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Social Functioning and Autonomic Nervous System Sensitivity Across Vocal and Musical Emotion in Williams Syndrome and Autism Spectrum Disorder

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Abstract

Both Williams syndrome (WS) and autism spectrum disorders (ASD) are associated with unusual auditory phenotypes with respect to processing vocal and musical stimuli, which may be shaped by the atypical social profiles that characterize the syndromes. Autonomic nervous system (ANS) reactivity to vocal and musical emotional stimuli was examined in 12 children with WS, 17 children with ASD, and 20 typically developing (TD) children, and related to their level of social functioning. The results of this small-scale study showed that after controlling for between-group differences in cognitive ability, all groups showed similar emotion identification performance across conditions. Additionally, in ASD, lower autonomic reactivity to human voice, and in TD, to musical emotion, was related to more normal social functioning. Compared to TD, both clinical groups showed increased arousal to vocalizations. A further result highlighted uniquely increased arousal to music in WS, contrasted with a decrease in arousal in ASD and TD. The ASD and WS groups exhibited arousal patterns suggestive of diminished habituation to the auditory stimuli. The results are discussed in the context of the clinical presentation of WS and ASD.

Keywords

auditory processing; autism spectrum disorder; autonomic nervous system; emotion; music; social behavior; vocalizations; Williams syndrome

INTRODUCTION

Individuals with Williams syndrome (WS) and autism spectrum disorder (ASD) typically experience difficulties in interpreting others' emotional nonverbal displays, which have been linked to abnormalities in social behavior (Braverman, Fein, Lucci, & Waterhouse, 1989; Järvinen-Pasley et al., 2010a). WS stems from a hemideletion of 25–28 genes on chromosome 7 (Ewart et al., 1993; Hillier et al., 2003), and results in increased social drive, low social anxiety, and bias toward social over nonsocial information (Järvinen, Korenberg, & Bellugi, 2013; Järvinen-Pasley et al., 2008a; Martens, Wilson, & Reutens, 2008; Riby & Hancock, 2008, 2009). ASD is a pervasive neurodevelopmental disorder associated with disordered language and communication, empathy, and social cognition, and avoidance of social interaction (APA, 2013; Lord, Cook, Leventhal, & Amaral, 2000). Significant heterogeneity in the clinical presentation of both WS (Little et al., 2013; Porter & Coltheart, 2005) and ASD (Cohen, Masyn, Mastergeorge, & Hessler, 2015) exist.

Whereas social impairment is pivotal to ASD diagnosis, evidence highlighting such deficits in WS has begun to emerge. Klein-Tasman, Mervis, Lord, and Phillips (2007) found that 50% of their sample of children with WS showed impaired social interaction behaviors. Studies utilizing the Social Responsiveness Scale (SRS; Constantino & Gruber, 2005) have discovered more pronounced social-cognitive (prevalence rate of ~80%) than pro-social deficits in participants with WS (Klein-Tasman, Li-Barber, & Magargee, 2011; Riby et al., 2014; van der Fluit, Gaffrey, & Klein-Tasman, 2012). van der Fluit et al. (2012) further demonstrated that the ability to interpret social dynamics was negatively associated with problems in real-life social reciprocity in WS.

How do the contrasting social tendencies and associated dysfunction of individuals with WS and ASD impact their processing of auditory emotion varying on social relevance, i.e., vocalizations and music? Investigations of emotional vocalization processing in ASD have yielded mixed results, ranging from global emotion recognition deficits (Boucher, Lewis, & Collis, 2000; Peppé, McCann, Gibbon, O'Hare, & Rutherford, 2007; Philip et al., 2010; Stewart, McAdam, Ota, Peppé, & Cleland, 2013) to specific difficulties limited to one emotion or to longer stimuli (Järvinen-Pasley, Peppé, King-Smith, & Heaton, 2008; Jones et al., 2011) to unimpaired performance relative to TD (Baker, Montgomery, & Abramson, 2010; O'Connor, 2007). While studies examining prosodic processing in WS have reported deficits (Catterall, Howard, Stojanovic, Szczerbinski, & Wells, 2006; Plesa-Skwerer, Faja, Schofield, Verbalis, & Tager-Flusberg, 2006), aberrant processing has been suggested to be limited to negatively valenced information (Järvinen-Pasley et al., 2010b; see also Plesa Skwerer et al., 2006).

