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TECHNICAL REPORT

Auditory perception in atypical development: From basic building blocks to higher-level perceptual organization

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Auditory perception in atypical development: From basic building blocks to higher-level perceptual organization

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Abstract

We examined auditory pattern perception in the neurodevelopmental disorder Williams Syndrome (WS), investigating cues and strategies used in organizing sound patterns into coherent units. In Experiment 1, we investigated the streaming of sound sequences into perceptual units, on the basis of pitch cues, in a group of children and adults with WS compared to normal controls. Participants had to judge whether or not a sound sequence contained a target integrated rhythm (a galloping pattern). We showed that individuals with WS were sensitive to the same pitch cues as typical children and adults when streaming these patterns. This led to Experiment 2, in which we evaluated differences in reliance on pitch and contour cues in unfamiliar melody perception in a group of adults with WS relative to normal control children and adults. Participants judged if two pure tone sequences were the same or different on the basis of pitch and contour cues. Unlike controls who demonstrated greater proficiency when contour cues were available, adults with WS showed no such advantage. Based on these findings, we suggest a preliminary integrative account of how auditory functioning in WS relates to the uneven profile observed in this developmental disorder, in which language and music eventually emerge as relative strengths.

Introduction

Different domains vary substantially in the nature of their inputs and their specific computational goals, e.g., recognize a familiar tune or parse a linguistic structure. Furthermore, there is little controversy that the adult brain employs specialized mechanisms dedicated to handling specific inputs where certain regions are consistently activated in response to stimulus categories corresponding to these domains. There remain, however, unresolved questions not only regarding the extent to which different domains share common underlying mechanisms, but also in relation to how the adult brain reaches this specialized and complex state.

These questions have been fruitfully addressed in a body of literature comparing the development and processing of language and music. Despite their clear differences, both domains appear to be universal, their inputs are highly structured, and both become progressively specialized in the adult brain. Theoretical models have proposed that commonalities between language and music are reflected in various ways, including similar processing mechanisms in the adult brain (Friedereci, Maess, Koelsch, & Gunter, 2001; Heiser, Iacoboni, Maeda, Marcus, & Mazziotta, 2003; Kan & Thompson-Schill, 2003; Patel, 2003; Patel, Gibson, Ratner, Besson, & Holcomb, 1998; Peretz & Coltheart, 2003), shared developmental mechanisms (McMullen & Saffran, 2004; Trehub, 2003), and common evolutionary origins as forms of communication (Huron, 2003; Wallin, Merker, & Brown, 2000).

In adults, for example, the processing of absolute pitch, which is frequently viewed as a uniquely musical gift, turns out to be modulated by language experience. Deutch and colleagues (2004) found that speakers of tone languages like Mandarin make use of absolute pitch as a linguistic cue in lexical processing rather than only as a musical cue as do speakers of other languages like English (Deutsch, Henthorn, & Dolson, 2004). Furthermore, linguistic and musical syntax overlap with respect to their neural correlates (Patel et al., 1998; Maess et al.,

2001). In a study using Event-Related Potentials (ERPs), Patel and colleagues found similar patterns of neural activation in the same group of participants in response to violation of rule-governed phrase structures for both music and language, (Patel et al., 1998). Interestingly, the notion of shared processing mechanisms of music and language is accepted even in theoretical models which view these domains as functionally separable, or 'modular', in the adult brain (e.g., Peretz & Coltheart, 2003). The latter models differ in the extent to which different domains overlap, where it is proposed that a few shared general mechanisms process all forms of auditory input but these inputs are subsequently processed by domain-specific components.

The possibility that shared mechanisms underlie these different domains has also become increasingly a focus in the developmental literature. McMullen and Saffran (2004) reviewed commonalities in the acquisition process of music and language in infancy. More than adults, naïve infant learners whose task is discover structure in any form of perceptual input, are likely to find these domains more similar than they are different. Both domains are acquired on the basis of incidental exposure, where the infant is provided with a rich set of cues that can be used in discerning the elements and the structure in those two domains. These cues are available from various sources, including distributional, rhythmic, and prosodic cues. A number of studies have documented the remarkable learning capacities both infants and adults employ in tracking statistical patterns in various forms of input including linguistic and musical input in order to discover patterns and regularities (Newport & Aslin, 2000; Saffran, 2003). Furthermore, various studies (reviewed in Jusczyk, 1997; 1999) have highlighted an important role of prosodic and rhythmic cues in language acquisition, beginning in the prenatal period (Mehler et al., 1988). Infants appear to use prosodic and rhythmic information, which correlates with syntactic boundaries, to segment continuous speech streams into its component units. Not only are infants sensitive to statistical and prosodic cues signaling boundaries, but caregivers appear to further enhance the intelligibility of some cues through providing patterned, repetitive, and exaggerated linguistic and musical inputs (Trehub, 2003).

In contrast with the notion that developmental or on-line processing mechanisms can be either domain-specific or domain-general, these adult and developmental approaches emphasize that common, domain-relevant computations are important precursors to the specialized mechanisms which progressively emerge over developmental time (Karmiloff-Smith, 1998). Common to these approaches is the view that for each of these domains the brain is presented with a similar problem where it has to form coherent percepts through discovering perceptual units and discerning patterns in highly structured auditory input.

These approaches have also demonstrated that shared processes and developmental origins of language and music can be elucidated through examining how general auditory input is structured and organized. For instance, auditory pattern perception is one of the domain-relevant mechanisms deemed important in the development as well as the on-line processing of language and music. Presented with continuous inputs, auditory pattern perception serves not only to segment musical or linguistic streams into units but also to discern the relations among the elements.

