Mapping the developmental trajectory and correlates of enhanced pitch perception on speech processing in adults with ASD.

Jennifer L. Mayer,[1] Ian Hannent[2] & Pamela F. Heaton[2]

Abstract: Whilst enhanced perception has been widely reported in individuals with Autism Spectrum Disorders (ASDs), relatively little is known about the developmental trajectory and impact of atypical auditory processing on speech perception in intellectually high-functioning adults with ASD. This paper presents data on perception of complex tones and speech pitch in adult participants with high-functioning ASD and typical development, and compares these with pre-existing data using the same paradigm with groups of children and adolescents with and without ASD. As perceptual processing abnormalities are likely to influence behavioural performance, regression analyses were carried out on the adult data set. The findings revealed markedly different pitch discrimination trajectories and language correlates across diagnostic groups. While pitch discrimination increased with age and correlated with receptive vocabulary in groups without ASD, it was enhanced in childhood and stable across development in ASD. Pitch discrimination scores did not correlate with receptive vocabulary scores in the ASD group and for adults with ASD superior pitch perception was associated with sensory atypicalities and diagnostic measures of symptom severity. We conclude that the development of pitch discrimination, and its associated mechanisms markedly distinguish those with and without ASD.

Keywords: Autism Spectrum Disorders, Pitch Discrimination, Auditory Processing, Developmental Trajectory

[1]University of Roehampton, Department of Psychology, Whitelands College, Holyborne Avenue, London, SW15 4JD, UK.
[2]Goldsmiths College, Department of Psychology, 8 Lewisham Way, New Cross, London SE14 6NW, UK.
CORRESPONDING AUTHOR: Jennifer Mayer
email address: Jennifer.mayer@roehampton.ac.uk, phone: +44 (0)20 8392 3661

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Introduction

Autism Spectrum Disorders (ASDs) are neurodevelopmental disorders characterized by impairments in social communication/interaction and restricted/repetitive interests and/or behaviours. The Diagnostic and Statistical Manual of Mental Disorders (DSM-5) further defines the range of communication abilities in ASD, requiring an additional specifier to indicate whether the disorder has occurred “with or without accompanying language impairment” (American Psychiatric Association, 2013, p. 53). Although the presentation of language impairments is extremely diverse across the population, they appear to be a key feature in predicting the course the disorder will take in an individual (Rutter, 1970; Venter, Schopler & Lord , 1992).

While not specifically mentioned in previous diagnostic criteria, sensory processing atypicalities across modalities are common in individuals with ASD (Leekhman, Nieto, Libby , Wing & Gould, 2007) and the DSM-5 has included hyper or hypo sensitivity to sensory information within the diagnostic criteria under the restricted, repetitive patterns of behaviour cluster (APA, 2013). These atypicalities have an estimated prevalence of between 69% and 90% (Kern et al., 2007) and may affect auditory processing and pain sensitivity thresholds as well as responses to visual, and olfactory stimuli (Gerland, 2003). There is an increasing consensus that sensory abnormalities may predispose an early avoidance of social stimuli, thereby constraining the development of social and cognitive abilities (Ben-Sasson, Cermak, Orsmon, Tager-Flusberg, Carter, Kadlec & Dunn, 2007). Eye-gaze and joint attention behaviours are important precursors for language skills (Norbury, Brock, Cragg, Einav, Griffiths & Nation, 2009) and avoidance of social stimuli may well be associated with delayed language acquisition and atypical language skills in children with ASD (Luyster, Kadlec, Carter & Tager-Flusberg, 2008). Recently, Mayer & Heaton (2014) found that increased levels of sensory sensitivity negatively impacted on speech encoding in verbally able, high-functioning adults with ASD and this suggests that the effects of sensory difficulties are not limited to those with marked language delays and abnormalities and also do not end with childhood.

An important strand of research that has relevance to questions about the behavioural correlates of sensory processing atypicalities has specifically focused on auditory processing in ASD. Two recent review articles (Haesen, Boets, Wagemans , 2011; O’Connor, 2012) provide an in-depth review of the behavioural, neurological and neuroanatomical research on auditory processing in ASD. Overall they present evidence for a diverse range of auditory processing abnormalities in this group. For example, atypical orientation to auditory stimuli, atypical perception of pure tones, loudness, complex stimuli and prosody, as well as difficulties processing auditory information in noise have all been demonstrated. In support of the Enhanced Perceptual Functioning (Mottron & Burack, 2001; Mottron, Dawson, Soulières, Hubert & Burack, 2006) theory of autism, are results from a number of behavioural studies showing enhanced pitch discrimination and memory for simple and complex tones in ASD compared with typically developing control groups (Applebaum, Egel, Koegel & Imhoff, 1979; Bonnel, Mottron, Peretz, Trudel, Gallun, & Bonnel, 2003; Bonnel, McAdams, Smith, Berthiaume, Bertone, Ciocca, Burack, et al., 2010; Heaton, 2003, 2005; Heaton, Hermelin & Pring, 1998, 1999; Heaton, Hudry, Ludlow & Hill , 2008; Heaton, Williams, Cummins & Happé, 2008; Jones et al., 2009; Mottron, Peretz & Menard, 2000; O’Riordan & Passetti, 2006). However, whilst these studies report superior pitch discrimination at the group level in ASD, some evidence suggests that it is limited to subgroups and may be associated with levels of language impairment. For example, Bonnel et al., (2010) observed superior pitch discrimination in adults with autism, but not Asperger syndrome, and studies of adolescents with ASD have reported superior discrimination of complex and pure tones in subgroups with low vocabulary scores and delayed speech onset (Heaton, et al., 2008; Jones et al., 2009).

Although pitch information serves important pragmatic functions in speech (Patel, 2007), evidence from studies of congenital amusia show that relatively normal language development can occur when aspects of pitch discrimination are compromised (Goulet, Moreau, Robitaille & Peretz, 2012; Lebrun, Moreau, Mc-Nally-Gagnon, Goulet & Peretz, 2012). Whilst this suggests that fine-grained pitch discrimination ability is not advantageous for speech perception, and experimental studies have associated enhanced pitch with poor language outcomes in ASD (Bonnel et al., 2010; Heaton et al., 2008; Jones et al., 2009), the results from one study suggest that questions about such an association should be considered in a developmental context.

