

Auditory Discrimination and Auditory Sensory Behaviours in Autism Spectrum Disorders

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Abstract

It has been hypothesised that auditory processing may be enhanced in autism spectrum disorders (ASD). We tested auditory discrimination ability in 72 adolescents with ASD (39 childhood autism; 33 other ASD) and 57 IQ and age-matched controls, assessing their capacity for successful discrimination of the frequency, intensity and duration differences in pairs of sounds. At the group level, auditory discrimination ability did not differ between the adolescents with and without ASD. However, we found a subgroup of 20% of individuals in the ASD group who showed 'exceptional' frequency discrimination skills (defined as 1.65 SDs above the control mean) and who were characterised by average intellectual ability and delayed language onset. Auditory sensory behaviours (i.e. behaviours in response to auditory sensory input) are common in ASD and we hypothesised that these would relate to auditory discrimination ability. For the ASD group, poor performers on the intensity discrimination task reported more auditory sensory behaviours associated with coping with loudness levels. Conversely, those who performed well on the duration discrimination task reported more auditory sensory behaviours across the full range measured. Frequency discrimination ability did not associate with auditory sensory behaviours. We therefore conclude that (i) enhanced frequency discrimination is present in around 1 in 5 individuals with ASD and may represent a specific phenotype; and (ii) individual differences in auditory discrimination ability in ASD may influence the expression of auditory sensory behaviours by modulating the degree to which sounds are detected or missed in the environment.

Introduction

Autism spectrum disorders (ASD) are complex neurodevelopmental disorders, characterised by social and communication impairments and rigid and repetitive behaviours (DSM-IV-TR, American Psychiatric Association, 2000; ICD-10, World Health Organization, 1993).

Attempts to tease apart the multifarious factors that contribute to the expression of ASD has led to interest in how fundamental differences in basic perception may have a bottom-up effect on particular behaviours and difficulties, as well as explain perceived strengths such as in detail-focussed processing (e.g. Milne et al., 2002; Milne et al., 2006; Mottron et al., 2007; Pellicano, Gibson, Maybery, Durkin, & Badcock, 2005). Particularly, there is increasing interest in both the research and academic communities in documenting atypical auditory processing in ASD (see Kellerman, Fan, & Gorman, 2005; Nieto Del Rincón, 2008; Samson et al., 2006; for recent reviews). Auditory perceptual processing can be measured by assessing the ability to discriminate between pure tones that vary systematically according to parameters such as frequency, intensity or duration. To date, it has been reported that individuals with high-functioning autism (mean age 18 years) are superior to controls at frequency discrimination ('same/different' judgement) and frequency categorisation ('high/low' judgement) (Bonnell et al., 2003). The majority of research using event-related potentials (ERPs) suggests enhanced neural detection of frequency changes in ASD at the pre-attentive level (mismatch negativity: MMN) (Ferri et al., 2003; Gomot, Giard, Adrien, Barthélémy, & Bruneau, 2002; Kujala et al., 2007), although a clear consensus has yet to emerge for higher-order processing of auditory stimulus changes (P3 waveform) (Ceponiene et al., 2003; Ferri et al., 2003; Gomot et al., 2002). This evidence of enhanced frequency discrimination is further supported by evidence of superior identification of and memory for pitch compared to controls (Heaton, Hermelin, & Pring, 1998), as well as enhanced sensitivity for the pitch direction of closely spaced notes (Heaton, 2005).

These findings prompt speculation that fundamental differences in the perception of sounds may be part of the autistic profile. However, the limited research in this area suggests a need for replication and clarification. In particular, more recent work suggests that enhanced frequency perception may only characterise a subgroup of individuals on the spectrum (Heaton, Williams, Cummins, & Happé, 2008), and the relatively small sample sizes used in the studies thus far (e.g. $n=12$ in Bonnell et al., 2003; $n=15$ in Heaton, 2005; $n=32$ in Heaton et al., 2008) suggest the need for further exploration. Further, it is untested whether enhanced perceptual processing can be observed across a range of stimulus characteristics (e.g. frequency, intensity, duration), or whether the effect is limited to

frequency discrimination. Thus, the first aim of our study is to characterise the auditory discrimination profile in a large sample of adolescents with ASD, at the age at which neurophysiological maturation of the auditory cortex is complete (e.g. Moore & Guan, 2001).

Atypical auditory sensory experiences (e.g. oversensitivity to loud noises or particular sounds) are highly characteristic of individuals with ASD and can cause significant impairment and distress, interfering with ongoing processing of the (social) environment and impacting negatively on adaptation. Related to this, parents and caregivers consistently reports high levels of atypical sensory behaviours, including in the auditory domain (e.g. placing hands over ears to protect from sound; preoccupation with particular sounds), in individuals with ASD (Baranek, David, Poe, Stone, & Watson, 2006; Kern et al., 2006; Kientz & Dunn, 1997; Tomchek & Dunn, 2007; Watling, Deitz, & White, 2001). Sensory experience is critical in the development of neuronal circuitry (Hensch, 2004; Jiao, Zhang, Yanagawa, & Sun, 2006). A child who consistently shuts out essential sensory input or focuses on particular sensory experiences is likely to compromise the typical neuro-developmental trajectory, with down-stream implications for perceptual, cognitive and social maturation (Mottron et al., 2007).

The causes and consequences of atypical sensory behaviours remain poorly understood. An intuitive, yet unexplored, hypothesis is that fundamental differences in the processing of auditory information can lead to atypical auditory sensory experiences and the expression of auditory sensory behaviours. For example, auditory sensory behaviours may arise from enhanced or reduced sensitivity to differences between sounds, i.e. a neurophysiological system of auditory discrimination that is more ‘finely tuned’ or ‘blunted’. There has been very little work to date that has tried to link sensory behaviours to perceptual correlates. Minshew and Hobson (2008) found no association between measures of tactile sensory perception and self-report of tactile sensitivities in children and adults with high functioning ASD. However, the auditory domain remains unexplored. Thus, the second aim of our study was to investigate the association between auditory discrimination sensitivity and self-reported auditory sensory behaviours in adolescents with ASD.

Method

The study was approved by the South East Research Ethics Committee (05/MRE01/67).

