

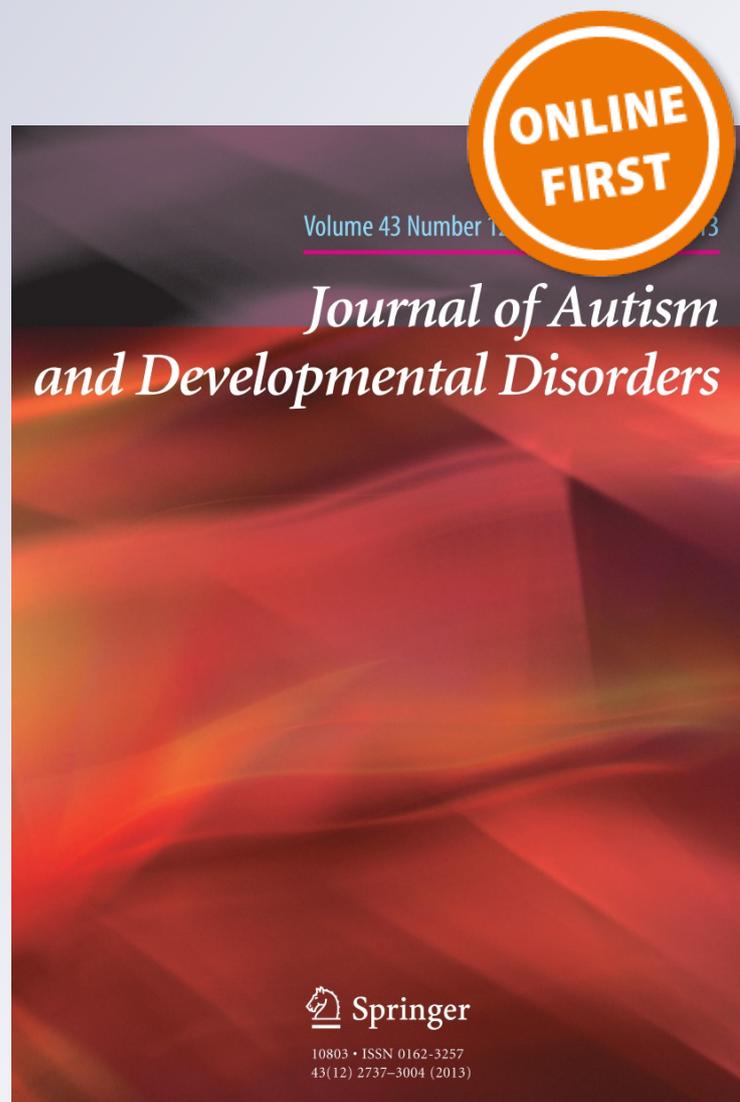
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Auditory Stream Segregation in Autism Spectrum Disorder: Benefits and Downsides of Superior Perceptual Processes

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Abstract Auditory stream segregation allows us to organize our sound environment, by focusing on specific information and ignoring what is unimportant. One previous study reported difficulty in stream segregation ability in children with Asperger syndrome. In order to investigate this question further, we used an interleaved melody recognition task with children in the autism spectrum disorder (ASD). In this task, a probe melody is followed by a mixed sequence, made up of a target melody interleaved with a distractor melody. These two melodies have either the same [0 semitone (ST)] or a different mean frequency (6, 12 or 24 ST separation conditions). Children have to identify if the probe melody is present in the mixed sequence. Children with ASD performed better than typical children when melodies were completely embedded. Conversely, they were impaired in the ST separation conditions. Our results confirm the difficulty of children with ASD in using a frequency cue to organize auditory

perceptual information. However, superior performance in the completely embedded condition may result from superior perceptual processes in autism. We propose that this atypical pattern of results might reflect the expression of a single cognitive feature in autism.