Despite these promising directions in adult and developmental research, investigations of developmental disorders such Williams Syndrome (WS) have in the main focused on describing the end products of development in terms of patterns of proficiency and impairment in higher-level domains (e.g., Bellugi, Lichtenberger, Jones, Lai, & St. George, 2000; Pinker, 1999). Williams Syndrome has presented a paradox to the scientist: whereas in adulthood language and music processing stand out as particularly proficient relative to other domains, in infancy and toddlerhood WS language is seriously delayed. Yet young children with WS are fascinated by sound, while at the same time often suffering from an unusual sensitivity to sound, generally known as hyperacusis (Levitin, Cole, Lincoln, & Bellugi, 2005). Can theoretical approaches targeting processes of segmentation and integration help resolve this paradox?

It has long been claimed that domain-relevant mechanisms must influence behavioral outcomes across domains and over developmental time, in both typical and atypical circumstances (Elman et al., 1996; Karmiloff-Smith, 1998). Atypical strategies in organizing environmental input are likely to shape the emerging cognitive and behavioral profiles in atypical development. Hence, understanding how individuals with developmental disorders perceive and organize input, and what alternative constraints govern this organization, would clarify the process by which higher-level outcomes are achieved. Elucidating alterations in these domain-relevant

mechanisms in WS relative to normal controls help in understanding what gives rise to the uneven behavioral profile observed in this clinical population.

Interestingly, atypical behavioral and brain processes are found not only in areas of serious impairment but also in domains of relative proficiency like language and music processing in WS. In addition to being generally delayed in acquiring language, infants and toddlers with WS do not display the same order of appearance of various milestones such as pointing, gesturing, and the onset of the vocabulary spurt (Laing et al., 2002; Mervis & Bertrand, 1997; Singer-Harris, Bellugi, Bates, Jones & Rossen, 1997). In adulthood, language emerges as a strength relative to other abilities like visuospatial processing, yet it is marked by various processing problems which emerge when finer measures are used. Language in WS is more accurately described as a domain of relative proficiency, but not of sparing, of certain aspects like phonological representations, vocabulary, and some syntax, the result of which is language marked by superficial fluency. However, once the demands of the language task become more complicated, particularly in aspects of syntax and semantics, performance begins to suffer.

Less is understood regarding musical processing in Williams Syndrome. Anecdotal evidence and questionnaire reports indicate that individuals with WS are more attracted and exposed to music relative to controls (Levitin et al., 2005). Some learn to sing and play instruments, with a few achieving high levels of proficiency, which has led some researchers to describe music as a preserved domain in WS (e.g., Lenhoff, 1998; Levitin & Bellugi, 1998; Levitin, Cole, Chiles, Lai, Lincoln, & Bellugi, 2004). Two studies have evaluated musical abilities using standardized tasks and found that while individuals with WS displayed similar performance to verbal-age matched controls, they were still overall significantly worse at all tasks relative to what is expected for their chronological age (Don, Schellenberg, & Rourke, 1999; Hopyan, Dennis, Weksberg, & Cytrynbaum, 2001). Levitin and colleagues (Levitin et al., 2004) disagree with the results of these studies, citing methodological limitations of the tasks. Their own informal study of rhythm production (Levitin & Bellugi, 1998) showed that a group of musically-trained individuals with WS was able to reproduce rhythms of varying complexity at a level comparable to mental age-matched controls who were also musically trained. A functional brain imaging study of music perception in WS showed that patterns of

activation when listening to music vs. noise were less distinct in individuals with WS than in controls, with the clinical group showing more variable and diffuse patterns of cortical activation, together with abnormally high levels of activation in the amygdala and cerebellum (Levitin et al., 2003). While more research is needed to resolve these differing claims, it appears that at least some individuals with WS develop musical proficiency, albeit relying on atypical pathways.

The combined results from research on language and music indicates that explaining the behavioral profile in WS needs to go beyond describing patterns of proficiency or impairment in various domains. Instead, targeting domain-relevant mechanisms of developmental change would highlight the differences in how input is perceived and organized, providing the basis for explaining the resulting behavioral profiles. The goal of the current study was to examine the processes which structure and organize perceptual input in the auditory domain in WS. The rationale is that targeting processes of auditory pattern perception in this syndrome can provide important, albeit indirect links to language and music processing in this clinical group.

To investigate these processes, we used two auditory paradigms which involve the manipulation of important cues used by typically developing individuals to structure auditory sequences. The aim was to test whether individuals with WS benefit from these cues to the same extent as healthy controls. If we find differences in reliance on some cues relative to others, this would provide evidence for atypical strategies used in discovering structure in auditory input, which will in turn have important implications for the development and processing of language and music in this population.

In the first paradigm (Experiment 1), we investigated the use of pitch cues in perceptual streaming, i.e., the perception of segregated or integrated patterns. In the second paradigm (Experiment 2), we investigated differences in reliance on pitch and contour cues in the processing of unfamiliar melodies. Highlighting potential differences in reliance on different cues is only one goal of our investigation. The second is to integrate findings from the present study with previous findings in the literature targeting domain-relevant mechanisms in WS to motivate a developmental account of why language and music (although the latter is less understood) turn out to be relative strengths in this population. We specifically selected the tasks in these experiments because they

employ auditory patterns which do not fit with the structure of Western music. The serves not only to control for differences in exposure to music as documented by other studies (Levetin et al., 2004) and in musical training, but also because we wanted to specifically target processes likely to be shared across domains, and not ones that are specific to music and would depend heavily on musical exposure and skills.