Participants

Seventy-two adolescents with an ASD (mean age = 15 years 6 months, SD 5.7 months) and 48 adolescents without an ASD (mean age = 15 years 6 months, SD 5.9 months) were tested.

The 72 participants with an ASD (39 childhood autism; 33 other ASD) and 22 of the participants without an ASD were recruited from the Special Needs and Autism Project cohort (SNAP; Baird et al., 2006). For this cohort, consensus clinical ICD-10 diagnoses were made by 3 experienced clinicians (GB, ES, TC) using information from the ADI-R (Lord, Rutter, & Le, 1994) and ADOS-G (Lord et al., 2000) as well as IQ, language and adaptive behaviour measures (see Baird et al., 2006 for details). Of the 33 cases meeting criteria for ‘other ASDs’; 3 met ICD-10 criteria for ‘atypical autism’ due to late onset; 28 met ICD-10 criteria for ‘other pervasive developmental disorder’ due to sub-threshold symptomatology and 2 met ICD-10 criteria for ‘pervasive developmental disorder unspecified’ due to lack of information (incomplete assessment, adopted children for whom early history was not available). The 22 participants assigned to the non-ASD group were adolescents who had a statement of special educational needs (the UK term for children with identified learning and/or behavioural problems in school) at age 9–10 years but who did not reach clinical criteria for an ASD (Baird et al., 2006). Rather, they had a range of primary ICD-10 diagnoses (13 mild intellectual disability; 2 moderate intellectual disability; 3 specific reading/spelling disorder; 2 AD/HD; 1 expressive/receptive language disorder; 1 no diagnosis).

The remaining non-ASD participants ($n = 26$) were recruited from local mainstream schools. It was confirmed through parent and teacher report that all 26 individuals were typically developing; none had a psychiatric or developmental diagnosis, a statement of special educational needs or were receiving medication. The social communication questionnaire (SCQ; Rutter, Bailey, & Lord, 2003) was collected from 22 of the 26 adolescents; no individual scored 15 or above, the cut-off for ASD. Measures of IQ were obtained using the Wechsler Abbreviated Scale of Intelligence-UK (WASI; Wechsler, 1999), with full scale IQ of the total cohort ranging from 52 to 133. There were no group differences between the ASD and non-ASD participants for age or IQ (t -test, all $p > .10$; see Table 1).

Design and procedure

Hearing screen

The 120 participants completed either an audiologist-led hearing screen in a hospital clinic or, if unavailable, a researcher-led screen using a portable audiometer. Fifty-three of the participants (10 non-ASD; 43 ASD) were assessed by an audiologist in a purpose built sound-proofed room on a battery of three tests of hearing function: pure tone audiometry to assess whether hearing levels were within the normal range; otoacoustic emissions to assess cochlear

function; tympanometry to assess middle ear function. Audiometric thresholds were established using Test Method A as outlined by the British Society of Audiology (1981). A threshold score of 20 dB had to be achieved between 500 and 4000 Hz, i.e. within normal speech limits. To pass the hearing screen, participants had to pass all three measures. For some individuals travelling to the assessment centre was not possible and they were seen at school, home or at an alternative testing location. Therefore, for the remaining 67 participants (38 non-ASD; 29 ASD) a portable audiometer (Earscan 3) was used by a trained researcher in the school or home environment, in the quietest room available. Thresholds were ascertained in an identical manner, but the absence of a sound-proofed environment meant that the pass threshold was lowered to a score of 30 dB between 1000 and 4000 Hz. It was not possible to measure cochlear or middle ear function in these individuals. All 120 participants passed the hearing screens, which was a condition of being included in the study. An additional 20 participants (4 non-ASD (2 clinical assessment, 2 portable assessments); 16 ASD (9 clinical assessments, 5 portable assessments)) failed the screen and have not been reported in this paper.

Auditory discrimination tasks

Each participant completed three tasks of auditory discrimination: frequency discrimination, intensity discrimination and duration discrimination, in a random order over 2 days of testing (interspersed with other tasks). The lag between the two testing sessions averaged at 28 days (SD: 33 days). All three tasks followed the same format, a two forced choice procedure, using the “Dinosaur” software programme created by Dorothy Bishop (Oxford University). The participant was shown two cartoon dinosaurs, one standing on a red box and one standing on a yellow box, presented on a sound-calibrated Hewlett Packard laptop with a 15 in. LCD display screen. They were told that each dinosaur would make a “funny sound”, and that their job was to decide which dinosaur makes a “higher” (frequency discrimination), “louder” (intensity discrimination) or “longer” (duration discrimination) sound respectively. The dinosaurs always made their sounds in the same order (left (yellow box) dinosaur, followed by right (red box) dinosaur), with a circle appearing around the dinosaur to denote which one was making a noise. The participants were told to indicate their response by pointing to the dinosaur, or calling out the colour of its box. The experimenter then inputted the response, which initiated visual feedback (a cartoon icon appearing and remaining in the left hand side of the screen if correct, a black cross temporarily appearing in the dinosaur’s box if incorrect) alongside congruent auditory feedback (a positive sounding noise vs. a “sighing” sound).

Each task started with a five-trial practice, to ensure that the participant understood the instructions. The participant wore headphones (Sennheiser HD 280 pro) for both the tasks and practice trials.

The parameters of the tasks are listed in Table 2. In each dinosaur pairing, the participant was presented with one ‘standard’ stimulus (randomly assigned to either dinosaur on each trial), which did not change across the particular task, and a probe stimulus that varied according to an adaptive procedure based on the participant’s current performance (more virulent PEST: Parameter Setting by Sequential Estimation, Findlay, 1978). PEST begins with presentation of the standard stimulus and the probe stimuli that is most different from the standard. Probe stimuli vary in equal log increments and can be perceived as a ladder of sounds between the standard stimuli and starting probe; all stimuli were .wav files. PEST uses a 2up1down procedure (after 2 correct trials the perceptible difference between the two stimuli reduces; after 1 incorrect trial the perceptible difference between the two stimuli is increased) to determine the threshold at which the participant is correct on 75% of trials. The task is terminated after 6 reversals (change in direction in the 2up1down procedure) or after 40 trials, and the final threshold score is the mean threshold value from the 4th reversal.