Keywords Autism · Audition · Stream segregation · Perception

Introduction

Autism spectrum disorder (ASD) is a neurodevelopmental variant, diagnosed on the basis of altered peer-oriented behaviors and communication, restricted and stereotyped interests and atypical perception (APA 2000). In the auditory modality, individuals with ASD show superior capacities in discriminating sounds that vary in frequency (Bonnell et al. 2010; Bonnell et al. 2003; Jones et al. 2009) or intensity (Khalifa et al. 2004). They also possess superior musical abilities. Indeed, superior performance for melody discrimination (Heaton 2005; Mottron et al. 2000) and note labeling (Heaton 2003) have been reported in participants with ASD, who also show a particular interest in musical stimuli (Molnar-Szakacs and Heaton 2012). Most cognitive theories in autism have interpreted these results as a superior ability to process local elements, but they differ on how they account for its origin. The enhanced perceptual functioning (EPF) model (Mottron et al. 2006) proposes that superior pitch discrimination is one among multiple examples of superior low and mid level perceptual processes in autism. The weak central coherence theory (WCC) claims that this superiority is due to a more detail-focused cognitive style and a difficulty to link elements together (Happé and Frith 2006). However, both theories

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agree that the superior process of local elements is a major component of autistic cognition, likely to influence autistic behavior.

Despite their superior abilities for processing pure tones and musical stimuli, autistic individuals have difficulty in perceiving speech in noise. At the behavioral level, this difficulty is often highlighted through a clinical caregiver questionnaire, the Short Sensory Profile (Dunn and Westman 1997). Indeed, children with ASD are typically reported as having difficulty to filter auditory information based on reports like “is distracted or has trouble functioning if there is a lot of noise around” or “appears to not hear what you say” (Tomchek and Dunn 2007). A few studies have experimentally evidenced the difficulty of autistic people to perceive speech in noise. They reported that participants with ASD required a higher signal-to-noise ratio than controls to correctly detect a syllable (Alcántara et al. 2004), or perceive a monosyllabic word or a sentence in a noisy background (Bhatara et al. 2013; DePape et al. 2012). These findings suggest difficulties in filtering auditory information, and are supported by data from auditory ERPs (Russo et al. 2009).

The ability to filter auditory information refers to a more general ability for auditory stream segregation (i.e., the ability to segregate sounds produced by distinct sources) that is critical for distinguishing speech in a noisy environment. An auditory stream is “the percept of a group of successive and/or simultaneous sound elements as a coherent whole, appearing to emanate from a single source” (p.919, Moore and Gockel 2012). To perceive distinct streams, the auditory system performs an auditory scene analysis (Bregman 1990). The auditory scene analysis relies on the simultaneous and sequential organization of auditory information. The present study focuses on auditory information sequential organization, which refers to the ability to perceptually link successive auditory events coming from the same source over time (i.e., integration), together with segregating them from sounds coming from distinct sources. Stream segregation can rely on the analysis of different acoustical cues such as sound frequency, timbre, or tempo (Bregman 1990). Sounds sharing similar acoustic cues (i.e., frequency) are perceived as coming from the same auditory source whereas sounds with different cues are analyzed as coming from distinct sources and are thus segregated by the auditory system.

Bregman (1990) assumes that streaming involves both primitive and schema-based processes. The primitive process is a perceptual mechanism, based on psycho-acoustic cues like frequency, intensity or perceptual harmonic coherence, which allows initial parallel processing of acoustic signals coming from different sources. This bottom-up mechanism decomposes the incoming sound into distinct auditory streams. The secondary, schema-based

mechanism requires attention and is a knowledge-driven, top-down selection mechanism. It helps to match the incoming stream with memory-stored knowledge. Shinn-Cunningham and Best (2008) proposed that correct identification of an auditory object (i.e., a sound that can be differentiated from another) results from the competition between perceptual information and top-down mechanisms (which features to attend to); the more unique the target object's features, the more effective the selection (driven by attention processes).

A single study has investigated auditory stream segregation in ASD. Using electroencephalography, Lepistö et al. (2009) explored stream segregation of pure tones varying in frequency in Asperger and typical children. When two distinct streams were presented, Asperger children showed a mismatch negativity, reflecting the automatic detection of an auditory change, of lesser amplitude than typical participants. No group difference was observed in the single stream condition. This result was interpreted as evidence of less efficient auditory segregation in Asperger children. As acknowledged by the authors, this difficulty to segregate streams varying in frequency was rather unexpected, considering that preserved auditory abilities are well documented in autism, including preserved frequency processing (for a review see O'Connor 2012). Based on previous evidence that stream segregation was not only dependent on frequency discrimination ability (Rose and Moore 2005) but further involved higher-order processes, like attention, integration of prior context or schematic knowledge (Snyder and Alain 2007), the authors interpreted their findings as reflecting a limitation in the influence of top-down processes in Asperger children.