Individuals with WS and ASD display interest in and unimpaired capacity to enjoy music (WS: Dykens, Rosney, Ly, & Sagun, 2005; Hopyan, Dennis, Weksberg, & Cytrynbaum, 2001; Levitin et al., 2004; ASD: Blackstock, 1978; Heaton, 2003). Evidence indicates consistent and remarkable enhancements of social communication and emotional responsivity resulting from music-based interventions in ASD (Whipple, 2004; Wigram & Gold, 2006), and improved social functioning as indexed by the SRS resulting from music relative nonmusic-based social skills training intervention techniques (LaGasse, 2014). The

typical deficits in facial emotion identification in ASD have largely not been replicated for music (Heaton, Pring, & Hermelin, 1999; Quintin, Bhatara, Poissant, Fombonne, & Levitin, 2011), which may reflect the fact that music is not primarily social in nature (cf. Heaton et al., 1999; Quintin et al., 2011). Studies examining musical emotion identification in WS have indicated specific deficits in recognizing sad and fearful affect (Järvinen et al., 2012).

Literature into autonomic nervous system (ANS) function in WS (Järvinen & Bellugi, 2013) and ASD (Cheshire, 2012) is increasingly suggesting linkages with psychosocial behavior (Appelhans & Luecken, 2006). The polyvagal theory (Porges, 2007) postulates that variability in human social–emotional capacities across both typical (Quintana, Guastella, Outhred, Hickie, & Kemp, 2012) and atypical populations (Porges et al., 2013) is linked to resting state heart rate variability (HRV), a biomarker reflecting capacity for social interactive and social–emotional behaviors (Quintana et al., 2012).

The only known study examining autonomic sensitivity to affective auditory stimuli (happy, fearful, and sad vocalizations and music) found uniquely increased HRV to vocalizations compared to music in WS relative to TD. This suggested that human vocalizations appeared more engaging than music for individuals with WS. Increased HRV to happy stimuli, indexing greater vagal involvement, was also reported for the WS group. This is consistent with their positive bias (Dodd & Porter, 2010), as positive emotional stimuli are specifically socially engaging and promote approach-related motivations (Porges, 2007). In one study with children with ASD, higher resting skin conductance level (SCL) and increased autonomic sensitivity to tones, as compared to TD was found (Chang et al., 2012). Greater behavioral auditory–sensory processing impairments were associated with increased sympathetic activation at rest and to auditory stimuli. Abnormally high sympathetic involvement may thus underpin the atypical auditory reactivity in ASD. Given the scarceness of evidence of autonomic basis of auditory emotion processing profiles associated with WS and ASD, it is important to examine ANS activity as a potential regulator of associations between emotion sensitivity and social functioning in WS and ASD.

The current study examined the contribution of emotion processing profiles to patterns of social dysfunction in children with WS and ASD. A relationship between processing nonlexical affective auditory information and social functioning (the SRS) in both WS and ASD was hypothesized. However, while this relationship was specifically predicted between emotional sensitivity to social (i.e., vocal) information and social function in WS, for ASD, sensitivity to musical emotion was hypothesized to impact social functioning more strongly. For WS, this prediction was founded upon previous evidence indicating a positive relationship between social functioning and ability to interpret social situations (van der Fluit et al., 2012), and a bias toward social stimuli (e.g., Järvinen-Pasley et al., 2008a). For ASD, the hypothesis was based on studies indicating a greater effect of music-based than nonmusic-based social skills training interventions upon social functioning (SRS) (LaGasse, 2014), as well as a natural bias toward music over voice stimuli (Blackstock, 1978), which may enhance music-related processing abilities.