Each stimulus was labelled by the numeric code given to its .wav file (0.wav as the standard, increasing to 1.wav, 2.wav for the probes, and so on). Threshold scores were calculated from the .wav values, generating a mean .wav score since the 4th reversal, where the closer the score is to 0 (i.e. 0.wav), the better the performance. Translating these mean values to Hz, dB or ms values loses sensitivity (i.e. rounding the mean .wav value up or down to the nearest integer). Thus, the data were analysed based on the .wav values. However, we also included translated scores in our Section 2 to aid interpretation of the data.

Self report of auditory sensory behaviours

The adolescents who were able to complete self-report measures were given the Adolescent/Adult Sensory Profile (AASP: Brown & Dunn, 2002). 45 individuals with ASD and 27 individuals without an ASD returned completed questionnaires. Four adolescents missed out items, meaning auditory sensory behaviour scores could not be calculated for one or more quadrants. As we were interested in statistically comparing across the quadrants, these individuals were excluded, so analysis was confined to 42 individuals with an ASD (from a total of 72 that did the auditory perceptual tasks) and 26 individuals without an ASD (from a total of 48 that did the auditory perceptual tasks). The mean IQ scores were

calculated for these groups who, because of the demands of the questionnaire, were at the higher end of the ability range. For the non-ASD group they were: verbal IQ = 95.9 (13.0); performance IQ = 104.4 (14.5); full scale IQ = 100.1 (14.5) (SD in brackets). For the ASD group they were: verbal IQ = 91.4 (13.2); performance IQ = 102.1 (11.8); full scale IQ = 96.4 (11.4). Independent *t*-tests confirmed that there was no significant difference between the groups ($p > .10$).

The AASP is divided into quadrants, each reflecting a different combination of an individual's neurological threshold and style of behavioural response. The scores on the auditory items for each quadrant were summed to give four separate measures of auditory sensory behaviours. The low registration quadrant (item52: having trouble following conversation, 55: not noticing when your name is called, 59: asking people to repeat things) and sensation seeking quadrant (50: making noises such as humming, singing or whistling, 58: seeking events with music) both reflect a high neurological threshold for noticing and responding to stimuli. Low registration behaviours denote a passive behavioural response of acting in accordance with the neural underresponsivity, whereas sensation seeking behaviours denote an active behavioural response of pursuing or creating stimuli to try and counteract the high threshold. The sensory sensitivity quadrant (51: a strong startle response, 54: distracted by noise, 60: difficulty with background noise) and sensation avoiding quadrant (53: leaving the room when the TV is on or turning down the volume, 56: strategies to drown out sound such as hands over ears, 57: avoiding noisy environments) both reflect a low neurological threshold for noticing and responding to stimuli. Sensory sensitivity denotes a passive behavioural response to this neural overresponsivity, whereas sensation avoiding denotes an active behavioural response of reducing and preventing exposure to sounds. The AASP is scored on a 5 point scale, with a high score indicating a high level of sensory behaviours.

Results

Auditory discrimination thresholds

For reasons of time constraints, two participants in the ASD group completed only two of the tasks and one participant from both groups only completed one task. As a result, the frequency discrimination task was completed by ASD = 71 and non-ASD = 47, the intensity discrimination task by ASD = 70 and non-ASD = 47 and the duration discrimination task by ASD = 71 and non-ASD = 48. The threshold data (using .wav file values) for the three tasks

were not normally distributed. A log transformation (to base 10) normalised the duration discrimination and intensity discrimination data, but did not normalise the frequency discrimination data. For simplicity, all subsequent data are presented on log-transformed scores.

The data were tested for significant group effects by comparing means, using independent-samples *t*-tests for the intensity and duration discrimination tasks and a non-parametric Mann–Whitney *U* test for the frequency discrimination task. None of the tests was significant ($p > .20$), indicating that the ASD and non-ASD groups did not differ in their auditory discrimination ability. The mean scores as log transformed .wav values and translated Hz, dB and ms values are presented in Table 3.

There has been some discussion in the literature that enhanced frequency discrimination in ASD may be confined to a subgroup (Heaton et al., 2008). Given the heterogeneity of the disorder, we investigated whether the mean scores were masking a small subgroup of exceptionally good discriminators in the ASD group. As a low threshold score equates to better performance, the criterion for exceptional performance was a threshold score that was more than 1.65 SD below the mean of the control group (i.e. corresponding to the 5% percentile). For the frequency task, 14 individuals in the ASD group (19.7% of the ASD group) performed >1.65 SD below the control mean (range -1.67 to -2.83), compared to 4 individuals in the non-ASD group (8.5% of the non-ASD group; range -1.68 to -2.07). This difference in distribution approached significance (χ^2 (df = 1) = 2.75; $p = .097$). The size of the subgroups became more polarised when a stricter threshold of 2 SD was used, with 8 individuals in the ASD group performing >2 SD below the control mean (11.3% of the ASD population), compared to 1 individual in the non-ASD group (2.1% of the non-ASD population) (χ^2 (df = 1) = 3.35; $p = .067$). Of note, there were two further cases in the ASD group with exceptional frequency discrimination scores: one individual ($Z = -2.27$) did not have his hearing screened and the other ($Z = -2.07$) failed the portable audiometric assessment, hence both were excluded from the current analysis. For the intensity discrimination task, 3 individuals (4.3% of the ASD group) performed >1.65 SD below the control mean (all at $Z = -2.44$), whilst 3 individuals in the non-ASD group (6.4% of the non-ASD group) performed similarly well (range -1.78 to -2.44). This difference in distribution was not significant ($p > .6$). For the duration discrimination task, 10 individuals (14.1% of the ASD group) performed >1.65 SD below the control mean (all at $Z = -1.90$), whilst 4 individuals in the non-ASD group (8.3% of the non-ASD group) performed similarly

well (all at $Z = -1.90$). This difference in distribution was not significant ($p > .3$) and, unlike the other two ASD subgroups, no one performed 2 SD from the control mean. The overlap between individuals who excelled at the different types of discrimination task was limited. Twenty-two individuals in the ASD group fell into at least one subgroup but only four of these showed exceptional skill in more than one task (2 showing exceptional frequency and duration discrimination, 1 showing exceptional frequency and intensity discrimination and 1 showing all three types of exceptional ability). Ten individuals without ASD fell into at least one subgroup and only one of these showed exceptional skill in more than one task (exceptional frequency and intensity discrimination).