To further explore the auditory stream segregation capacity of children with ASD, we used an interleaved melody recognition task paradigm (Bey and McAdams 2002, 2003; Dowling 1973). In this paradigm, an unfamiliar melody (the probe) is presented first, followed by a mixed sequence of two melodies. The mixed sequence is composed of a similar or modified version of the probe melody interleaved with a distractor melody. The target and distractor melodies can be separated by different mean frequency values (see Fig. 1). Participants have to decide if the probe melody is present or not in the mixed sequence. Previous findings have shown that typical adult participants were unable to recognize the target melody when the mixed sequence was composed of two completely interleaved melodies. This failure indicated that they perceived the mixed sequence as a single stream (Bey and McAdams 2002). However, performance in recognition of the target melody improved when the differences in frequency between the two melodies increased. Better melody recognition in conditions involving frequency separation indicates that listeners were able to compare the target

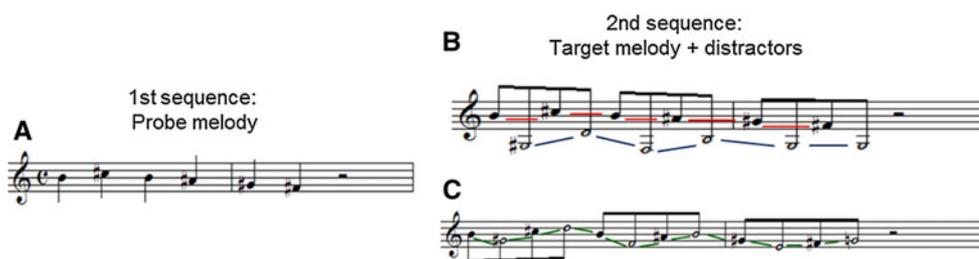


Fig. 1 Illustration of interleaved melody recognition task. First the probe melody is presented alone (**a**, filled circles, left side) followed by a mixture (**b** and **c**) made up of the target melody interleaved with a distractor melody (open circles). **c** represents the target and distractor melodies completely interleaved and **b** the target and

distractor melodies separated by a frequency value. When melodies are completely interleaved, a single stream is perceived (in green). When melodies are separated by a certain frequency, two streams are perceived (in red and in blue) (Color figure online)

melody to the probe melody, as the two melodies were segregated and thus perceived. The interleaved melody recognition task is thus viewed as providing an indirect but more objective measure of auditory stream segregation than the direct paradigm based on judgment of switching (Bey and McAdams 2002; Moore and Gockel 2012).

The single study exploring stream segregation among typical children with a direct paradigm showed that school-age children were able to perceive two streams when separated by 9 semitones (ST) (Sussman et al. 2007). In the absence of evidence from an indirect stream segregation paradigm similar to the one we used, we expected typical children to perform correctly in our task in conditions where melodies are separated by more than 9 ST. Moreover, based on the findings of Lepistö et al. (2009), showing that autistic participants have difficulty in using frequency as a cue to segregate streams, poorer performance was expected in all conditions of frequency separation in children with ASD compared to typical children, regardless of stimuli type and distance frequency between streams. In contrast, no difference between groups is expected when the two melodies are completely interleaved, as even typical adults fail to recognize the target melody in this condition (Bey and McAdams 2002).