Heaton, Davis & Happé, (2008) described the case of A.C., an intellectually high-functioning man with ASD with absolute pitch (AP) naming skills that had been established early in childhood and before the emergence of phrase speech at six years. A.C.'s parents reported that his early sensitivity to pitch had resulted in difficulties understanding that the same word, when spoken by a male or a female speaker was not a different word. In the study A.C. and musically trained typical adults with A.P. were asked to name pure and complex tones and to extract and name frequencies in simple French and English words. Whilst A.C. performed at a higher level than controls across all conditions, his superiority emerged most strongly on the linguistic condition, indicating domain-general pitch processing that persisted into adulthood. However, at the time of testing A.C. was fluent in several European and Asian languages and it appeared that whilst enhanced pitch had impaired his early language acquisition it may have facilitated the acquisition of new languages in adulthood (Heaton, Davis et al., 2008). This suggests that in ASD the interaction between auditory processing and language acquisition is atypical, and changes across development.

The first aim of the current paper was to investigate discrimination of speech and non-speech pitch across child, adolescent and adult cohorts and to determine whether this is negatively associated with current receptive vocabulary at the different stages of development in individuals with and without ASD. The study builds on an earlier investigation in which children with ASD demonstrated a superior ability to make same/different judgments about pitch changes across word, non-word, and analogue pitch contour stimulus pairs (Heaton, Hudry et al., 2008). The experiment was completed by groups of adolescents and adults with autism and the analysis was carried out on these data and the data from the earlier child study.

STUDY 1: EXPLORING THE DEVELOPMENTAL TRAJECTORY OF PITCH DISCRIMINATION IN INDIVIDUALS WITH AND WITHOUT ASD

Methods

Participants

Data were collected from high functioning adults with ASD and typically developing controls and compared with pre-existing data using the same paradigm with groups of children and adolescents, with and without ASD. All of the participants were asked to confirm that they did not have any diagnosed hearing difficulties. The children and adolescents received age-appropriate prizes for their participation and the adult participants were paid £8/hour for taking part.

Child Cohorts

The pre-existing child ASD sample consisted of 14 children, all male (Heaton, Hudry, et al., 2008). The participants' ASD diagnoses were confirmed through inspection of their Special Educational Needs (SEN) Statements. Their chronological ages ranged between 6 years 11 months and 14 years 9 months. 14 control children, 13 male and 1 female, were group matched based on age and receptive vocabulary scores to the ASD group. The age and intelligence matched control children where either typically developing or had moderate learning difficulties. Their chronological ages ranged between 5 years 0 months and 14 years 1 month. All of the children completed a receptive vocabulary test (British Picture Vocabulary Scales (BPVS); Dunn, Whetton & Burley, 1997) and the raw scores obtained were converted into standard scores (table 1). The two groups did not differ significantly on either of the measures on which they were matched (Chronological age, CA $t(27)=-0.306$, *ns;* British Picture VS, $t(27)=0.870$, *ns*).

Adolescent Cohorts

14 adolescents diagnosed with ASD, 13 males and 1 female, were previously recruited and participated in the experiment (Mayer, 2009). All of the adolescents in the ASD group were either recruited from a secondary school for children with ASD in England or from local support groups. The participants' ASD diagnoses were confirmed through inspection of their SEN Statements. Their chronological ages ranged between 9 years 8 months and 16 years 5 months. 14 control adolescents, all male, were group matched based on age and non-verbal IQ scores to the ASD group and participated in the experiment. All had previously been recruited from a mainstream secondary school in England. Their chronological ages ranged between 12 years 0 months and 16 years 9 months.

All of the adolescents were administered the BPVS (table 1). Participants were also administered the Ravens Progressive Matrices to assess their non-verbal IQ. Raw scores for the ASD group ranged between 20 and 50 (5^{th}-75^{th} percentile) with a mean of 34.67, and between 27 and 41 (5^{th}-50^{th} percentile) with a mean of 35.20 in the control group. The two groups did not significantly differ on either of the measures on which they were matched (CA, $t(27)=0.506$, *ns;* Ravens, $t(27)=-0.199$, *ns*). However, a significant difference was found between the two groups on verbal mental age, derived from the BPVS, $t(27)=-5.891$, $p<0.001$ with the control group achieving higher scores than the ASD group.

Adult Cohorts

19 adults with ASD, 4 females and 15 males, were recruited for the current study. Their chronological ages ranged between 23 years 9 months and 59 years 8 months. All of the adults in the ASD group were recruited from local support groups or from previous research at Goldsmiths College and City University, London, England. All ASD participants' had been previously diagnosed by clinicians using DSM-IV criteria and these diagnoses were confirmed by the first author using ADOS module 4. Of the 19 ASD participants recruited, two did not meet overall diagnostic criteria on the ADOS. However, as all participants had previously been diagnosed by a clinician and the results from the background assessments and the experimental task did not change if those individuals were excluded from the analysis, they were retained in the final sample. 19 adults with typical development, 4 females and 14 males, were group matched based on age and receptive vocabulary to the ASD group. The Peabody Picture Vocabulary Test (PPVT) (Dunn & Dunn, 1997), includes norms for adults and has a similar format to the BPVS, therefore this test was used with the adult group. Their chronological ages ranged between 25 years 1 month and 52 years 8 months. Control participants were recruited from the University of London and the local community.