Although not the focus due to our a priori predictions, there were three individuals in the ASD group who showed exceptionally poor performance (i.e. 1.65 SD above the control mean) on the intensity discrimination task (range 1.68–1.83) and four individuals in the ASD group who showed exceptionally poor performance on the duration discrimination task (range 1.67–1.95). All other individuals (ASD and non-ASD) performed within 1.65 SD.

Characteristics of the subgroup with exceptional frequency discrimination and ASD

For the participants with ASD, those with enhanced frequency discrimination were the most notable subgroup and we were motivated to explore their characteristics further. Particularly, we were interested in diagnosis, intellectual ability and language development (see Table 4 for a summary). Eight individuals within the subgroup had strictly defined autism (21.1% of the 38 with autism) compared to 6 with other ASD (18.2% of the 33 with other ASD). Using Chi-square analysis, the difference in distribution of diagnoses between those with and without exceptional frequency discrimination was not significant ($p > .8$). Only one individual in the enhanced frequency discrimination subgroup had a full scale IQ below average (i.e. outside of 1 SD from the mean). Splitting the ASD group in to those with a full scale IQ either ≥ 80 (high IQ) or < 80 (low IQ), we found that thirteen individuals with a high IQ (27.1% of the 48 with a high IQ) had exceptional frequency discrimination compared to 1 individual with a low IQ (4.3% of the 23 with a low IQ). This difference in distribution was statistically significant ($\chi^2 (df = 1) = 5.07; p = .024$), suggesting that the incidence of exceptional frequency discrimination is more common amongst more intellectually able individuals. Using ADI criteria for defining delayed onset of first single words (> 24 months), 8 individuals with delayed first words (33.3% of the 24 with delayed first words) had exceptional frequency discrimination compared to 6 without delayed first words (13.0% of

the 46 without delayed first words). This difference in distribution was statistically significant (χ^2 (df = 1) = 4.06; $p = .044$), suggesting that the incidence of exceptional frequency discrimination is more common amongst those with delayed first words. Notably, diagnosis, FSIQ and presence of delayed first words failed to distinguish the ASD subgroups with and without enhanced discrimination in the intensity and duration domains (all $p > .7$).

Auditory sensory behaviours

The data are presented in Fig. 1. A mixed factorial ANOVA was used to statistically test the data: 4 (AASP auditory quadrant) \times 2 (group). A main effect of AASP auditory quadrant (F (2.62, 172.57) = 12.48; $p < .001$) illustrates that mean scores differed across quadrants, which is to be expected as the sensation seeking quadrant only had two items (max. score = 10) and the other three quadrants had three items each (max. score = 15). The significant main effect of group (F (1, 66) = 10.71; $p = .002$) illustrates that the degree of self-reported auditory sensory behaviours was higher in the ASD group. Also significant was the quadrant by group interaction (F (2.62, 172.57) = 12.91; $p < .001$), reflecting that the non-ASD group reported more auditory sensory behaviours in the sensation seeking quadrant compared to the ASD group, whilst the ASD group reported more behaviours in the other three quadrants. Post hoc independent t tests were used to explore the significance of group differences in each quadrant (N.B. with a Bonferroni corrected p threshold of 0.017). These confirmed that the ASD group reported significantly more behaviours in three quadrants (low registration (t (66) = 4.85; $p < .001$); sensory sensitivity (t (66) = 2.67; $p = .010$); sensation avoiding (t (66) = 3.45; $p = 0.001$). The non-ASD group reported significantly more auditory behaviours in the sensation seeking quadrant (t (66) = -2.83; $p = 0.006$). The results of the sensation seeking quadrant were unexpected. However, closer inspection of the items (“I hum, whistle, sing, or make other noises” and “I like to attend events with lots of music”) suggest that they may be confounded by sociability and social interest, which would disadvantage the ASD group, particularly in the adolescent age-range tested. As we question whether the sensation seeking quadrant is truly reflecting auditory sensory behaviours for this particular population, we have excluded it from subsequent analyses.

Association between auditory discrimination thresholds and auditory sensory behaviours

The participants with ASD who completed the AASP were divided into two subgroups based on their auditory discrimination scores using a mean split method. For the frequency discrimination task, the mean score was 0.83 (SD 0.49), with 19 individuals scoring

Below the mean and performing relatively well ('low threshold' scorers) and 23 individuals scoring above the mean and performing relatively poorly ('high threshold' scores). For the intensity duration task, the mean score was 0.80 (SD 0.50), with 18 below the mean and 23 above. For the duration discrimination task, the mean score was 0.60 (SD 0.35), with 19 below the mean 23 above. Using this division we ran a 3 (AASP auditory quadrant)×2 (high or low discrimination subgroup) mixed factorial ANOVA for each task (see Fig. 2). The frequency discrimination task showed a significant main effect of AASP quadrant ($F(2, 80) = 7.68; p = .001$) but no significant main effect of subgroup or subgroup by quadrant interaction. Thus, frequency discrimination ability is not associated with degree of auditory sensory behaviours in ASD. The intensity discrimination task showed a main effect of AASP quadrant ($F(2, 78) = 10.15; p < 0.001$) but no significant effect of subgroup. However, there was a significant quadrant by subgroup interaction ($F(2, 78) = 3.14; p = .049$), reflecting minimal difference between low and high threshold scorers on the low registration quadrant, compared to the poor performers displaying more sensory sensitivity and sensation avoiding behaviours than the good performers. Post hoc t tests showed that this difference was significant for the sensation avoiding quadrant ($t(39) = 2.34; p = .024$), although this is below significance at the Bonferroni threshold of 0.05/3. Thus, the association between intensity discrimination and auditory sensory behaviours in ASD depends on whether the behaviour is low registration or sensory sensitivity and sensation avoiding. The duration discrimination task showed a main effect of AASP quadrant ($F(2,80) = 7.73; p = .001$). There was also a main effect of subgroup ($F(1,40) = 6.69; p = .013$), indicating that those who showed more proficient auditory duration discrimination abilities self reported more auditory sensory behaviours. There was no significant AASP quadrant by subgroup interaction.