Methods

Participants

Autism Spectrum Disorder (ASD) Group

Fourteen children with ASD were initially recruited in two expert centers in the Grenoble area: the “Centre Alpin de Diagnostic Précoce de l’Autisme” (CADIPA, Grenoble, France) and the “Centre d’Evaluation Savoyard de l’Autisme” (CESA, Chambéry, France). Exclusion criteria included personal or family history of psychiatric disorders other than autism, and/or a full scale IQ inferior to 70. All

children attended ordinary school and were therefore able to understand the instructions and feedback during the experiment. Data from two participants were excluded from the analyses because their error rates were 2 SD above their group performance in 3 of the 4 experimental conditions. The 12 remaining children with ASD (11 males) had an average age of 9.5 (SD = 2.02, range 7–13). IQ was measured using the Wechsler scale (WISC, WPPSI), except for one child who was administered the K-ABC scale that highly correlates with Wechsler measures in the French population (Laffaiteur et al. 2001). The ASD group showed a mean Verbal IQ of 90.45 (SD = 13.93, range 74–118) and a mean Performance IQ of 96.27 (SD = 20.53, range 77–138). Autism diagnosis was made using the Autism Diagnostic Interview Revised (ADI-r, Lord et al. 1994). Nine participants had cut-off scores in all three sections of the ADI-r. Two participants scored under the cut-off in the repetitive behavior section. For these two participants, the diagnosis of autism was confirmed using the Autism Diagnostic Observation Schedule-Generic (ADOS-G, Lord et al., 2000). For one participant, the ADI-r and ADOS-G scores were not available. The diagnosis was made by autism experts through clinical evaluation based on DSM-IV criteria. All participants had normal auditory acuity. Informed written consent was obtained from the children’s parents.

Control Group

Fourteen typically developing children were recruited from the urban community of Grenoble (France). Parents gave informed consent for the children’s participation. No IQ measure was available for control children but they all attended regular school and had normal schooling. They had normal auditory acuity and no history of neurological or psychiatric disorder. Performance of one control participant was excluded because of answering at random during the experiment. Mean age of the remaining group of 13 control participants (9 males) was 9.3 years (SD = 1.4,

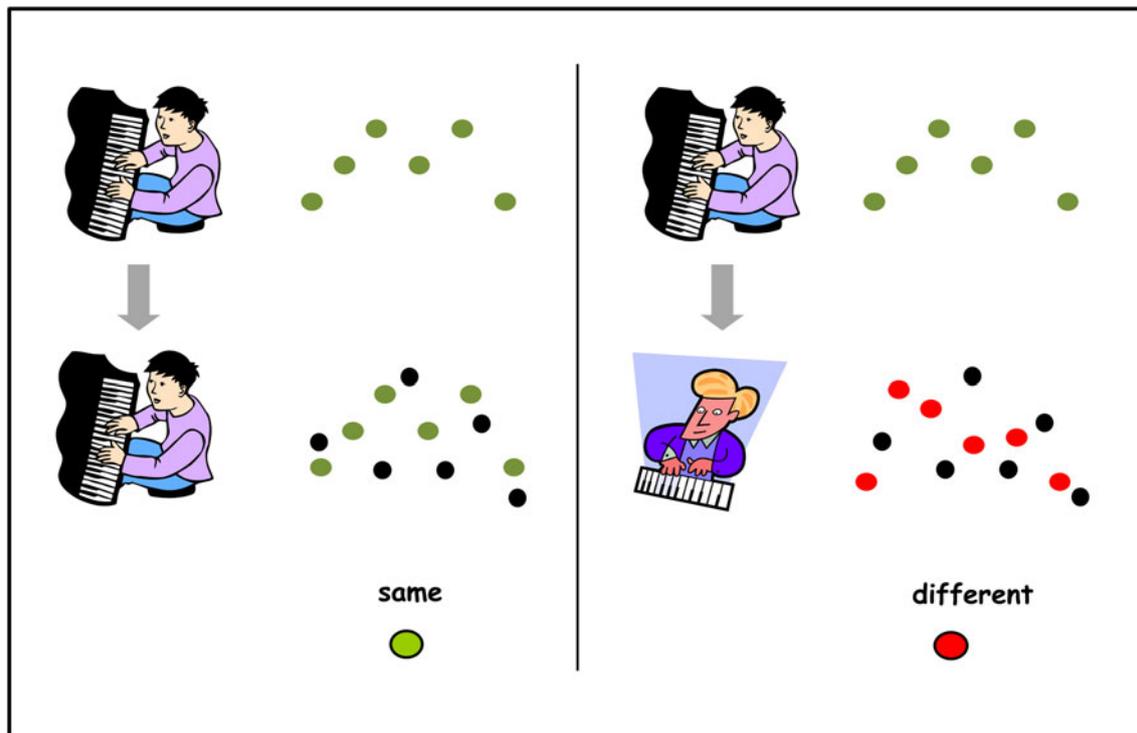


Fig. 2 Schematic illustration of the instructions provided to participants

range 7–13). The autistic and control group did not differ in either chronological age ($p = .77$, ns) or level of schooling (i.e., between first and fifth grade).