Experiment 1: Perceptual Streaming in Williams Syndrome

When presented with a sound sequence, listeners rely on a variety of cues to discover which elements are grouped and which are segregated. Among the most important cues are the spectral and rhythmic characteristics of sound sequences. When high and low tones are alternated continuously they either segregate perceptually into two streams or they fuse into a single coherent stream (Bregman, 1990). There are general principles according to which such auditory sequences are perceptually organized. These general principles of auditory processing also apply to language and music (Bregman, 1990; Deutch, 1999). For instance, the perception of these 'galloping' patterns, illustrated in Fig. 1, is influenced by multiple factors, including the frequency separation of the two tones and the rate of presentation. Frequency separation of the high and low tones, i.e., how different the two tones are, is the factor which has the greatest weight in altering perception of these patterns between a gallop (a single stream) or two segregated high and low streams (Bregman, Ahad, Crum, & O'Reilly, 2000). Streaming is the result of the integration of elements that are similar in frequency, which overrides their temporal proximity. The larger the frequency separation the more likely it is for the high and low tones to segregate into two perceptually separate streams. Streaming requires the integration of spectral information over a period of time, and is viewed as an auditory binding mechanism, resulting in optimal units for subsequent pattern perception (Bregman, 1990).
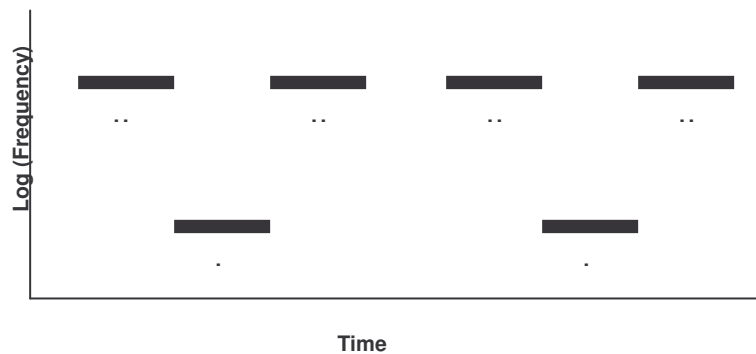

Figure 1. Galloping patterns used in Experiment 1. Depending on the frequency separation of the high (H) and low (L) tones, the sequences are perceived as an integrated galloping stream (HLH-HLH) or two separate high and low streams.

The aim of this experiment was to examine stream segregation in WS. More specifically, we aimed to discover whether children and adults with WS are sensitive to the same constraints as normal controls in perceiving these patterns. Although well-documented in the normal adult literature, to our knowledge this streaming phenomenon has never been investigated in children. Hence, we adapted the standard adult paradigm (e.g., Bregman et al., 2000) for use with normal children as well as individuals with mental retardation

Method

Participants

A group of 17 individuals with WS aged 9;11 to 56;10 (mean = 28;11) was recruited through the Williams Syndrome Foundation of the United Kingdom. Diagnosis was confirmed clinically and on the basis of genetic screening (FISH) confirming the deletion of Elastin. Normal hearing status was verified based on reports of audiological assessment.

The verbal mental age of the participants ranged from 3;1 to 17;0 (mean = 10;3) as measured on the British Picture Vocabulary Test (Dunn, Dunn, Whetton, & Pinitilie, 1997). A control group of 27 typical children and adults aged 8;6 to 54;1 years (mean = 19;1) was also recruited. The children in the group came from a primary school in North London. Normal hearing status was verified through reports of audiological assessment done at the school within the year. The adults were staff volunteers (mostly non-academic) from the UCL Institute of Child Health in London. Normal hearing status for the adults was determined on the basis of self-report. None of the participants had any extensive training in music.

Stimuli

Stimuli were triplets of high and low tones separated by 75ms gaps (HLH-HLH-….) as illustrated in Fig. 1. Each individual tone had a duration of 100 ms (20 ms onset and decay). On each trial, a sequence of 6 cycles was presented. Fig. 1 shows an example of the first two cycles of a sequence presented on a given trial. Five sequences were constructed using a baseline frequency of 500 Hz for the low tone and frequency separation levels of either 2, 6, 10, 14, or 18 semitones for the high tone. These differences between the high and low tones in each stimulus resulted in sequences which ranged perceptually from highly integrated to highly segregated. Stimulus presentation was done via headphones at a comfortable level of around 60 dB SPL.

Procedure

The experiment began with a familiarization phase in which participants were presented with the galloping pattern. They were given an explanation about the difference between a single galloping stream and two separate streams, using clearly segregated or clearly integrated stimuli. To help maintain focus of attention on the pattern itself, participants were instructed to listen for the gallop, and to try to judge if one was present or not on each trial. The familiarization phase continued until the experimenter ensured that the participant understood the difference between the two patterns. Their understanding was then verified by having them label three exemplars as 'Galloping' or 'Not Galloping'. A practice phase was then administered in which participants judged 10 sequences presented in random order, half of which were highly segregated (frequency separation = 20 semitone) and the other half were integrated (frequency separation = 1). In the test phase, the participant initiated the trials. A single pattern was presented on each trial, and the participant had to decide whether the galloping pattern was present or not, by responding verbally or by pressing one of two keys corresponding to these choices. In a given session, each frequency separation level was presented five times for a total of 25 trials.

Results and Discussion

Individual performance curves were constructed as a function of frequency separation to determine the Segregation point for each participant. This was defined as the frequency separation level at which the galloping pattern could not be detected in at least 4 out of 5 trials. Three participants from the WS group were excluded from subsequent analysis because their performance was not sensitive to the different levels of frequency separation. Hence, for these three participants, the key result, i.e., the Segregation point could not be determined.

Remaining data are shown in Fig. 2. The figure illustrates the proportion of responses corresponding to the perception of a gallop as a function of frequency separation for the two groups. The effects of Group membership as a categorical variable and Age as a continuous variable on the Segregation point were examined in a linear regression analysis. The Segregation point for the WS group (mean = 2.57, SD = 0.65) and for the typically developing group (mean = 2.67, SD = 0.68) did not differ significantly [$F(1, 37) = 0.35$, $P = 0.56$]. Age, too, was not a significant predictor of performance in the overall group [$F(1, 37) = 1.93$, $P = 0.17$].