Discussion

This study is the first to investigate auditory discrimination ability in ASD across a range of parameters and to explore the functional impact of auditory discrimination sensitivity. Seventy two adolescents with an ASD and representing a wide range of IQs were assessed, making it by far the largest exploration of auditory discrimination ability to date. Whilst at a group level auditory discrimination abilities were not different in individuals with ASD compared to controls, enhanced auditory discrimination was present in around 1 in 5 individuals with ASD in the frequency domain. Further, these individuals were characterised by average intellectual ability and delayed language, suggestive of a specific phenotype. In

addition, we found that auditory duration and intensity discrimination ability related to the degree of self-reported auditory sensory behaviours in ASD.

Atypical auditory frequency discrimination, but not intensity or duration discrimination, in ASD

We did not find a difference in frequency discrimination performance between the ASD and non-ASD participants at the group level, unlike a previous study with a much smaller sample ($n=12$) and a different mode of presenting and analysing the data (Bonnell et al., 2003). In particular, our protocol gave strategic support (via the stepwise procedure, which gets easier if the individual is struggling) and feedback, which was not present in the Bonnell et al. (2003) study. O’Riordan and Passetti (2006) also report evidence of enhanced sensitivity for detecting differences in pitch in ASD. Their paradigm involved pressing a button when two continually repeating tones became indistinguishable in pitch, i.e. a longer response indicated greater sensitivity for detecting differences in pitch. However, slower reaction time is a well-documented phenomenon in ASD (e.g. Schmitz, Daly, & Murphy, 2007), so the data could also be confounded by general effects of slower cognitive processing/decision making.

The heterogeneity of ASD means that it is circumspect to consider the possibility of subgroups and there is a precedent for this within the perceptual literature (e.g. Heaton et al., 2008; Milne et al., 2006). Of note, Heaton et al. (2008) found a small subgroup of adolescents (3 out of 32) with ASD who were superior (4–5SDs above the mean) at judging pitch distance (deciding how ‘far’ a tone was from a standard using a learnt spatial scale), despite no overall group difference between individuals with and without ASD. The cut-off for exceptional performance on the frequency discrimination task in our own study was defined as 1.65 SD from the control mean performance. We found evidence of enhanced auditory processing of the frequency component of sound in a subgroup of fourteen individuals on the autism spectrum (19.7% of ASD group) compared to a similar performance in four individuals (8.5%) in the non-ASD group, although this missed statistical significance ($p = .097$). This suggests that around 1 in 5 individuals with ASD may exhibit exceptional frequency discrimination skills. Notably, the subgroups became more disparate in size when the more conservative threshold of 2 SD was used (11.3% of the ASD group compared to 2.1% of the non-ASD group). In addition, a further two cases in the ASD group had exceptional frequency discrimination but were not included in the analysis due to either

not completing or failing an audiometric assessment, suggesting we may have under-reported the true size of our subgroup. We therefore find our data supportive of enhanced frequency discrimination being present in a meaningful subgroup of individuals on the autism spectrum. The percentage of performers with exceptional intensity and duration discrimination ability was not significantly different across groups and for duration discrimination did not include any individuals over the 2 SD threshold. Additionally, the subgroups of enhanced performers were largely mutually exclusive. Proponents of the enhanced perceptual functioning model (Mottron & Burack, 2001; Mottron, Dawson, Soulieres, Hubert, & Burack, 2006) argue that low level perceptual processing is overdeveloped in ASD. As the first study to explore perceptual discrimination in ASD across a range of parameters, we conclude that enhanced auditory perceptual processing is a specific effect within the frequency domain.

Compared to the remainder of individuals with ASD, the fourteen individuals with ASD and with enhanced frequency discrimination were significantly more likely to have a higher IQ ($FSIQ \geq 80$) and to have delayed onset of first words. There was no difference in severity of diagnosis (autism vs. other ASD). Congruent with this, the two previous studies showing enhanced perceptual processing in ASD at the group level both assessed high functioning individuals (Bonnell et al., 2003; O’Riordan & Passetti, 2006). Like us, Heaton et al. (2008) tested across the range of intellectual ability and two of their three exceptional outliers were high functioning. Recently, it has been suggested that an over-focus on perceptual cues, particularly pitch, during speech negatively impacts upon linguistic processing (Jarvinen-Päsley, Pasley, & Heaton, 2008a; Jarvinen-Päsley, Wallace, Ramus, Happé, & Heaton, 2008b). As far as we are aware, this is the first study to directly demonstrate a behavioural association between language delay and enhanced auditory perceptual processing in ASD, although this association has been previously explored and confirmed in the visual domain using the block design paradigm and age of onset of first word and first phrase (Mottron, Soulieres, Meilleur, & Dawson, 2008). It remains to be determined whether the frequency sensitivity of our subgroup interfered with their language development or whether this was due to other factors. Neural responses to frequency change in speech sounds are atypical in ASD, with enhanced responses at the pre-attentive (MMN) level but diminished involuntary attention switching (P3a) to frequency changes in speech sounds (Lepistö et al., 2005; Lepistö et al., 2006). This could be interpreted as enhanced automatic detection of frequency changes in speech in tandem with reduced significance being appropriated to speech sounds. Recently, it has been proposed that homogeneous

cognitive and behavioural subgroups (as compared to the heterogeneous broader group of individuals with ‘the autisms’ who happen to meet current clinical diagnostic criteria) might be useful in uncovering the genetic basis of this range of neurodevelopmental disorders (Abrahams & Geschwind, 2008). Our data suggests that those with enhanced frequency discrimination may represent such a phenotype and this is given more credence by the lack of similar defining characteristics for individuals with enhanced intensity or duration discrimination. Further work is needed to delineate why these particular features hang together in certain individuals on the autism spectrum.