Table 1. *Child, Adolescent, and Adult Cohort's Age and Receptive Vocabulary Scores*

	CA (months)		Receptive Vocabulary [a]	
	Mean (SD)	Range	Mean (SD)	Range
ASD Child	126.07 (47.53)	83-177	82.36 (18.00)	50-105
Control Child	126.28 (28.47)	60-169	77.71 (13.94)	53-106
ASD Adolescent	165.64 (23.46)	116-197	71.50 (22.94)	46-126
Control Adolescent	162.93 (10.54)	144-201	100.07 (16.14)	72-129
ASD Adult	482.79 (136.00)	285-716	105.63 (12.07)	76-123
Control Adult	459.79 (108.64)	301-632	106.05 (10.24)	84-125

Note: CA=chronological age, ASD= autism spectrum disorders
[a]British Picture Vocabulary Scales (BPVS), standard score (Dunn et al., 1997) (child and adolescent data) or Peabody Picture Vocabulary Test (PPVT), standard score (Dunn & Dunn, 1997) (adult data)

Experimental Methods

Experimental Stimuli

The two stimulus types in the present study assessed discrimination of pitch changes in speech and non-speech stimuli. The paradigm and stimuli were developed and utilized in a behavioural study carried out with children and adolescents with and without ASD (Heaton, Hudry, et al., 2008), and revealed significantly increased sensitivity to changes in speech pitch contours in ASD participants.

Pitch discrimination was assessed at different levels of complexity. Stimuli were either pairs of monosyllabic words (e.g. boot, got, hit), or pitch contours derived from these words. In each trial, words or analogue pairs were presented at either the same pitch or at a distance corresponding to 2, 3 or 6 semitones in the musical scale. In each of the two conditions (word and analogue tones) there were 40 trials, 10 at each pitch interval, resulting in a total of 80 stimuli that were presented to the participants in a computer generated random order. For more detail regarding development of stimuli see Heaton, Hudry, et al. (2008).

Procedure

For each condition participants were administered 10 practice trials in which a recorded instruction stated "Listen carefully, are these two the same?" followed by the stimulus pair. The participant was instructed to indicate whether the two words in the pair were the same or a different pitch by pressing a button on a computer keyboard labelled "S" or "D". During the practice trails, participants received feedback after each stimulus pair indicating whether or not they had answered correctly. Following the 10 practice trials, 40 experimental trials were administered in the same format, but without the recorded instruction or feedback. The order of presentation of the two conditions was counterbalanced across sessions and the experimenter sat with the participant offering encouragement regardless of their performance on the task. Raw scores for each of the tasks were obtained by counting the number in which the participant's had responded correctly with a maximum of 40.

Results

A mixed factorial analysis of variance (ANOVA) was conducted with within-subjects factors of stimulus type (2 levels; words and analogue contours of words) and pitch interval (4 levels; same, small, medium, and large pitch differences) and a between-subjects factor of group (6 levels; ASD adult, ASD adolescent, ASD child, Control adult, Control adolescent, and Control child). The dependent variable was the percentage of correct responses made by each participant across the 10 trials at each pitch interval in each of the two stimulus types (table 2).

Table 2. *Mean percentage correct scores, standard deviations and ranges on experimental task*

ASD Child	Words			Analogue Contours		
N=14	Mean (SD)	Range	% Ceiling	Mean (SD)	Range	% Ceiling
Same	89.28 (13.28)	60.00-100.00	50%	89.28 (12.69)	60.00-100.00	50%
Small	61.43 (29.05)	00.00-100.00	14%	77.14 (28.94)	20.00-100.00	50%
Medium	77.14 (22.68)	30.00-100.00	28%	85.71 (19.50)	40.00-100.00	50%
Large	85.71 (28.48)	00.00-100.00	64%	88.57 (17.91)	50.00-100.00	57%
Total	78.39 (18.54)	35.00-100.00	7%	85.18 (16.74)	50.00-100.00	28%
Control Child	Words			Analogue Contours		
N=14	Mean (SD)	Range	% Ceiling	Mean (SD)	Range	% Ceiling
Same	79.28 (23.36)	30.00-100.00	36%	85.00 (17.87)	50.00-100.00	36%
Small	35.00 (28.22)	00.00-90.00	0%	43.57 (29.77)	10.00-100.00	14%
Medium	43.57 (32.49)	00.00-100.00	14%	46.43 (28.98)	00.00-100.00	14%
Large	52.14 (31.91)	00.00-100.00	14%	74.28 (18.28)	40.00-100.00	14%
Total	52.50 (22.81)	25.00-97.50	0%	62.32 (18.22)	37.50-100.00	14%
ASD Adolescent	Words			Analogue Contours		
N=14	Mean (SD)	Range	% Ceiling	Mean (SD)	Range	% Ceiling
Same	90.00 (16.17)	50.00-100.00	57%	85.71 (17.85)	40.00-100.00	36%
Small	56.43 (30.03)	10.00-100.00	7%	70.00 (29.61)	20.00-100.00	36%
Medium	65.71 (29.80)	10.00-100.00	14%	77.86 (26.94)	20.00-100.00	43%
Large	81.43 (25.97)	10.00-100.00	43%	86.43 (18.65)	50.00-100.00	50%
Total	73.39 (21.49)	27.50-100.00	0%	80.00 (19.83)	50.00-100.00	21%
Control Adolescent	Words			Analogue Contours		
N=14	Mean (SD)	Range	% Ceiling	Mean (SD)	Range	% Ceiling
Same	95.71 (6.46)	80.00-100.00	64%	90.71 (17.30)	40.00-100.00	64%
Small	36.43 (22.05)	00.00-80.00	0%	52.86 (37.09)	00.00-100.00	21%
Medium	50.00 (21.48)	20.00-90.00	0%	65.00 (26.53)	20.00-100.00	21%
Large	77.14 (15.41)	60.00-100.00	21%	81.43 (16.57)	50.00-100.00	21%
Total	64.82 (13.03)	52.50-97.50	0%	72.50 (20.02)	40.00-100.00	14%
ASD Adult	Words			Analogue Contours		
N=19	Mean (SD)	Range	% Ceiling	Mean (SD)	Range	% Ceiling
Same	92.10 (16.18)	40.00-100.00	68%	95.79 (9.01)	70.00-100.00	79%
Small	40.00 (27.28)	10.00-100.00	5%	64.74 (34.05)	20.00-100.00	42%
Medium	63.16 (28.49)	10.00-100.00	21%	73.68 (33.37)	10.00-100.00	42%
Large	87.37 (20.23)	40.00-100.00	63%	91.05 (15.60)	50.00-100.00	68%
Total	70.66 (17.48)	40.00-100.00	5%	81.31 (20.84)	37.50-100.00	31%
Control Adult	Words			Analogue Contours		
N=19	Mean (SD)	Range	% Ceiling	Mean (SD)	Range	% Ceiling
Same	95.79 (9.61)	60.00-100.00	74%	98.42 (5.01)	80.00-100.00	89%
Small	53.16 (27.29)	10.00-100.00	5%	73.15 (26.68)	30.00-100.00	32%
Medium	73.68 (25.21)	30.00-100.00	31%	84.74 (21.18)	30.00-100.00	47%
Large	93.68 (11.64)	60.00-100.00	68%	95.79 (6.07)	80.00-100.00	63%
Total	79.08 (14.49)	50.00-100.00	0%	88.03 (13.53)	55.00-100.00	31%