We were further interested in verifying whether behavior during the task, i.e., responses across the different levels of frequency separation, depended on Age and Group. A repeated measures ANOVA revealed a main effect of Frequency separation as a within subjects variable [$F(1, 37) = 170.5$, $P < 0.001$], but no significant main effect of Group [$F(1, 37) = 1.1$, $P = 0.303$] or Age [$F(1, 37) < 1$, $P = 0.346$]. Furthermore, none of the interactions of Frequency separation and Group [$F(1, 37) = 1.1$, $P = 0.292$] or Frequency separation and Age [$F(1, 37) = 1.7$, $P = 0.200$] was significant. Because the groups did not differ overall, further analyses based on verbal mental age were not pursued.

The results show that typically developing children do not differ significantly from adults in their perceptual streaming on the basis of pitch characteristics of the sequences, supporting a pattern

of early developmental stability of this ability within the age ranges which were examined. This was evident not only from the values of the segregation point, but also from overall performance on the task. Performance of individuals with WS was also similar to that of typical children and adults, indicating that

the organization of sounds into perceptual streams on the basis of pitch has reached proficiency in this clinical group. But does this competence extend to all aspects of auditory processing in WS? We addressed this question in Experiment 2.
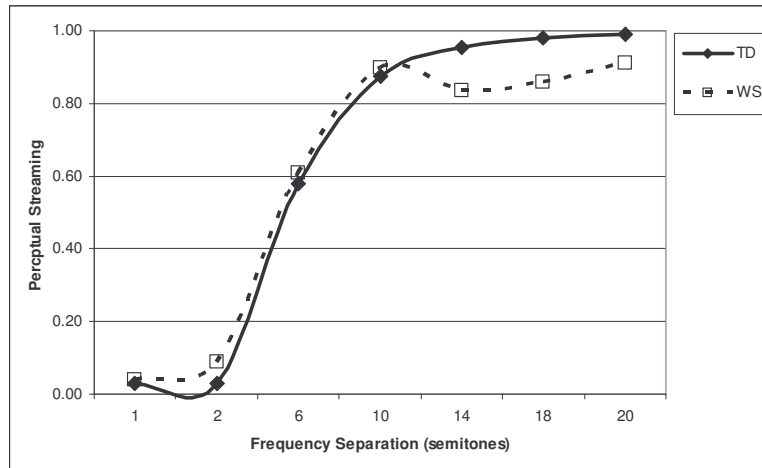

Figure 2. Perception of the galloping pattern (0 = gallop, 1= no gallop) as a function of frequency separation of the high and low tones in the WS and typically developing group (TD) in the practice and test phase in Experiment 1.

Experiment 2: Perception of Unfamiliar Melodies in WS

The previous experiment showed that individuals with WS are sensitive to the same cues as healthy controls when streaming sound sequences on the basis of pitch. But what about even higher levels of complexity, when pitch patterns are not only streamed, but organized into melodic patterns? Unfamiliar melodies provide the listener with various auditory cues: (a) absolute pitch values of the individual notes in the melody, (b) interval information, which is the exact relationship between adjacent units, and (c) contour information, which is the pattern of rise and fall of the notes. Typical infants as well as adults perform better when discriminating melody transformations in which the contour is violated than melody transformations in which the contour is preserved, i.e., the interval is violated (Dowling, 1978; Trehub & Trainor, 1993). Recently, Deruelle and colleagues (Deruelle, Schon, Rondan, & Mancini, 2005) tested a group of children with WS and an age matched control group on discrimination of unfamiliar melody pairs. The

control group performed better in the contour-violating condition relative to the contour-preserving condition, whereas children with WS showed equivalent levels of performance in both conditions. The goal of the current experiment was to extend the results of Deruelle et al. (2005) by testing the developmental stability of the differences in the utilization of interval and contour cues in a group of adults with WS.

Method

Participants

Fourteen individuals with WS (8 males), were recruited through the Williams Syndrome Foundation of the United Kingdom or through the Irish Williams Syndrome Association. The ages of the participants ranged from 16;4 to 56;9 years (mean = 31;7). Mean verbal mental age as assessed using the British Picture Vocabulary Test (Dunn et al., 1997) ranged from 4;9 to 17;0 (mean = 11;6).

Two control groups of typically developing individuals were also recruited. The first group was

composed of 14 children (6 males) aged 8;6 to 12;11 years (mean = 10;7). The children were recruited from a primary school in North London. The second control group comprised 14 adults (7 males) aged 19;5 to 58;10 years (mean = 34;7). The remaining participant characteristics were the same as in the previous experiment.
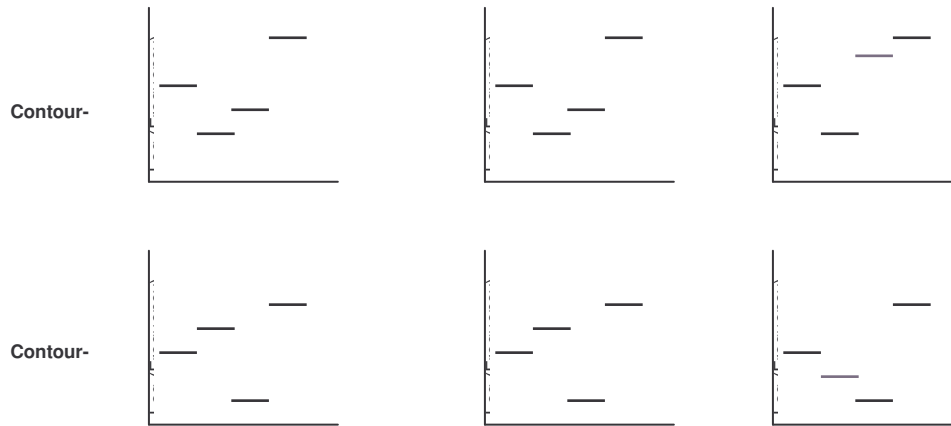

Figure 3. Examples of items in the unfamiliar melody perception task in Experiment 2.