Material

Seventy-two melodies inspired from Bey and McAdams (2003)'s study were created using Final software (www.finalemusic.com). Among these 72 melodies, 36 were target melodies and 36 were distractor melodies. They were composed of six notes of 400 ms duration, each played with piano timbre for a total duration of 2,400 ms. Within each melody, intervals between notes varied from -3 to $+2$ ST¹ around the A4 note. Each target melody was presented in an original and a modified version. In the modified version, the second and fourth or the third and fifth notes were modified within a range of ± 4 semitones. Changes altered the original melodic contour. Two interleaved melodies, the 6-tone target melody and a 6-tone distractor melody, composed the mixed sequences. The target melody and the distractor could be separated by four ST values (0, 6, 12, 24) obtained by transposing the distractor sequence to lower frequencies (see Figure 1). The

first note of the mixed sequence always belonged to the target melody.

Procedure

The assessment of the ASD group took place at home, in a quiet room. Control children were tested individually at school in a quiet room. Assessment began with a training phase, followed by a test phase. In the two phases, a probe melody was first displayed followed 1,000 ms later by a mixed sequence composed of a similar or modified version of this same melody, called the target melody, interleaved with distractor tones.

Before each trial, the word “Ecoutez” (listen) was visually presented for 2,000 ms associated with a bip sound (1a4) lasting 300 ms. The instructions were orally given to the participants while they were presented a schematic illustration of the task (see Fig. 2). The instructions were as follows: “you are going to hear a first melody, followed by a mixed sequence made of two interleaved melodies. In some cases, the exact first melody will be repeated in the mixed sequence but interleaved with a distractor melody. In other cases, the first melody is not repeated.” The participants were told to press the green button when the target melody was present in the complex stream and the red button if it wasn't. They were asked to listen carefully because it was sometimes difficult to recognize the first melody in the complex stream. The training phase was

¹ A semitone is the smallest musical interval in the Western tonal musical range. On a piano keyboard, a semitone is the distance between two adjacent keys.

made of 16 trials (8 different) of increasing difficulty (4 trials by conditions of ST separation starting with the 24 ST condition). The training melodies were not used in the test session. Visual feedback (the words “Bravo” in blue or “You Lose” in red) was provided to participants during the training phase only. The test phase was composed of 48 trials, with 12 trials by ST condition (6 different), with no feedback. To avoid the succession of two identical target melodies, trials were pseudo randomly chosen among the 64 possibilities in each four ST conditions for the identical and different trials. During the test phase, the four conditions of ST separation were presented to participants in a counterbalanced order. The experiment lasted about 15 min. Experiments were controlled by E-prime software and stimuli were presented via Sennheiser HD 212Pro headphones at 60 dB SPL.

Results

Hit rates (proportion of “different” answers when the probe and the comparison melodies were actually different) and false alarm rates (proportion of “different” answers when the two melodies were actually the same) were computed across the four conditions for each participant. From these rates, two indices measuring perceptual sensitivity (the d' index) [$d' = Z(\text{hits}) - Z(\text{false alarms})$] where Z is the inverse of the normal distribution function] and decision criterion (the c index) [$c = -0.5 * (Z(\text{hits}) + Z(\text{false alarms}))$] were computed according to the “Signal Detection Theory” (Macmillan and Creelman 1991). Performing analysis on d' rather than raw scores is particularly recommended in discrimination tasks to control for participants' errors.