Note: Mean percentage correct scores (out of a maximum of 100)
Child data from Heaton, Hudry, et. al (2008); Adolescent data from J. Mayer's MSc Dissertation (2009)

The ANOVA results were consistent with Heaton, Hudry, et al.'s (2008) findings in showing that participants in all groups experienced more difficulty discriminating stimuli with semantic content and that correct discrimination of 'different' pitches significantly improved as the size of the pitch interval difference increased (all comparisons $p<0.001$) (N.B. with a Bonferroni corrected p threshold). Further post hoc pairwise comparisons revealed that participants made significantly more correct decisions on the "small", t(93)=-6.21, p<0.001, "medium", t(93)=-4.40, p<0.001, and "large" t(93)=-2.91, p<0.01 pitch intervals during the analogue contour stimuli than during the word stimuli (Fig 1). However, there was no significant difference in the participants' performance between the two tasks when the pitches were the same, t(93)=-0.45, p=0.65. It was noted that the verbal IQ scores were lower in the adolescent cohort than in the child and adult cohorts and we conducted a second ANOVA, excluding this group. This analysis failed to change the pattern of results across groups and conditions (table 3).

Table 3. *Main Effects and Interactions for the full ANOVA and ANOVA excluding the adolescent cohorts*

	Analyses with All Groups	Analyses without Adolescents
Stimulus Type ME	***F*(1, 93)=38.16, *p*<0.001**	***F*(1, 65)=33.56, *p*<0.001**
Pitch Interval ME	***F*(1.94, 93)=125.63, *p*<0.001**	***F*(2.02, 65)=81.75, *p*<0.001**
Group ME	***F*(5, 93)=4.89, *p*<0.001**	***F*(3, 65)=7.59, *p*<0.001**
Stimulus x Pitch Interaction	***F*(2.55, 93)=10.15, *p*<0.001**	***F*(2.46, 65)=5.74, *p*<0.001**
Stimulus x Group Interaction	*F*(5, 93)=0.25, *p*=0.937	*F*(3, 65)=0.27, *p*=0.854
Pitch x Group Interaction	***F*(15, 93)=2.81, *p*<0.001**	***F*(9, 65)=3.04, *p*<0.01**
Stimulus x Pitch x Group Interaction	*F*(15, 93)=1.38, *p*=0.154	*F*(9, 65)=1.88, *p*=0.073

Note: Significant main effects and interactions are in bold

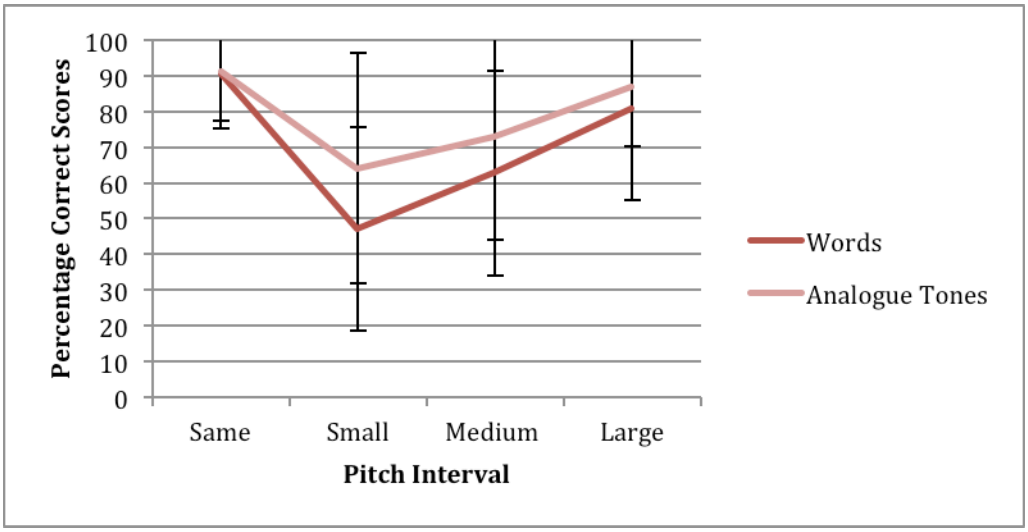

Figure 1. *Study 1 ANOVA Pitch Interval x Stimulus Type Interaction*

The primary aim of the present study was to examine the developmental trajectory of pitch discrimination abilities therefore the main effect of group was examined in greater detail. The analysis revealed a significant main effect of group F(5, 93)=4.89, p<0.001, (Fig 2) with the children without autism, obtaining lower scores than all of the other groups. As this figure suggests unique developmental trajectories for the ASD and non-ASD groups, a trend analysis using linear contrasts to examine the change in performance across cohorts was carried out for each of the 2 diagnostic groups. Within the non-ASD groups there was a significant linear trend, indicting that as age group progressed, pitch discrimination abilities increased proportionately. This trend was present in word, F(1, 44)=20.31, p<0.001, and analogue tone, F(1, 44)=18.27, p<0.001, stimulus pairs as well as in their total pitch discrimination scores, F(1, 44)=23.19, p<0.001. However, within the ASD groups, there were no significant linear trends, indicating that as age group progressed pitch discrimination abilities remained relatively stable, regardless of whether the stimulus pair contained words, F(1, 44)=1.33, p=0.255, or analogue tones, F(1, 44)=0.32, p=0.575, and was also true of their total pitch discrimination scores, F(1, 44)=0.81, p=0.372.