Stimuli

Stimuli were pairs of pitch sequences separated by one second gaps similar to those used by Foxton and colleagues (Foxton, Dean, Gee, Peretz, & Griffiths, 2004). The sequences spanned either 4- or 5-notes. Individual notes were 250 ms each (20 ms rise and fall), generated from a logarithmic octave split into seven equally spaced notes. The lowest pitch was randomly selected from one of the following values: 250, 268, 287, 308 or 330 Hz. In the first condition (contour-preserving condition), same pairs contained identical sequences, whereas different pairs contained a standard melody and a melody that was changed by one note. The change was two notes in magnitude, and could occur at any position apart from the first and the last notes. The note change in this condition did not violate the pattern of rise and fall of pitch in the sequence. The second condition (contour-violating condition) was identical to the first condition except that the change in melody in this condition violated the pattern of rise and fall in the pitch sequence. Hence the contour-preserving condition provided the listener with interval cues only, whereas the contour-violating condition provided the listener with both interval and contour cues. Examples of the sequences in the two conditions are shown in Fig. 3.

Procedure

In each trial, participants judged if two melody sequences were the same or different, either by responding verbally or by pressing one of two keys which corresponded to 'same' or 'different'. Individual participants were tested on the two conditions (contour-preserving and contour-violating) in two separate blocks of 40 trials. Each block contained 20 identical pairs and 20 different pairs. Half of the participants in each group were randomly assigned to the 4-note condition, and the other half to the more difficult 5-note condition. A practice phase with 6 pairs of sequences (half the same and half different), preceded testing. Stimuli were presented through headphones at a comfortable level of around 60 dB SPL.

Results and Discussion

Fig. 4 presents mean accuracy expressed in d-prime scores for each group in the contour-preserving and in the contour-violating conditions. Results were transformed into d-prime scores to control for response bias among participants. This analysis compares hits relative to false alarms for each participant. Two questions were of interest in the analysis: First, how does overall proficiency of the WS group compare to that of typical children and

adults? Second, does performance of the WS group relative to the other groups differ in the contour-preserving vs. contour-violating condition? To answer these questions, a repeated measures ANOVA examined the effects of Condition (contour preserving, contour violating), Group (control children, control adults, WS), and Difficulty (4-notes, 5-notes) on performance accuracy.
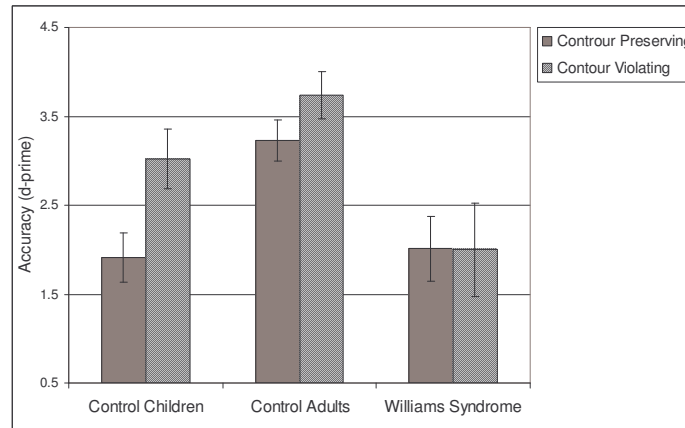

Figure 4. Accuracy scores for the three groups in the contour-preserving and contour-violating conditions in Experiment 2

Pertaining to the first question, overall accuracy levels over the two conditions were significantly different among the three groups [$F(2, 36) = 5.84$, $P = 0.006$]. Overall performance of the WS group was significantly different from control adults but not from control children. This indicates that individuals with WS have very limited proficiency in processing unfamiliar melodies. Overall performance of adults with WS in our study resembled that of the younger group of control children who had an equivalent verbal mental age. It is still possible, however, that the worse performance of the WS group on this task is in part due to the fact that unfamiliar melodies were used which do not fit with standard Western musical structure. Since individuals with WS are more exposed to music (Levitin et al., 2005), it is possible that the use of such stimuli was more detrimental to them relative to the typically developing group.

As far as the second question is concerned, the groups also differed in their performance in the contour-preserving relative to the contour-violating condition, evident by the significant interaction of Condition and Group [$F(2, 36) = 3.7$, $P = 0.035$]. Individuals with WS performed equally in the contour-violating and contour-preserving condition [$t(1, 13) < 1$, $P = 0.965$], unlike control children who showed greater proficiency in the contour-violating condition [$t(1, 13) = 4.5$, $P = 0.001$]. Performance of the control adults resembled that of control children, but the difference between the conditions was less pronounced [$t(1, 13) = 1.8$, $P = 0.096$]. This is probably because both conditions were relatively easy for adult controls. The lack of sensitivity to contour cues in the WS group relative to the control groups did not differ in the 5-notes version of the task relative to the easier 4-note version. A 3-way interaction of Condition, Group, and Notes was not significant [$F(2, 36) < 1$, $P = 0.624$]. Moreover, a separate repeated measures ANOVA for the WS group revealed no significant interaction of Condition and Notes [$F(1, 12) < 1$, $P = 0.703$].