What also remains unclear is why frequency is the singular aspect of sound where individuals with ASD can show exceptional ability. The answer may be related to fundamental differences in how different aspects of sound are processed and represented at the neurophysiological level. There is ample evidence that the frequency of sound is represented in a tonotopic organisation in the auditory cortex (e.g. Lockwood et al., 1999; Talavage et al., 2004) and suggestion that the intensity of sound may be represented in the auditory cortex in a separable, amplitopic manner (Lockwood et al., 1999). Perhaps the most established finding for sound intensity is that the extent of neural activation in auditory cortex is closely coupled with the level of intensity (e.g. Jäncke, Shah, Posse, Grosse-Ryken, & Müller-Gärtner, 1998). Distinct from these, the basal ganglia and associated dopaminergic system have been implicated in temporal processing, including duration discrimination (e.g. Coull, Nazarian, & Vidal, 2008; Jahanshahi, Jones, Dirnberger, & Frith, 2006; Rao, Mayer, & Harrington, 2001). Indeed, performance on frequency and duration discrimination tasks have been dissociated from each other in patient groups (e.g. Harrington, Haaland, & Hermanowicz, 1998; Tramo, Shah, & Braida, 2002). There is no understanding of how these separable neural responses for each of the sound parameters might be different (or similar) in individuals with ASD, both with and without enhanced frequency discrimination. Research to date has generally focused on ERPs, but with limited comparison across frequency, intensity and duration within the same individuals, and none in pre-identified subgroups with superior auditory perception. Investigations of frequency sensitivity typically report enhanced pre-attentive neural responses in ASD (e.g. Ferri et al., 2003; Gomot et al., 2002; Kujala et al., 2007; Lepistö et al., 2005), but there is no evidence of enhanced MMN to intensity changes (Kujala et al., 2007) and mixed reports of both enhanced (Kujala et al., 2007) and diminished MMN responses to duration change (Lepistö et al., 2005; Lepistö et al., 2006). One theoretical avenue that could be borne in mind is the distinction made between the processing of spectrally and temporally ‘simple’ vs. ‘complex’

stimuli in ASD and how this hypothesis fits with the findings of the current study (e.g. Bertone, Mottron, Jelenic, & Faubert, 2005; Groen et al., 2009; Samson et al., 2005).

Enhanced pitch processing in ASD has also been established during the processing of complex sequences of notes and of chords, which has been related to pitch memory and labelling abilities (e.g. Heaton et al., 1998; Heaton, 2003, but see Altgassen, Kliegel, & Williams, 2005 for a non-replication) and to the capacity for segmenting and perceiving perceptual units as distinct from the gestalt (e.g. Foxton et al., 2003), rather than reflecting increased sensitivity to aspects of sound per se. When drawing conclusions about the presence of enhanced perceptual ability, it is important to distinguish perceptual sensitivity from overarching processing styles that may facilitate performance, particularly in more elaborate tasks. Our study suggests that enhanced frequency discrimination affects only a subgroup of individuals in ASD, but superior performance may be more widespread when the task employs additional layers of perceptual or cognitive processing. Notably, Foxton et al. (2003) found that their ASD group were not superior to controls on a simple task of pitch discrimination (using a train of five notes, where the probe sequence was either the same as the standard or differed in the pitch of one note), despite showing an atypical lack of susceptibility to gestalt interference effects during more complex sequences. Also, Jarvinen-Päsley and Heaton (2007) report no difference in ability between children with and without ASD on a task requiring same/different pitch discrimination of four-tone musical sequences. However, when one or both stimuli were four-syllable words, the performance of the ASD group was impervious to the typical profile of relatively degraded performance. This suggests an over-focus of attention towards simple perceptual information and resilience to the distracting effect of linguistic content. Further research comparing performers with ‘enhanced’ and average basic frequency perception on more complex tasks of pitch processing may help disambiguate the relative influence of bottom-up enhanced perceptual sensitivity and cognitive factors such as memory, disembedding ability and weak central coherence (e.g. see Happé & Frith, 2006).

The association between auditory discrimination ability and auditory sensory behaviours

There has been a large body of work detailing parent and caregiver report of increased levels of sensory behaviours in ASD compared to comparison populations (e.g. Baranek et al., 2006; Kern et al., 2006; Kientz & Dunn, 1997; Tomchek & Dunn, 2007; Watling et al., 2001). We demonstrate a significantly greater degree of *self-reported* auditory sensory behaviours in ASD. This concurs with two recent studies that have demonstrated that high

functioning children and adults with ASD self-report more sensory sensitivities and sensory behaviours, collapsed across a range of modalities, than those without ASD (Crane, Goddard & Pring, 2009; Minshew & Hobson, 2008).

The causes of atypical sensory behaviours in ASD remain poorly understood. We hypothesised that auditory discrimination ability would relate to the degree or pattern of self-reported auditory sensory behaviours and found that this association held true for duration and intensity discrimination, but not for frequency discrimination. As the ASD group reported a significantly greater degree of auditory sensory behaviours but did not differ in their intensity or duration discrimination ability, then a simple association or causal link between the ability to discriminate sounds and the expression of auditory sensory behaviours is unlikely. However, auditory discrimination proficiency could serve as a ‘risk factor’ for auditory sensory behaviours by modulating the degree to which sounds are detected or missed in the environment, which are then subject to atypical physiological or psychological response. This hypothesis precludes the alternative interpretation that sensory behaviours affect the ability to discriminate sounds (e.g. avoidance behaviours could limit refinement of a discrimination system). The direction of the relationship between auditory discrimination ability and auditory sensory behaviours could be explored using a developmental study that charts the emergence of auditory sensory behaviours alongside auditory discrimination proficiency.

For the intensity task, there was no difference in discrimination ability for individuals with both high and low levels of low registration behaviours, which reflect a difficulty in perceiving relevant sounds. However, for both quadrants that implicate a hypersensitised system and an aversive reaction to noise (sensory sensitivity and sensation avoiding), those with poor intensity discrimination skills reported more auditory sensory behaviours. This pattern was demonstrated in the significant interaction between intensity discrimination subgroups and AASP quadrant. Intensity modulation is the process by which sensory responses to incoming sounds of different intensity are regulated and Bruneau, Bonnet-Brilhault, Gomot, Adrien, and Barthélémy (2003) and Bruneau, Roux, Adrien, and Barthélémy (1999) have demonstrated absence of the typical modulation of the N1 waveform to sounds of increasing intensity in the left hemisphere of children with ASD. Bruneau et al. (2003) failed to establish a link between the amplitude of the N1c and a caregiver-rated observation of unusual responses to auditory stimuli, although they discuss that their observation item may have been too broad and, further, there was limited variability in the possible scores (1–5). An inability to modulate incoming intensity levels could not only mean

relatively poor discrimination ability (the result of a less finely tuned system) but could mean that behavioural reactions to sounds are augmented due to the up-stream implications of a threshold mechanism that does not adequately filter and sort (and therefore make adaptive sense of) incoming stimuli.