METHODS

Participants

The sample comprised 12 children with WS (2 males; mean age = 11.4 years, range: 9.6–13.9 years), 17 children with ASD (13 males; mean age: 10.6 years, range: 7.5–13.7 years), and 20 typically developing (TD) control children (8 males; mean age: 10.7 years, range: 7.5–13.0 years). The participants did not differ in terms of CA ($p = .39$). The genetic diagnosis of WS was established using fluorescence in situ hybridization (FISH) probes for elastin (ELN) (Ewart et al., 1993). Participants with WS exhibited the medical and clinical features of the WS phenotype (Bellugi, Lichtenberger, Jones, Lai, & St. George, 2000). All participants were recruited as a part of a multi-site multidisciplinary program of research addressing hierarchical levels of neural and cognitive processing in children with different neurodevelopmental disorders. The inclusion criteria for the children with high functioning ASD were as follows: monolingual English speaking; idiopathic/nonsyndromic autism diagnosed by the Autism Diagnostic Observation Schedule (ADOS) (Lord, Rutter, DiLavore, & Risi, 1999) and/or the Autism Diagnostic Interview-Revised (ADI-R) (Lord, Rutter, & Le Couteur, 1994) criteria; ability to participate in cognitive testing; and PIQ portion of the Wechsler Intelligence Scale for Children (WISC-III) above 70. The TD children were screened for history of brain trauma, psychiatric concerns, and central nervous system disorders, and were required to be native English speakers. All participants were administered a threshold audiometry test calibrated to ANSI s.3.21 (2004) standards to confirm normal hearing. Written informed consent and assent were obtained from the participant and their caregivers prior to the study. Experimental protocols were approved by the Institutional Review Boards at the Salk Institute and the University of California, San Diego.

Materials

Cognitive Functioning.—Cognitive functioning was assessed using the WISC-III (Wechsler, 1991). The groups varied on verbal intelligence quotient (VIQ), performance IQ (PIQ), and full-scale IQ (FSIQ) (all p -values $< .001$), with both the TD and ASD groups scoring higher on VIQ, PIQ, and FSIQ than WS (all p -values $< .001$). The TD group scored higher as compared to the ASD group on VIQ ($p = .001$) and FSIQ ($p < .001$), while the groups did not differ on PIQ ($p = .07$). The FSIQ score was subsequently covaried in the emotion identification data analyses to account for the lack of MA-matched controls for participants with WS and ASD.

In psychophysiology analyses, emotion identification accuracy was controlled for in our modeling approach as autonomic activity was recorded passively, independent of the active responses, which may convolute the data. Between-group IQ differences were not controlled for as they have been suggested not to contribute to meaningful differences in autonomic responses to valence and social content in neurodevelopmental conditions (Cohen et al., 2015). For the psychophysiological portion of the study, the sample comprised 11 children with WS (two males; mean age = 11.63, $SD = 1.56$), 17 children with ASD, and 20 TD participants (samples as above), as it was necessary to exclude data from one child with WS from analyses due to excessive movement artifacts.

Experimental Measure.—The vocal condition comprised 24 segments of nonlinguistic vocal sounds (2–3 s/segment) taken from the “Montreal Affective Voices,” a standardized set of vocal expressions without confounding linguistic information (available at <http://vnl.psy.gla.ac.uk/info.php?file=mav>), eight segments for each emotion (happy/fearful/sad). The musical condition included 24 segments of novel, normed musical pieces, eight segments eliciting each of three possible emotions (fearful/happy/sad), which have been specifically composed by Marsha Bauman of Stanford University for studies of musical ability in WS (see Järvinen et al., 2012; Järvinen-Pasley et al., 2010c). More detailed description of the experimental stimuli has been provided elsewhere (Järvinen et al., 2012).

Psychophysiology Recordings and ANS Measures.—EDA and electrocardiogram (ECG) measures were recorded during the passive portion of the experiment using BioPac MP150 Psychophysiological Monitoring System (BioPac systems, Inc., Santa Barbara, CA) at a 1,000 Hz sampling rate. Raw ECG signal was filtered and used to calculate HR and inter-beat interval (IBI) measures, after classifying the *R* peaks of the heart beat cycles. Besides the mean HR and IBI, standard deviation of the inter-beat interval (sdIBI) was extracted to assess variability in heartbeats for each experimental condition, as suggested by Mendes (2009). Quantification of the mean IBI was used in conjunction with mean HR, it representing a more sensitive and direct measure of parasympathetic and sympathetic systems activity (Bernston, Cacioppo, & Quigley, 1995).

For all ANS measurements, we sampled 7 s subsequent to stimulus presentation and a 3 s prestimulus baseline on a trial-by-trial basis, to compute event-related change scores. This enabled weighted trial-specific percentage variations of autonomic activity to be calculated, thus minimizing the influence of large-scale tonic fluctuations, and assess small-scale ANS reactivity and sensitivity.