These findings extend the results of Deruelle et al. (2005) with children, demonstrating that the lack of sensitivity to contour cues is a developmentally stable phenomenon in WS that does not change even by adulthood. The results suggest that when processing unfamiliar melodies, individuals with WS do not take advantage of available cues to the same extent as normal controls. Adults with WS were able to detect changes in pitch which preserve the overall contour only in a manner similar to children with the same mental age. However, when additional cues were made available such as violation in the contour, normal controls showed greater proficiency, whereas individuals with WS failed to derive any such advantage.

General Discussion

The current investigation of auditory pattern perception in Williams Syndrome extends our understanding of the complex nature of auditory skills in this developmental condition. Certain aspects of auditory pattern perception are proficient in WS, while others fail to reach the levels of normal controls. Our findings reveal that when sorting auditory sequences into perceptual streams, individuals with WS are sensitive to the same pitch constraints as normal controls (Experiment 1). However, as the demands of auditory tasks become more complex, as in unfamiliar melody discrimination tasks, performance of individuals with WS begins to suffer. These differences cannot be simply explained on the basis of performance decrement in response to more difficult tasks since our study revealed different, atypical strategies for processing auditory input compared to controls in two conditions matched for overall difficulty. While individuals with WS are able to use absolute pitch cues, they exhibit a developmentally stable failure to use contour cues when discriminating melodies, even in adulthood (Experiment 2). Taken together, the two experiments indicate that absolute pitch cues are used proficiently in segregating streams where differences in pitch are constant over time. However, when the task involves tracing varying levels of pitch over time and relative pitch cues are available, the latter cues are not used effectively by individuals with WS.

While it might be appealing to describe these findings in the auditory domains in the usual terms of the juxtaposition of patterns of sparing and impairment in describing the phenotype of this clinical group, this would, in our view, be misleading. The goal of our study was not simply to describe task-specific patterns of proficiency or impairment in the auditory domain, but above all to speculate on how these domain-relevant developmental mechanisms of auditory pattern perception help to explain the gradual emergence of the adult phenotype in this developmental condition.

The notion that domain-relevant mechanisms such as those in the auditory domain help to explain the emerging behavioral phenotype in WS is not to conjecture direct or causal links between these two levels. In our view, these auditory mechanisms are important, particularly in early development, in altering the experience and the perception of input, providing a possible mechanism by which developmental outcomes are reached. What are the implications of these results in the auditory domain for understanding the relative proficiency of language in WS? We now examine a few possibilities, deriving predictions from theoretical accounts of how auditory pattern perception relates to both language and music. Different models make different predictions depending on the extent to which they propose that shared auditory mechanisms underlie the development and online processing of language and music.

An extreme theoretical view of modularity would predict that there is little relationship between auditory pattern perception and either language or music, since dedicated mechanisms are thought to underlie development and processing in each domain. According to this view, our findings reflect perceptual biases, which have no serious implications outside low-level perception. It could be further argued that the atypical processing biases found in the auditory domain bear no relation to language or music since similar biases are found in the visual domain and in the face processing domain, where individuals with WS are biased towards using local or featural cues over global or configural information. However, we reviewed in the introduction a substantial body of adult and developmental literature that such extreme views are not supported by experimental evidence.

Alternative theoretical models, while retaining a modular view of the organization of language and music, propose that these domains can be regarded as functionally modular, yet nevertheless sharing some components. In the model proposed by Peretz and Coltheart (2003), stream segregation (Experiment 1) would be considered an early component in processing, the output of which is subsequently passed onto dedicated mechanisms for language and music processing. The implication of our findings with the WS group is that such rudimentary analysis is performed in the typical fashion in this population and provides adequate input for language and music processing.

On the other hand, in this model, contour analysis (Experiment 2) is a mechanism primarily important for music. The prediction based on our findings is that WS individuals should have some deficits in musical processing in cases where relative contour information is required. Generally speaking, this implies that the organization of pitch patterns in music will predominantly rely on absolute pitch information in WS. These predictions are consistent

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with studies of music perception showing enhanced absolute pitch perception in WS (Lenhoff, 1998).

In relation to language, the model proposed by Peretz and Coltheart (2003) does not rule out an effect of contour on language but deems it to be weak and indirect. By contrast, other models adopting even less stringent views of modularity make slightly different predictions. Some claim that although linguistic and musical knowledge may be represented in different areas in the brain, they still share various processing resources, mainly those responsible for processing of structural relations unfolding over time (Patel et al., 1998; Patel, 2003). According to this model, pattern perception mechanisms like stream segregation (Experiment 1) and contour analysis (Experiment 2) are relevant for processing both language and music. This model predicts that in both language and music, syntactic relations which rely on local dependencies should be proficient in WS whereas those relying on long-distance dependencies, where the integration of relational information over time is required, would be affected. Although no study to date has directly examined these structures in either domain, partial support can be found in studies examining the neural correlates of atypical language processing. A study using event-related potential (ERP) examined the online processing of sentences ending either in a probable way or in an anomalous way e.g., my fingers are in the moon. Individuals with WS did not show the typical left hemisphere asymmetry for grammatical words which typically indexes the integration of the elements of the sentence nor did they show the typical processing difference between content words and grammatical words (Mills et al., 2000). These predictions are also consistent with more general patterns of linguistic processing in this population where, although individuals with WS appear to master quite a few formal rules of phonology and syntax, their representations are nevertheless fragile, and impairments are often masked by their superficial fluency (Grant, Valian, & Karmiloff-Smith, 2002; Thomas et al., 2001; Volterra, V., Capirci, O., Pezzini, G., & Sabbadini, 1996).

Developmental models of the relationship between language and music suggest consistent predictions to those derived from adult models like those described above. These models emphasize that while a modular organization may be plausible in adulthood, shared developmental mechanisms are very likely to come into play, including mechanisms of auditory pattern analysis (McMullen & Saffran, 2004; Newport & Aslin, 2000; Trehub, 2003; Trehub & Trainor, 1993).