Perceptual Sensitivity (d')

Data (d') was analyzed with a 2×4 ANOVA with Group (AS, control) as between-subject factor and ST condition (0, 6, 12, 24) as within-subject factor. Results are presented in Fig. 3. A main effect of ST condition was found, $F(3,69) = 3.06$, $p < .05$, $\eta_p^2 = .11$. Performance increased linearly with the number of ST between melodies, $F(3,63) = 7.34$, $p < .05$. Analysis also revealed that the control group performed better than the ASD group, $F(1,23) = 18.71$, $p < .001$, $\eta_p^2 = .45$ [$M dif = 0.64$, 95 % CI (0.33, 0.95)]. The Group by ST condition interaction was significant, $F(3,63) = 9.02$, $p < .001$, $\eta_p^2 = .28$. In order to explore this interaction and compare the group's performances in each condition, between group t tests adjusting for multiple comparisons (Bonferroni correction, $\alpha = 0.05/4 = .0125$) were carried out. The ASD group showed better performance in the 0 ST condition compared

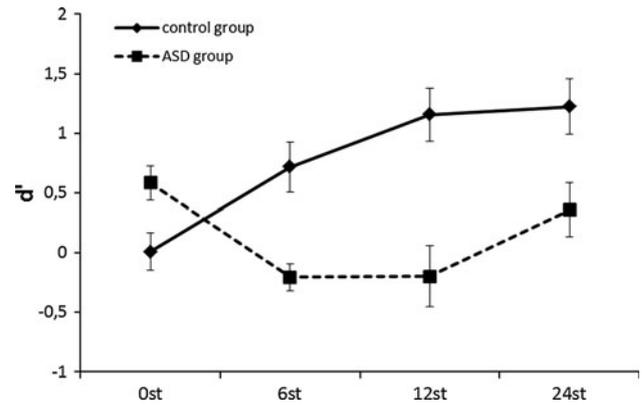


Fig. 3 Mean performance (d') in the segregation task as a function of the four conditions of mean frequency separation [0, 6, 12 and 24 ST] for the control (solid line) and ASD group (dashed line). Vertical bars represent standard errors

to the control group, $t(23) = 2.71$, $p = .012$, $M dif = 0.57$, 95 % CI (0.13, 1.01). In contrast, the control group performed better than the ASD group in both the 6 ST condition, $t(23) = -3.86$, $p < .001$, $M dif = -0.92$, 95 % CI (-1.41, -0.42), and the 12 ST condition, $t(23) = -4.01$, $p < .001$, $M dif = -1.35$, 95 % CI (-2.05, -0.66). In the 24 ST condition, the difference between groups was marginally significant, $t(23) = -2.66$, $p = .014$, $M dif = -0.86$, 95 % CI (-1.54, -0.19). Lastly, whereas the control group performance was above chance level in all conditions ($p < .005$) except for the 0 ST condition ($p = .95$), the ASD group showed the reverse pattern. ASD participants performed at chance level in the 6, 12, and 24 ST conditions that were easier for the controls ($p > .1$) but they performed above chance level in the 0 ST condition ($p < .005$) that was the most difficult for controls.

Response Bias or Decision Criterion (c)

A negative c indicates a bias towards “Different” answers and a positive c indicates a bias towards “Same” answers. A c close to 0 indicates an absence of bias. To explore whether the ASD or control groups showed any response bias, c values were first compared to 0. The ASD group showed no bias in any ST condition (0, 6 and 24 ST condition, $p > .2$; 12 ST condition, $p > .05$). The control group showed a negative bias in the 0 ST condition ($p < .001$) but a positive bias in the 6, 12 and 24 ST conditions ($p < .001$). C values were then analyzed with a 2×4 ANOVA with Group (AS, control) as between-subject factor and ST condition (0, 6, 12, 24) as within-subject factor. Results are presented in Fig. 4. There was a main effect for the ST condition, $F(3,69) = 18.43$, $p < .001$, $\eta_p^2 = .44$. Compared to the other conditions, participants showed a negative response bias in the 0 ST