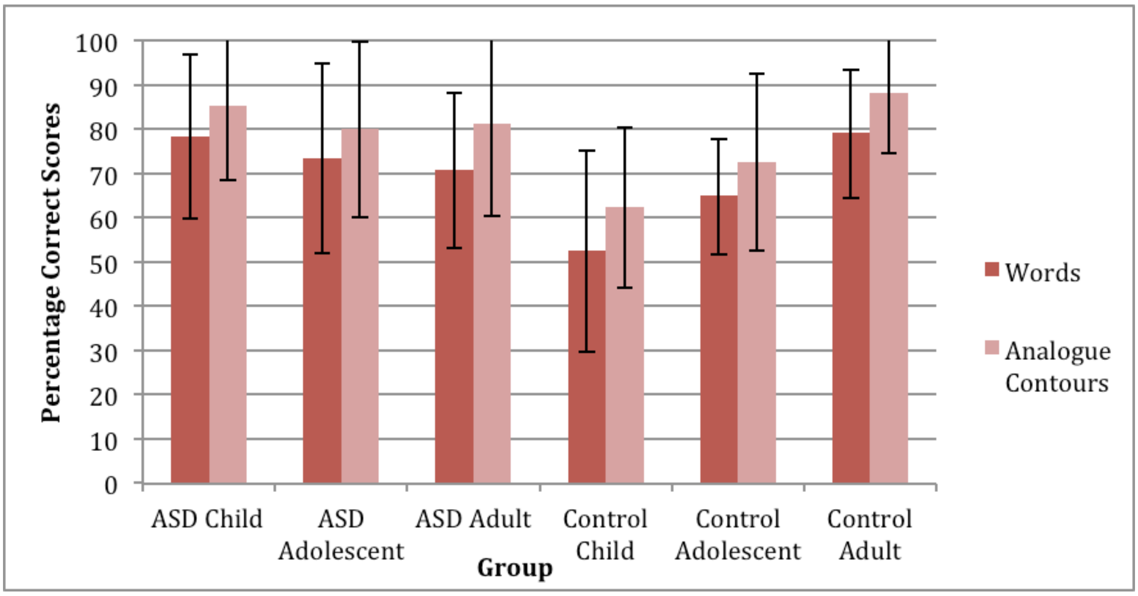

Figure 2. *Study 1 ANOVA Main Effect of Group*

The results show that whilst typical and autistic adults show similar levels of perceptual discrimination on the experimental task, the developmental trajectories leading to these performance levels distinguish the two groups.

Exploring the Different Developmental Trajectories

In order to further explore the different developmental trajectories, a correlation analysis was performed. Participants' total percentage correct scores on word pairs, analogue tone pairs, and total pitch discrimination scores along with participants' receptive vocabulary scores and age in months were used in the correlation. Within the ASD groups there were no significant correlations between participants' pitch discrimination scores and their receptive verbal abilities and age. However, within the control groups both age and verbal IQ were significantly positively correlated with performance on word and analogue tone pairs as well as on total discrimination scores (table 3). Importantly these results remained the same when the adolescent cohorts, with different baseline verbal abilities, were removed from the analyses.

Table 3. *Summary of correlations between experimental task performance and age and verbal IQ*

	ASD		Control	
	Verbal Ability	Age	Verbal Ability	Age
Word Total	0.040	-0.115	0.476*** (**)	0.384**
Analogue Tone Total	0.077	-0.025	0.531***	0.426**
Total Discrimination	0.062	-0.073	0.536***	0.431**

*Note: *p<0.05, **p<0.01, ***p<0.001 (two-tailed); (**) represents a change in significance level when the adolescent cohorts were removed from the analyses.*

Summary

In summary, the results from study 1 revealed a significant increase in pitch discrimination abilities from childhood and adolescence into adulthood within typically developing individuals whilst the performance within the ASD groups did not change over time. This suggests that the failure to observe superior pitch discrimination in the adult ASD cohort resulted from a significant increase in pitch discrimination ability in typical adults. Most striking was the apparent stability of pitch discrimination ability across age and verbal skill in the ASD group. Superior pitch discrimination has previously been observed in adults with ASD (Bonnel et al., 2010; Heaton et al., 2008) and is consistent the EPF model of ASD (Mottron et al., 2006). Many of the adults in the ASD group were intellectually high functioning and possessed good language skills and it is plausible to suggest that their apparently normative performance on the pitch discrimination task reflects a shift from perceptual to higher order auditory information processing. However, this suggestion is discounted by our failure to observe a significant negative relationship between pitch discrimination and receptive verbal abilities in the ASD group. Taken together the results from Study 1 suggest that sensitivity to pitch information increases with age and is positively associated with language skills in typical development. In contrast, they also suggest that pitch discrimination is enhanced in childhood in ASD, remains stable over development, and may not be associated with language skills at any developmental stages.

STUDY 2: EXPLORING THE UNDERLYING MECHANISMS INVOLVED IN SPEECH PROCESSING IN ADULTS WITH ASD AND TYPICAL DEVELOPMENT

According to Kellerman et al.'s (2005) review, previous studies examining auditory processing in ASD have identified both enhanced and impaired discrimination performance. These contradictory findings may reflect differences in the types of stimuli used in the studies (e.g. sine/complex tones) or may result from heterogeneity characteristic in ASD. The results from Bonnel et al. (2010) associated diagnostic factors (high-functioning autism/Asperger syndrome) with enhanced pitch and Jones et al. (2009) observed increased sensory abnormalities in her participants with enhanced pitch discrimination. Therefore the primary aim of study 2 is to identify the cognitive, clinical, and behavioural correlates of enhanced pitch sensitivity in the adult sample.

Methods

The ASD and typically developing adults were closely matched on all cognitive measures, and the ASD group obtained significantly higher scores on all behavioural and clinical measures of autism symptoms (table 4).

Cognitive Correlates

The Weschler Abbreviated Scales of Intelligence (WASI) (Wechsler, 1999) was used as a measure of intellectual and cognitive functioning and the Peabody Picture Vocabulary Test (PPVT) (Dunn & Dunn, 1997) was used as a measure receptive vocabulary. In order to assess participants' working memory capacity, the backwards digit span subtest from the Weschler Adult Intelligence Scale – Fourth Edition (WAIS-IV) (Wechsler, 2008) was used.