As noted in the introduction, prosodic contours provide infants with rich cues marking syntactic boundaries since these contours correlate probabilistically with syntactic structures in language. If, as our findings suggest, infants with WS turn out to be less sensitive to contour information, this is likely to contribute to the significant delay in language acquisition in this population. It is also consistent with findings specifically related to infants' segmentation of the speech stream revealing serious delay in toddlers with WS relative to typical controls (Nazzi, Paterson, & Karmiloff-Smith, 2003). Perhaps in the clinical group a lack of sensitivity to these prosodic contour cues requires a more extended period of exposure to linguistic input for the discovery of structure to get off the ground.

Of course much remains to be understood not only with respect to auditory pattern perception in WS, but also in relation to additional domain-relevant mechanisms shown to be important precursors to language such as memory, joint attention, and social referencing, where we know that toddlers with WS already display early impairments (Laing et al., 2002; Vicari et al., 1996). Furthermore, none of the theoretical views discussed preclude the influence of domain-specific processes in the development and online processing of language and music. Despite commonalities between language and music, their various differences in communicative value or in hierarchical structure still pose inherently different problems for the brain. Hence, multiple and converging factors, both within and outside the domain in question, need to be invoked to understand how these factors exert an impact on the emerging phenotype, in an interactive rather than an additive fashion.

Such preliminary integrative accounts of how domain-relevant mechanisms relate to subsequent competence in higher-level domains are essential in explaining the resulting adult profiles. Elements of such preliminary developmental accounts for language or other domains can be examined rigorously and verified independently or in combination, as we have done in the current study, but must be integrated theoretically. This would shift our understanding of this developmental condition from a mere description of patterns of proficiency and impairment towards a coherent picture of the dynamics of how these patterns are achieved over developmental time.

References

Bellugi, U., Lichtenberger, L., Jones, W., Lai, Z., & St. George, M. (2000). The neurocognitive profile of Williams Syndrome: a complex pattern of strengths and weaknesses. *Journal of Cognitive Neuroscience, 12 supplement*, 7-29.

Bregman, A. (1990). *Auditory Scene Analysis.* Cambridge, MA: MIT Press.

Bregman, A., Ahad, P., Crum, P., & O'Reilly, J. (2000). Effects of time intervals and tone durations on auditory stream segregation. *Perception and Psychophysics, 62*, 626-636.

Deruelle, C., Schon, D., Rondan, C., & Mancini, J. (2005). Global and local music perception in children with Williams syndrome. *Neuroreport, 25,* 631-634.

Deutsch, D. (1999). Grouping mechanisms in music. In D. Deutsch (Ed.), *The psychology of music* (2nd ed., 349–411). San Diego: Academic Press.

Deutsch, D., Henthorn, T., & Dolson, M. (2004). Absolute Pitch, Speech, and Tone Language: Some Experiments and a Proposed Framework. *Music Perception, 21*, 339–356.

Don, A., Schellenberg, E. G., & Rourke, B. P. (1999). Music and language skills of children with Williams syndrome. *Child Neuropsychology, 5,* 154–170.

Dowling, W. J. (1978). Scale and contour: two components of a theory of memory for melodies. *Psychological Review, 85*, 341-354.

Dunn, L. M., Dunn, L. M., Whetton, C., & Pinitilie, D. (1997). *British Picture Vocabulary Scales.* Windsor, UK: NFER-Nelson.

Elman, J. L., Bates, E., Johnson, M. H., Karmiloff-Smith, A., Parisi, D., & Plunkett, K. (1996). *Rethinking Innateness: A Connectionist Perspective on Development.* Cambridge, MA: MIT Press.

Foxton, J., Dean, J., Gee, R., Peretz, I., & Griffiths, T. (2004). Characterization of deficits in pitch perception underlying 'tone deafness'. *Brain, 127,* 801-810.

Friedereci, A., Maess, B., Koelsch, S., & Gunter, T. C. (2001). Musical syntax is processed in the area of Broca: A MEG study. *Nature Neuroscience, 4*, 540-545.

Grant, J., Valian, V., & Karmiloff-Smith, A. (2002). A study of relative clauses in Williams syndrome. *Journal of Child Language, 29,* 403–416.Heiser, M., Iacoboni, M., Maeda, F., Marcus, J., & Mazziotta, J. C. (2003). The essential role of Broca's area in imitation. *European Journal of Neuroscience, 17,* 1123-1128.

Heiser, M., Iacoboni, M., Maeda, F., Marcus, J., & Mazziotta, J. C. (2003). The essential role of Broca's area in imitation. *European Journal of Neuroscience, 17,* 1123-1128.

Huron, D. (2003). Is music an evolutionary adaptation? In I. Peretz & R. J. Zatorre (Eds.), *The cognitive neuroscience of music* (pp. 57–75). Oxford: Oxford University Press.

Hopyan, T., Dennis, M., Weksberg, R., & Cytrynbaum, C. (2001). Music skills and the expressive interpretation of music in children with Williams–Beuren syndrome: Pitch, rhythm, melodic imagery, phrasing, and musical affect. *Child Neuropsychology, 7,* 42–53.

Jusczyk, P. (1997). *The discovery of spoken language.* Cambridge, MA: MIT Press.

Jusczyk, P. (1999). How infants begin to extract words from speech. *Trends in Cognitive Science, 3,* 323 -328.

Kan, I. P., & Thompson-Schill, S. L. (2003). Effect of name agreement on prefrontal activity during overt

and covert picture naming. *Cognitive, Affective & Behavioral Neuroscience, 4,* 43-57.