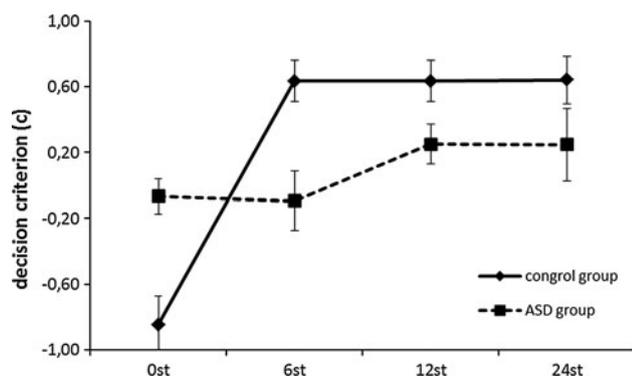


Fig. 4 Mean criterion decision (c) in the segregation task as a function of the four conditions of mean frequency separation [0, 6, 12 and 24 ST] for the control (*solid line*) and ASD group (*dashed line*). Vertical bars represent standard errors. A criterion of 0 indicates no response bias; a positive criterion indicates a bias towards “Same” response, a negative criterion indicates a bias towards “Different” response

condition, $F(1,23) = 41.51$, $p < .001$ [$M dif = 2.53$, 95 % CI (1.72, 3.35)]. No main Group effect was observed, $F(1,23) = 2.06$, $p = .16$. However, the Group by ST condition interaction was significant $F(3,69) = 10.92$, $p < .001$, $\eta_p^2 = .32$. To explore this interaction and compare the groups’ performance in each condition, we carried out between groups t tests adjusting for multiple comparisons, using the Bonferroni correction ($\alpha = 0.05/4 = .0125$). In the 0 ST condition, the control group showed a stronger negative bias, $t(23) = 3.79$, $p < .001$, $M dif = 0.78$, 95 % CI (0.35, 1.21) than children with ASD, while they showed a stronger positive bias in the 6 ST condition, $t(23) = -3.39$, $p < .005$, $M dif = -0.73$, 95 % CI (-1.17, -0.28). There was no significant difference between groups in the 12 and 24 ST conditions ($p > .03$).

Discussion

We used an interleaved melody recognition task as an objective and indirect measure of stream segregation (Bey and McAdams 2002) in order to explore streaming abilities in ASD. Based on previous evidence that Asperger children have difficulty in using frequency cues for segregating auditory streams (Lepistö et al. 2009), we expected children with ASD to perform more poorly compared to typical children in all conditions of frequency separation (6, 12, 24 ST). No difference between groups was expected in the 0 ST condition, as even typical adults fail to recognize the target melody in this later condition (Bey and McAdams 2002). When engaged in the melody recognition task, children with ASD discriminated completely embedded melodies (0 ST condition) more accurately than typical participants. In contrast, children with ASD showed poorer

discrimination performance when the target and distractor melodies were separated by 6–24 ST. They performed at chance level in the ST conditions that were easier for the controls, but above chance level in the 0 ST condition in which controls failed. Thus, current findings suggest that ST separation is used as an acoustical cue to segregate auditory streams in typical children but not in ASD participants. Globally, our results are consistent with previous findings reported by Lepistö et al. (2009), that contrary to controls, individuals with ASD do not use acoustical cues in stream segregation.

In order to investigate the involvement of bottom-up and top-down processes in stream segregation, Bey and McAdams (2002) used an interleaved melodies recognition task where the probe melody can be presented before (as in the current study) or after the mixed sequence. In the before condition, participants have a precise knowledge of the to-be-detected melody which was not the case in the after condition. Comparing these two conditions, the authors intended to investigate the influence of previous knowledge on stream segregation. They observed that when melodies of mixed sequence are completely interleaved participants performed similarly in both conditions. However, when melodies of the mixed sequence are separated by a least one ST, participants performed better when the target melody is presented before rather than after the mixed sequence. They concluded that top-down processes were more likely to operate when melodies were separated by a minimum value of one ST. In this case, listeners can use the frequency cue in order to group notes of the same melody together, to focus on the target melody and to ignore the distractor melody. However, in the 0 ST condition, organization of the auditory perception is not possible as melodies are completely embedded and perceived as belonging to the same stream. Consequently, in order to perform the task in this specific condition, participants can only rely on perceptual processes.