For the duration task, the more sensitive someone with ASD is to discriminating duration, the more auditory sensory behaviours they report across the three domains of low registration, sensory sensitivity and sensation avoiding, which are united by describing a behavioural reaction to a difficulty with processing sound. This is the reverse pattern to that found for intensity discrimination. As previously mentioned, it is important to remember that intensity, duration and frequency are distinct domains of perceptual processing with their own neural profile. Duration discrimination is also distinct as successful performance requires the individual to attend to the entire length of both intervals (see Casini & Ivry, 1999). Judgment of the frequency and intensity of sounds does not require this type of sustained attention and the length of the frequency and intensity sounds in this particular study were much shorter. Liss, Saulnier, Fein, and Kinsbourne (2006) have found that overreactive responses (e.g. negative reactions to noisy environments; covering eyes in visually complex environments), sensory seeking responses (e.g. seeks loud environments; waves fingers over eyes) and the overfocusing of attention cluster together in 43% of their sample with ASD, with those with the lowest sensory scores also showing the lowest levels of attentional overfocusing. Performance on the duration discrimination task could be tapping into a particular type of attentional vigilance that correlates with sensory behaviours, which suggests that duration discrimination ability may reflect rather than influence the auditory sensory experience.

Temporal processing of brief durations is modulated by the sensory qualities of a stimulus such as the modality of presentation (e.g. Wearden, Edwards, Fakhri, & Percival, 1998) and the stimulus content of the interval being timed (e.g. Pfeuty, Ragot, & Pouthas, 2008). Related to this, duration judgements of stimuli with a strong emotional valence are modulated by the physiological arousal that they induce (e.g. Droit-Volet, Brunot, & Niedenthal, 2004; Noulhiane, Mella, Samson, Ragot, & Pouthas, 2007). Speculatively, for individuals who are sensitive to sounds, increased physiological arousal could lead to heightened attention and subsequently superior performance (see Easterbrook, 1959). Low registration concerns difficulties in noticing pertinent stimuli; thus, you would expect a negative relationship with a task that was indexing arousal mediated attention. However, the quadrant specifically concerns noticing, attending to or following communication from other

people. One explanation could be that low registration responses are the result of a ‘shut down’ or disengagement from human auditory cues, following auditory perceptual overload from the environment (i.e. an extreme behavioural response to aversive heightened arousal) (e.g. Kootz, Marinelli, & Cohen, 1982). Although arousal hypotheses of sensory behaviours have received mixed support (see Rogers & Ozonoff, 2005), Hirstein, Iversen, & Ramachandran (2001) described two types of individual with ASD: type A (the majority) had excessively high skin conductance responses (i.e. hyperarousal) that stopped when engaged in self-stimulating behaviours and type B had either no skin conductance responses or responses that were only elicited by extreme self-stimulatory behaviour. The commonality to both groups was that the sensory behaviours were used to control an arousal system that was not well regulated. This study suggests a need for ambitious research that can accommodate a large number of participants and therefore have the capacity to explore heterogeneity and to use target stimuli that are pertinent to a particular child or adult’s sensory behaviour profile.

We did not find evidence that frequency discrimination ability related to reported auditory sensory behaviours. However, the auditory sensory behaviours that the individuals were questioned about speak mainly to difficulties with loud noises and with not noticing relevant stimuli. Further work needs to establish whether individuals with exceptional frequency discrimination ability exhibit behaviours that more closely reflect their finely tuned frequency processing (e.g. interest in sounds within a particular frequency spectrum; particularly positive or negative response to certain types of music; strong reactions to very specific sounds that are not based on loudness). We predict that behaviours related to music may be particularly prevalent given that pitch is the aspect of sound that seems to underpin the capacity for the appreciation and recognition of music (e.g. Hyde & Peretz, 2004).

Summary

This study found that differences in auditory discrimination are not characteristic of most individuals with ASD. However, auditory frequency discrimination is enhanced in a subgroup of individuals with ASD that share particular defining features, suggestive of a specific phenotype. Further work is now needed to discern why frequency processing is ‘special’ in ASD. Research has generally focussed on a unifying explanation for the expression of sensory behaviours. However, a recent population-based toddler twin study suggests that different genetic factors underpin sensory behaviours in different modalities

(Goldsmith, Van Hulle, Arneson, Schreiber, & Gernsbacher, 2006). Future research could benefit from further unpacking of the domain-specific processes that may contribute to atypical sensory experiences. Exploring the genesis of sensory behaviours in ASD is important in terms of understanding behaviours that can have a negative functional impact, but also in terms of establishing how the neural markers of these behaviours contribute to the expression (or development) of an autistic profile. This is the first study to explore the link between auditory perceptual processing and auditory sensory behaviours and has demonstrated the potential value in this approach. At this stage, interpreting the nature of this association is speculative and further investigation is necessary. In particular, more finely targeted measures of auditory sensory behaviours and the direct testing of more specific hypotheses would be of benefit.

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Table 1: Mean age (years; months), verbal IQ, performance IQ and full scale IQ (SD in brackets) for the non-ASD and ASD groups.