Behavioural Correlates

The Communication Checklist – Self Report (CC-SR) (Bishop, Whitehouse & Sharp, 2009) was administered to provide information on any difficulties in speech, language, or interaction. The CC-SR is a 70-item self-report questionnaire, which examines three factors of communication: Language Structure, Pragmatic Skills, and Social Engagement. Higher scores on the CC-SR indicate an increased level of communication difficulties.

Measures of sensory abnormalities using the Adult/Adolescent Sensory Profile (SP) test (Brown & Dunn, 2002) were also obtained. The SP is a 60 item self-report questionnaire that examines sensory processing patterns across six sensory processing categories including: taste/smell, movement, visual, touch, activity, and auditory processing. Participants' raw scores across the 6 categories are used to derive their quadrant scores identified as: Low Registration (“I don’t get jokes as quickly as others”), Sensation Seeking (“I like to wear colourful clothing”), Sensory Sensitivity (“I am distracted if there is a lot of noise around”), and Sensation Avoiding (“I stay away from crowds”). Higher scores within each quadrant represent increased sensory abnormalities.

Clinical Correlates

In order to assess the self-reported levels of autistic traits in participants the Adult Autism Spectrum Quotient (AQ) (Baron-Cohen, Wheelwright, Skinner, Martin & Clubley 2001) was administered. The AQ is a 50-item questionnaire that examines 5 factors: Social Skills, Attention Switching, Attention to Detail, Communication, and Imagination. Within the AQ autistic-like behaviour is characterised by poor social, communication, or imagination skills, exceptional attention to detail, and either poor attention switching or a strong focus of attention (Baron-Cohen et al., 2001).

ASD participants’ pre-existing diagnoses were confirmed by administering the Autism Diagnostic Observation Schedule (ADOS) module 4 (Lord, Rutter, DiLavore & Risi , 2001). The ADOS provides a score representing autistic symptom severity in the areas of: Communication, Reciprocal Social Interaction, Imagination and Creativity, and Repetitive Behaviours.

Table 4. *Study 2 Participant Background Data*

	ASD N= 19		TD N= 19		
	Mean (SD)	Range	Mean (SD)	Range	*p* values
CA	40y8m (11.33)	23y9m-59y8m	38y3m (9.00)	25y1m-52y9m	0.568
Cognitive Correlates					
WASI Full Scale[a]	113.37 (15.27)	78-133	118.95 (10.84)	87-134	0.203
WASI Verbal[a1]	111.16 (15.57)	71-132	115.58 (11.52)	83-135	0.326
WASI Performance[a2]	112.95 (12.97)	92-129	118.05 (12.21)	96-136	0.221
PPVT[b]	105.63 (12.07)	76-123	106.05 (10.24)	84-125	0.908
WM-Total[c]	19.68 (4.57)	13-30	19.16 (4.69)	13-28	0.728
WM-Forward[c1]	11.32 (2.43)	7-16	11.53 (2.32)	8-15	0.786
WM-Backward[c2]	8.37 (2.54)	4-14	7.63 (2.98)	4-13	0.418
Behavioural Correlates					
CC-SR-Total[d]	67.84 (33.28)	32-159	22.00 (13.81)	1-50	<0.001*
CC-Lang. Struct.[d1]	14.58 (12.19)	1-49	5.00 (3.97)	0-16	<0.01*
CC-Pragmatics[d2]	17.84 (11.35)	0-39	5.89 (6.71)	0-25	<0.001*
CC-Social Eng.[d3]	35.42 (12.58)	19-71	11.11 (5.65)	1-24	<0.001*
Sensory Profile-Total[e]	179.58 (26.09)	130-218	131.89 (28.36)	32-160	<0.001*
SP-Low Reg.[e1]	43.42 (10.41)	27-62	26.16 (6.23)	10-35	<0.001*
SP-Sensation Seek.[e2]	43.79 (8.29)	31-63	47.58 (9.88)	12-58	0.209
SP-Sensory Sens.[e3]	47.16 (10.19)	23-62	29.05 (8.18)	4-39	<0.001*
SP-Sensat. Avoid.[e4]	45.21 (9.54)	31-61	29.11 (9.31)	6-48	<0.001*
Clinical Correlates					
AQ-Total[f]	35.16 (7.59)	21-45	12.26 (5.45)	3-21	<0.001*
AQ-Social Skills[f1]	6.72 (2.58)	3-10	1.32 (1.38)	0-4	<0.001*
AQ-Atten. Switch[f2]	8.67 (1.37)	6-10	3.26 (1.79)	0-6	<0.001*
AQ-Atten. to Detail[f3]	7.22 (2.13)	1-10	3.58 (2.10)	0-7	<0.001*
AQ-Commun.[f4]	6.50 (2.55)	2-10	1.95 (1.39)	0-5	<0.001*
AQ-Imagination[f5]	6.22 (2.29)	2-10	2.16 (1.98)	0-7	<0.001*
ADOS-Diagnostic[g]	9.58 (3.55)	5-17	N/A	N/A	N/A
ADOS-Commun.[g1]	2.84 (1.54)	1-6	N/A	N/A	N/A
ADOS-Soc. Int.[g2]	6.74 (2.70)	3-12	N/A	N/A	N/A
ADOS-Imag.[g3]	1.05 (0.70)	0-2	N/A	N/A	N/A
ADOS-Rep. Behav.[g4]	1.58 (1.02)	0-3	N/A	N/A	N/A

