinaldi, & Brown,

1998). The results of the current study support the notion that sensory factors are not to blame for the lack of auditory attentiveness. In fact, we confirmed that many of the children with autism in this study demonstrated normal auditory function through the level of the brain stem. It would be of interest for future studies with this population to examine higher-order auditory processing skills, including those associated with auditory attention.

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