	Non ASD	ASD
n	48	72
Male:Female	46:2	66:6
Age	15;6 (5.9)	15;6 (5.7)
Verbal IQ	87.25 (19.70)	84.31 (17.41)
Performance IQ	92.83 (21.15)	93.36 (17.44)
Full scale IQ	89.33 (21.53)	87.79 (17.32)

Table 2: Parameters of the auditory discrimination tasks

	Frequency	Intensity	Duration
Standard stimuli	600 Hz	73 dB	640 ms
Starting probe	982 Hz	81.1 dB	400 ms
Difference between probes	10 Hz (2 Hz on lowest probe)	0.27 dB	8 ms
Frequency	variable	800Hz	500Hz
Intensity	73 dB	variable	73dB
Duration	25 ms	150 ms	variable
Total probe stimuli	39	30	30
ISI	500 ms	480 ms	480 ms

Table 3: Mean values for the auditory discrimination tasks (SD in brackets)

	Non-ASD	ASD
Threshold score: log transformed		
Frequency	1.12 (0.40)	1.02 (0.48)
Intensity	0.82 (0.34)	0.88 (0.31)
Duration	0.72 (0.38)	0.70 (0.40)
Threshold score: transformed into difference from standard		
Frequency (Hz)	174.43 (122.17)	154.96 (119.16)
Intensity (dB)	2.34 (1.60)	2.58 (1.65)
Duration (ms)	59.33 (44.43)	58.82 (52.08)

Table 4: Characteristics of the 14 individuals with ASD and exceptional frequency discrimination: diagnosis, verbal IQ (VIQ), performance IQ (PIQ), full scale IQ (FSIQ), onset of first words in months (1st words)

	Diagnosis	VIQ	PIQ	FSIQ	1st words
1	Autism	77	110	92	44*
2	Autism	80	119	98	40*
3	Autism	86	115	100	48*
4	Autism	112	107	111	18
5	Autism	100	106	104	48*
6	Autism	97	109	103	14
7	Autism	109	114	113	15
8	Autism	99	110	106	40*
9	Other ASD	60	55	54	24
10	Other ASD	77	119	97	15
11	Other ASD	120	106	115	10
12	Other ASD	99	96	99	30*
13	Other ASD	103	115	109	30*
14	Other ASD	88	93	88	30*
	Mean (SD)	94.3 (15.6)	105.3 (15.7)	99.2 (14.6)	29.0 (13.3)

*delayed first words (>24 months), according to ADI criteria

Figure 1: ASD group: mean scores (and standard error) for auditory items on the Adolescent/Adult Sensory profile, grouped into quadrants. A higher score indicates more self-reported auditory sensory behaviours. The maximum score on sensation seeking is 10; the maximum score on the other three items is 15.

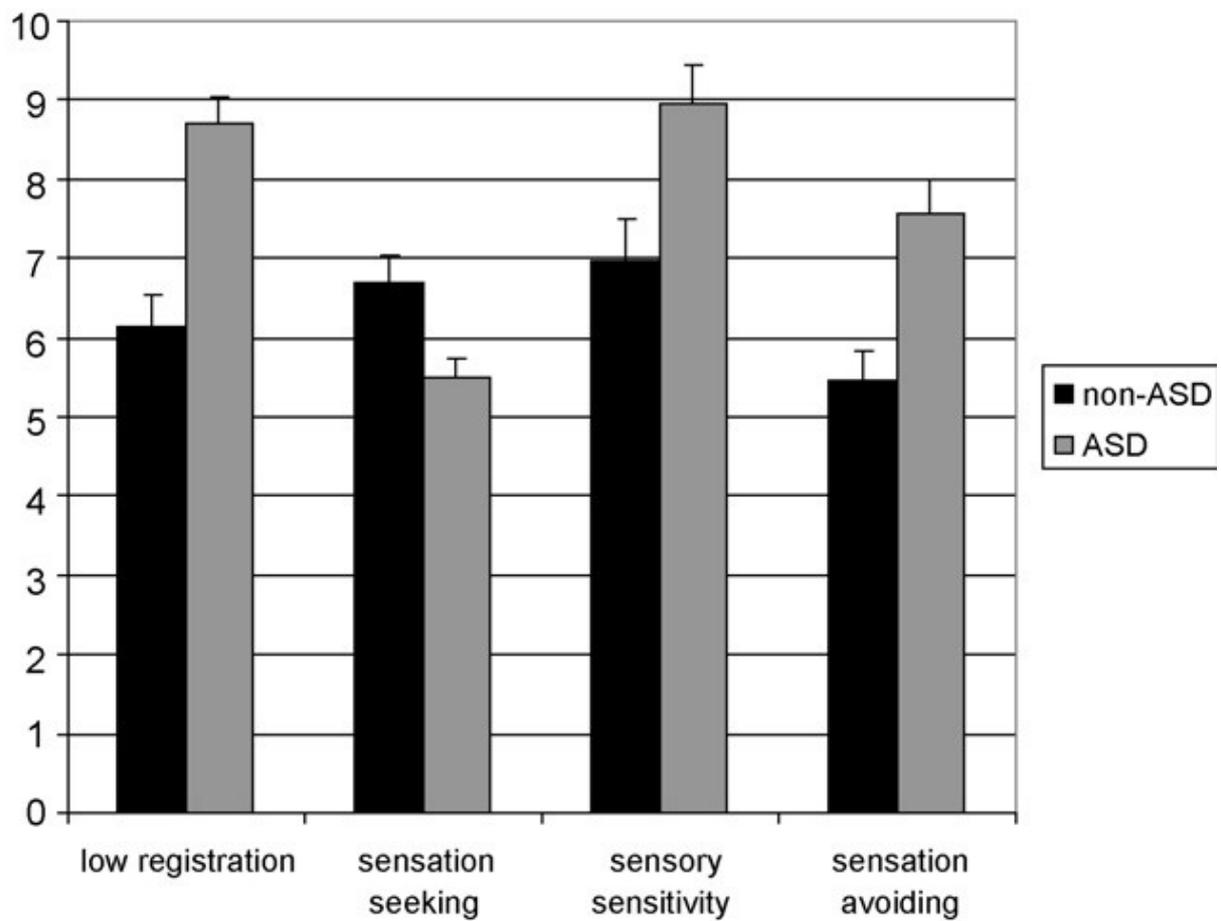


Figure 2: ASD group divided into individuals with a low threshold (indicating better performance) and with a high threshold (indicating poorer performance) on the auditory discriminations task. Groups are plotted for mean scores (and standard error) on auditory items on the Adolescent/Adult Sensory profile. (a) frequency (b) intensity (c) duration.