Note: CA= chronological age, ASD= Autism Spectrum Disorders, TD= typically developing
[a]Weschler Abbreviated Scales of Intelligence (WASI), standard score (Wechsler, 1999); [a1]WASI Verbal IQ; [a2]WASI Performance IQ. [b]Peabody Picture Vocabulary Test (PPVT), standard score (Dunn & Dunn, 1997). [c]Working Memory Digit Span (WM), Weschler Adult Intelligence Scales, (Wechsler, 2008); [c1]WM Forward Digit Span; [c2]WM Backward Digit Span. [d]Communication Checklist – Self Report (CC-SR), raw score (Bishop et al., 2009); [d1]CC-SR Language Structure; [d2]CC-SR Pragmatics; [d3]CC-SR Social Engagement.[e]Adult/Adolescent Sensory Profile (SP), (Brown & Dunn, 2002); [e1]SP Low Registration; [e2]SP Sensation Seeking; [e3]SP Sensory Sensitivity; [e4]SP Sensation Avoiding. [f]Adult Autism Spectrum Quotient (AQ), (Baron-Cohen et al., 2001); [f1]AQ Social Skills; [f2]AQ Attention Switching; [f3]AQ Attention to Detail; [f4]AQ Communication; [f5]AQ Imagination. [g]Autism Diagnostic Observation Schedule (ADOS), diagnostic total (Lord et al., 2001); [g1]ADOS Communication; [g2]ADOS Reciprocal Social Interaction; [g3]ADOS Imagination & Creativity; [g4]ADOS Repetitive Behaviours

Results

Correlation Analyses

Correlation analyses were carried out on the fine-grained pitch discrimination data and background data for the ASD and TD adults. Performance on the small and medium pitch difference conditions were highly correlated with each other within both the word, r=0.644, p<0.001, and analogue tone, r=0.847, p<0.001, tasks. The scores on the small and medium pitch differences were combined for each task to make two dependent variables, along with the cognitive, clinical, and behavioural variables (described above) for the correlation analyses. Background measures that were significantly correlated with word or tone pitch discrimination scores in either group are shown in table 5. These factors were then used in regression analyses carried out on the data from the word and analogue tone tasks.

Table 5. *Summary of significant correlations between enhanced pitch and background measures*

ASD; ***TD***	Word Pitch Discrimination	Tone Pitch Discrimination
Cognitive Correlates		
PPVT	NS	0.472*
Digit Span		
Total	NS	0.624**
Forward	NS	0.527*
Backward	NS	0.618**
Behavioural Correlates		
Sensory Profile		
Low Registration	0.468*	NS
Sensation Avoiding	0.555**	NS
Clinical Correlates		
AQ		
Attention to Detail	0.460*	NS
ADOS		
Reciprocal Social Interaction	NS	-0.687**
Diagnostic Score	NS	-0.624**

*Note: *p<0.05, **p<0.01, ***p<0.001 (two-tailed)*

Regression Analyses

Word Stimuli

In order to examine the extent to which the measures identified as pitch identification correlates accounted for the variance in performance on the word task in ASD and typically developing participants, two multiple linear regressions were performed on the two groups separately. The dependent variable was the pitch discrimination scores for the combined small and medium pitch differences in the word task. The predictor variables were individuals' scores on the low registration and sensation avoiding subscales of the Sensory Profile and their scores on the attention to detail subscale of the AQ. Due to the exploratory nature of this analysis, a backwards stepwise entry method was employed. In this method all of the potential variables are entered in the model initially and then individual variables are removed if their removal significantly improves the model.

The results revealed that there was no significant linear relationship between ASD participants' pitch discrimination scores on the word task and the predictor variables with a multiple correlation of 0.29, [$F(1,19)=1.05$, $p=0.322$; adjusted $R^2=0.00$]. Thus, there didn't appear to be a relationship between the predictor variables and pitch discrimination abilities on the word task in the ASD population.

There was however a significant linear relationship between typically developing participants' pitch discrimination scores during the word task and the predictor variables, with a multiple correlation of 0.55, [$F(1,19)=7.58$, $p<0.01$; adjusted $R^2=0.268$]. Thus, roughly 27% of the variability in typically developing participants' pitch discrimination scores during the word task were predicted by their scores on the sensation avoiding subscale of the Sensory Profile. An increase of one unit in the sensory atypicality score in the realm of sensation avoiding predicted an increase of 1.4% in a TD individual's pitch discrimination scores.

Analogue Tone Stimuli

In the multiple regression carried out on the analogue tone stimuli, the dependent variable was the pitch discrimination scores for the combined small and medium pitch differences. The predictor variables were individuals' scores on the PPVT, forward digit span, backward digit span, total digit span, and the reciprocal social interaction and diagnostic score subscales of the ADOS.

The results revealed a significant model for the predictor variables within the ASD group with a multiple correlation of 0.77, [$F(2,19)=11.84$, $p<0.001$; adjusted $R^2=0.546$]. Roughly 55% of the variability in ASD participants' pitch discrimination scores during the analogue tone task were predicted by their backwards digit span score and level of symptom severity on the reciprocal social interaction subscale of the ADOS. An increase in scores on the digit span test predicted an increase of 5% in an ASD individual's pitch discrimination scores, whereas an increase of one unit in reciprocal social interaction scores on the ADOS predicted a decrease of 6% in pitch discrimination scores.

The results also revealed a significant linear relationship between typically developing participants' pitch discrimination scores during the analogue tone task and the predictor variables, with a multiple correlation of 0.72, [$F(1,19)=4.488$, $p<0.05$; adjusted $R^2=0.177$]. Thus, roughly 17% of the variability in typically developing participants' pitch discrimination scores during the analogue tone task were predicted by their receptive vocabulary score on the PPVT. An increase of one unit in receptive vocabulary scores predicted an increase of 1% in a TD individual's pitch discrimination scores.

Discussion

Study 1 aimed to explore the developmental trajectory of pitch discrimination in children, adolescents and adults with and without ASD. Results from previous studies testing pitch discrimination using complex and simple tones have reported enhanced pitch discrimination in ASD (e.g. Heaton et al., 1998; Mottron et al., 2000) and more recent evidence suggests that atypical processing of pitch information generalises to speech (Jarvinen-Pasley & Heaton 2007; Jarvinen-Pasley, Pasley & Heaton, 2008; Heaton et al., 2008). Using a paradigm that had previously revealed enhanced discrimination of pitch contour changes in words and non-words in children with ASD (Heaton, Hudry, et al., 2008), study one compared data from child, adolescent and adult cohorts with ASD and matched controls. Whilst the three ASD groups were not matched on intelligence or symptom severity, they showed surprisingly similar levels and patterns of performance on the pitch discrimination task. This suggests that enhanced pitch is in evidence early in development in ASD and is stable over time. In contrast to the pattern observed across the ASD groups, the comparison groups without ASD, showed significant gains in discrimination performance across development. It was also noted that whilst pitch discrimination scores correlated with language scores in the control groups, such an association was not observed in the ASD groups.