Karmiloff-Smith, A. (1998). Development itself is the key to understanding developmental disorders. *Trends in Cognitive Sciences, 2,* 389-398.

Karmiloff-Smith, A., Brown, J., Grice, S., & Paterson, S. (2003). Dethroning the myth: Cognitive dissociations and innate modularity in Williams Syndrome. *Developmental Neuropsychology, 23,* 227-242.

Lenhoff, H. (1998). Insights into the musical potential of cognitively impaired people diagnosed with Williams syndrome. *Music Therapy, 16*, 33-336.

Laing, E., Butterworth, G., Ansari, D., Gsödl, M., Longhi, E. Panagiotaki, G., Paterson, S., & Karmiloff-Smith, A. (2002). Atypical development of language and social communication in toddlers with Williams syndrome. *Developmental Science, 5*, 233-246.

Levitin, D. J., & Bellugi, U. (1998). Musical abilities in people with WS. *Music Perception, 15,* 357–389.

Levitin, D. J., Cole, K., Lincoln, A., & Bellugi, U. (2005). Aversion, awareness, and attraction: investigating claims of hyperacusis in the Williams syndrome phenotype. *Journal of Child Psychology & Psychiatry, 46,* 514-523.

Levitin, D. J., Cole, K., Chiles, M., Lai, Z., Lincoln, A., & Bellugi, U. (2004). Characterizing the musical phenotype in individuals with WS. *Child Neuropsychology, 10,* 223-247.

Levitin, D. J., Menon, V., Schmitt, J. E., Eliez, S., White C. D., Glover, G. H., Kadis, J., Korenberg, J. R., Bellugi, U., & Reiss, A. L. (2003). Neural correlates of auditory perception in Williams syndrome: an fMRI study. *Neuroimage, 8,* 74-82.

Maess, B., Koelsch, S., Gunter, T., & Friederici, A. D. (2001). ''Musical syntax'' is processed in the area of Broca: An MEG-study. *Nature Neuroscience, 4,* 540–545.

McMullen, E., & Saffran, J. (2004). Music and Language: A Developmental Comparison. *Music Perception, 21,* 289–311.

Mehler, J., Dupoux, E., & Segui, J., (1990). Constraining models of lexical access: The onset of word recognition. In: G. T. M. Altmann (Ed.), *Cognitive models of speech processing.* NJ: Lawrence Erlbaum.

Mehler, J., Jusczyk, P., Lambertz, G., Halsted, N., Bertoncini, J., & Amiel-Tison, C. (1988). A precursor of language acquisition in young infants. *Cognition, 29,* 143–178.

Mervis, C. B., & Bertrand, J. (1997). Developmental relations between cognition and language: Evidence from Williams Syndrome. In: L. Adamson & M. Romski (Eds.), *Research on Communication and Language Disorders: Contributions of to Theories of Language Development* (pp.75-106). NY: Brooks.

Mills, D. L., Alvarez, T. D., St. George, M., Appelbaum, L. G., Bellugi, U., & Neville H. (2000). III. Electrophysiological studies of face processing in Williams syndrome. *Journal of Cognitive Neuroscience, 12 supplement,* 47-64.

Nazzi, T., Paterson, S., & Karmiloff-Smith, A. (2003). Early word segmentation by infants and toddlers with Williams syndrome. *Infancy, 4*, 251 – 271.

Newport, E. L., & Aslin, R. N. (2000). Innately constrained learning: blending old and new approaches to language acquisition. In: S. C. Howell, S. A. Fish, & T. Keith-Lucas (Eds.), *Proceedings of the 24th Annual Boston University Conference on Language Development* (pp. 1-21). Somerville, MA: Cascadilla Press.

Patel, A., Gibson, E., Ratner, J., Besson, M., & Holcomb, P. (1998). Processing syntactic relations in language and music: An event-related potential study. *Journal of Cognitive Neuroscience, 10,* 717–733.

Patel, A. (2003). Language, music, and the brain. *Nature Neuroscience, 6*, 674-681.

Peretz, I., & Coltheart, M. (2003). Modularity of music processing. *Nature Neuroscience, 6,* 688-691.

Pinker, S. (1999). *Words and rules.* London: Weidenfeld & Nicolson.

Saffran, J. R. (2003). Statistical language learning: Mechanisms and constraints. *Current Directions in Psychological Science, 12*, 110–114.

Singer-Harris, N., Bellugi, U., Bates, E., Jones, W., & Rossen, M. (1997). Contrasting profiles of language development in children with Williams and Down syndromes. *Developmental Neuropsychology, 13*, 345-370.

Thomas, M., Grant, J., Barham, Z., Gsödl, M., Laing, E., Lakusta, L., Tyler, L. K., Grice, S., Paterson, S., & Karmiloff-Smith, A. (2001). Past tense formation in Williams syndrome. *Language and Cognitive Processes, 2*, 143-176.

Trehub, S.E. (2003). The developmental origins of music. *Nature Neuroscience, 6*, 669-673.

Trehub, S. E., & Trainor, L. J. (1993). Listening strategies in infancy: The roots of music and language development. In: S. McAdams & E. Bigand (Eds.), *Thinking in sound: The cognitive psychology of human audition* (pp. 278-327). Oxford: Oxford University Press.

Wallin, N. L., Merker, B., & Brown, S. (Eds.). (2000). *The origins of music, Cambridge,* MA: MIT Press.

Vicari, S., Brizzolara, D., Carlesimo, G. A., Pezzini, G., & Volterra, V. (1996). Memory abilities in children with Williams syndrome. *Cortex, 32,* 503-514.

Volterra, V., Capirci, O., Pezzini, G., & Sabbadini, L. (1996). Linguistic abilities in Italian children with Williams syndrome. *Cortex, 32,* 663-677.