In the current study, we observed that typical and children with ASD used different response strategies in this 0 ST condition as indicated by analysis on criterion decision. Indeed, typical children showed a negative bias (higher false alarm rate) whereas children with ASD did not show any bias. This indicates that control children detected a difference that was not present, maybe in integrating distractor melody into the same stream of tones of the target melody. This suggests that contrary to children with ASD, typical children were not able to extract the target melody from the mixed sequence in this particular condition. However, in other ST conditions, typical children showed a bias toward the “same” answer. This bias contrasts with the negative bias observed in the 0 ST condition. This change of response criterion reflects a change of strategy of

typical children which was induced by the frequency separation between melodies.

In contrast, children with ASD showed a constant response strategy across conditions. They made false alarms and misses in the same proportion and thus showed no bias toward any specific answer. This indicates that children with ASD retain the same strategy during the whole task. This change of strategy response in typical children between 0 and 6 ST condition reflects the use of the frequency cue in order to organize their perception and to perceive distinct streams. Considering that we did not observe any change of strategy among children with ASD, we can consider that they failed to use this frequency cue in order to group notes of the same melody together.

According to two main accounts of autistic perception, EPF (Mottron et al. 2006) and WWC (Happé and Frith 2006), autistic cognition is characterized by superior process of local elements. This superiority could explain the superior performance of children with ASD in the 0 ST condition, where melodies are completely interleaved. Indeed, if children with ASD have a greater ability to process notes individually, they could perform in this condition more efficiently than controls, which was indeed found. The WCC theory (Happé and Frith 2006) supposes that this superior processing of local elements in autism is due to a weaker ability to perceive elements together. Thus, as individuals with ASD process notes separately, they could therefore have difficulty linking notes into a coherent stream. This might have prevented them from using the frequency cue in order to integrate the notes of the target melody into a distinct stream. Evidence of a difficulty to use a top-down strategy to group elements together in autism has been observed in gestalt tasks, especially on the grouping by similarity task (Bölte et al. 2007; Falter et al. 2010). Consequently, a superior capacity to process notes individually and a difficulty to group notes together might explain the performance pattern of the ASD group according to the WCC theory (Happé and Frith 2006).

Another explanation of the impaired performance of the children with ASD in conditions involving frequency separation (6, 12, and 24 ST conditions) is to consider it as the reflection of a selective attention deficit. Indeed, it has been reported that when engaged in an attention-requiring perceptual task, a large physical difference between standard and deviant stimuli can lead to enhanced distraction (Berti et al. 2004; Yago et al. 2001). In conditions involving frequency separation, as melodies become physically more distinct, they cause enhanced distraction. This superior distractibility involved in frequency separation cannot be ignored by ASD participants if they present selective attention impairment. Previous studies have reported an impairment in selective attention in ASD (Burack 1994; Ciesielski et al. 1990) as well as lower

resistance to interference from irrelevant elements when engaged in a flanker visual filtering task (Adams and Jarrold 2012; Christ et al. 2011). Impairment in selective attention processes has been recently attributed to superior perceptual capacities (Remington et al. 2009, 2012). Indeed, in manipulating various degrees of the perceptual load of the task (Lavie 2010), Remington and colleagues showed that autistic individuals required higher levels of perceptual load to successfully ignore distractors without diminishing overall performance. They interpreted this result as the consequence of superior perceptual capacities in autism in the EPF model (Mottron et al. 2006). This is relevant to the current study, as superior perceptual capacities could therefore have led children with ASD to process distractor notes in condition 6, 12 and 24 ST and to fail to establish a grouping attentional strategy. On the contrary, typical children were able to use the frequency cue as a grouping strategy and to inhibit distractor notes. In other words, superior perceptual capacities in autism might have a deleterious impact on task in which a grouping strategy is needed.

Our results confirm that ASD participants have difficulty using a frequency cue to segregate streams, as previously reported (Lepistö et al., 2009). As auditory abilities are preserved in autism (O'Connor 2012), we argued that a selective attention deficit combined with wider perceptual abilities in autism may account for the current findings. These results provide support for the different cognitive theories of autism (EPF and WCC) where a cognitive feature such as superior perceptual processes or a local bias could have beneficial or detrimental effects on performance depending on the difficulty of the task.

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