Study 2 explored this finding further and attempted to identity specific cognitive, behavioural and clinical factors associated with enhanced pitch in the ASD group. The analysis of the typical adult control data from study two confirmed the positive association between receptive vocabulary and pitch discrimination abilities observed in study one. For participants with ASD pitch discrimination scores were not associated with receptive vocabulary scores but were negatively associated with scores on the social interaction scale and positively associated with non-verbal intelligence and digit span scores. The results from study two also suggested links between enhanced pitch discrimination and the negative symptoms characterising ASD. However, this latter finding is currently difficult to interpret.

Strong negative correlations were found between ASD individuals' performance on the analogue tone task and their scores on the ADOS. Whilst these results appear to suggest that increasing levels of symptom severity negatively impact on the individual's pitch processing abilities, the absence of any significant correlation between discrimination scores and scores on the Communication Checklist failed to support this suggestion. It appears from our results, that ADOS scores may not be generally suitable for measuring symptom severity in the context of an empirical analysis. However, within the typically developing group positive correlations between pitch discrimination scores and scores on the AQ suggested that individuals with higher levels of ASD traits are better able to identify more subtle perceptual changes to linguistic stimuli than those with lower levels of ASD traits. This result provides support for the argument that the tail of the ASD spectrum extends to typically developing individuals.

The results from the two studies were consistent with research highlighting the positive association between good perceptual processing and language skills in typical development. For example, research has shown that musical training benefits language in typical development (Anvari, Trainor, Woodside & Levy, 2002) and our analyses revealed a strong link between good pitch discrimination and receptive vocabulary scores in the participants without ASD. Whilst a number of experimental studies have linked language deficits to atypical auditory processing in ASD (Kjelgaard & Tager-Flusberg, 2001; Järvinen-Pasley & Heaton, 2007; Järvinen-Pasley, Pasley, et al., 2008; Järvinen-Pasley, Wallace, et al., 2008) our results failed to reveal a negative correlation between receptive language and pitch discrimination scores in our ASD participants, and this suggests that enhanced pitch perception is not associated with constrained vocabulary acquisition.

Previous findings showing enhanced pitch perception in ASD have been interpreted in the context of the EPF theory (Bonnel et al., 2003, 2010) and we suggest that this model provides the best explanation for our findings. However, our results revealed superior pitch discrimination at early, but not later, developmental stages. We propose that developmental factors play a role in determining the consequences of enhanced pitch in ASD and therefore suggest the following theoretical framework.

Language acquisition is concerned with understanding and conveying meaning and during auditory processing typically developing infants' resources are focused on extracting semantic content from auditory information. Increases in cognitive and memory resources occur over time and attention can be simultaneously allocated to both perceptual and higher-order information when necessary. This enables the individual to process explicit meaning in words and sentences whilst being aware of those prosodic cues that provide additional information about a speaker's intentions. In ASD the initial setting is more perceptual than higher order, but as is the case in typical development, increasing memory and cognitive resources allow for the simultaneous processing of both. It has been suggested that overly selective attention towards the low-level perceptual components of speech may hinder the development of higher-level language processing and even in some cases language acquisition and development in ASD (Schreibman, Kohlenberg & Britten, 1986). Whilst this model is highly speculative, it is consistent with research showing that individuals with ASD and good pitch processing capacities are relatively insensitive to pitch-mediated semantic cues in prosody (Järvinen-Pasley, Pasley, et al., 2008), suggesting that pitch in speech is less strongly yoked to meaning than in typical development.

The model does not propose an overall difference in attentional resources across ASD and non-ASD groups. Instead it assumes an initial difference in the allocation of attentional resources across groups, and it also assumes that changes in allocation of attention will show different trajectories across groups. The model assumes that individuals without ASD allocate a substantial proportion of their attentional resources to meaning embedded in auditory information, and only minimal resources are allocated to psychoacoustic cues, such as pitch. However, during development attentional resources increase and can be allocated elsewhere. This then explains the significant increase in pitch discrimination observed in the typically developing cohorts. In contrast, individuals with ASD initially allocate a substantial proportion of their attentional processes to pitch, leaving fewer resources available for higher-order language processing. In our study levels of pitch discrimination were constant across ASD cohorts, and A.C.'s pitch discrimination skills remained in evidence, despite gains in language skills (Heaton, Davis, et al., 2008). For the adult participants with ASD, pitch discrimination scores were not correlated with language scores but were highly correlated with measures of memory, and it may be the case that auditory memory, and non-verbal intelligence have a direct effect on the efficiency with which they allocate their attentional resources.

Limitations

A potential limitation of study one is that it utilised cross-sectional data that was collected as part of three separate studies, and whilst matching criteria was consistent within each cohort, it was not consistent across cohorts. Most notably, the child and adult cohorts were both matched on receptive verbal abilities, whereas the adolescent cohorts were matched on non-verbal abilities. However, a confirmatory ANOVA was conducted to rule out any effects of lower language abilities in the adolescent groups and this failed to change the pattern of results. Additionally, both the child and adolescent cohorts included intellectually lower-functioning ASD individuals, whilst the adult groups were all intellectually high-functioning. There was also some overlap in chronological age between the child and adolescent cohorts. However, within each cohort groups were closely matched in terms of chronological age and intelligence and any inconsistencies in the findings arising from these factors would be expected to affect the ASD and control groups to a similar extent in the ANOVA and correlation analyses.

Conclusions

The findings from the present study represent the first attempt to examine the developmental trajectory of pitch discrimination and it's underlying mechanisms in individuals with and without ASD. Future studies should seek to employ a longitudinal, cohort design, in which ASD groups are carefully matched on diagnostic measures and verbal and non-verbal intelligence. In addition to more closely examining the relationship between sensory processing abnormalities and receptive vocabulary, future studies should also seek to examine the interaction between the development of these abilities and co-occurring perceptual and semantic processing biases in these individuals.

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