Measure of Social Functioning.—The SRS (Constantino & Gruber, 2005) is a 65-item parental/caregiver questionnaire, designed for children aged 4–18 years to screen for symptomatology associated with ASD. The items result in T-scores across the scales: social awareness, social cognition, social communication, social motivation, and autistic mannerisms, and a total score. T-scores below 60 indicate no clinically significant concerns in social functioning; T-scores of 60–75 indicate mild-to-moderate social dysfunction; and T-scores higher than 75 indicate severe social dysfunction.

Procedure

Experimental Measure.—To prevent autonomic habituation effects, the psychophysiological (i.e., passive) portion of the study was always administered first, followed by the active behavioral affect identification portion. The stimuli were presented on a desktop computer running Matlab (The MathWorks, Inc., Natick, MA), which delivered a digital pulse embedded in the recording at the onset of each stimulus. To measure physiological responses, following a fixation cross for 1,000 ms, each stimulus was presented for 5,000 ms, separated by an interstimulus interval (ISI) of 9,000 ms (blank screen) for autonomic activity to return to near baseline. The stimuli were presented in four blocks of 12 stimuli in random order out of 48 total stimuli. A blinking fixation cross

preceded each stimulus. The duration of the auditory clips, presented at the onset of a 5,000 ms blank screen, were slightly variable (vocalizations average = $1,353 \pm 642$ ms; music average = $1,355 \pm 539$ ms). Participants were told that they would hear short sounds that would either be a voice or music. For the passive task, participants were only instructed to listen to the sounds carefully while attending to a monitor displaying a fixation cross, and staying as quiet and still as possible. For the active task, participants were played the stimuli again sequentially, and asked to identify the emotion elicited by each sound at a forced-choice response screen. Prior to the onset of the active task, the experimenter showed the response screen to the participant, which listed the three possible emotions to ensure that the participant understood the response options (scary/scared, happy, sad). The participants responded verbally, and the experimenter operated the computer keyboard on the participant's behalf. Please see Järvinen et al. (2015) for a detailed description of the procedures involved in psychophysiological recording.

RESULTS

Behavioral Emotion Identification

The data are displayed in Table 1 (total number of trials per affect category = 8).

A repeated-measures analysis of covariance (ANCOVA), with the condition (vocal/musical) and emotion (fearful/happy/sad) entered as within-participants factors, group (WS/ASD/TD) as a between-participants factor, and FSIQ score as a covariate, failed to reveal significant effects (all p -values $> .065$). Expectedly, there was a significant main effect of FSIQ ($F(1,45) = 7.46, p = .009$).

ANS Measures and Statistical Analyses

Autonomic event-related change scores were analyzed by a linear mixed-effects models approach using *R* (R Development Core Team, 2008) and the *R* package *nlme* (Pinheiro, Bates, DebRoy, Sarkar, & R Development Team, 2013). The models accounted for random effects including between-participant individual differences and confounding covariates (gender, age), and overall behavioral affect identification accuracy. All trials containing outliers (> 2.5 SD above or below the mean) were excluded from analyses. Group (WS/ASD/TD), condition (vocal/musical), emotion (fearful/happy/sad), block (four levels), and trial number (12 levels) were included as fixed effects. To accurately investigate time-related changes while accounting for time-related confounds, the trial number was included as a discrete variable, and autocorrelations between subsequent trial measurements were modeled, which reflected a first-order autoregressive covariance matrix. Consistent with Pinheiro and Bates (2000), significance was assessed by conditional F -tests and report F and p -values of the Type III Sum of Squares computations. All pair-wise comparisons were Bonferroni corrected. The normality and homogeneity assumption for linear mixed-effects models was assessed by residuals.

ANS Analyses

The analyses of EDA data revealed a statistically significant interaction between group, emotion, and block ($F(12, 2037) = 2.09, p = .02$), and between groups, between condition

and trial number ($F(2, 2037) = 3.78, p = .02$). Pair-wise comparisons for blocks within each emotion category and group showed that children with ASD and TD demonstrated a gradually decreasing trend in mean EDA weighted changes to sad stimuli, while children with WS showed an inconsistent pattern of increasing and decreasing sympathetic activity. TD children consistently demonstrated negative modulations in EDA change scores, while WS and ASD groups tended to display different and less systematic patterns.

The mixed-effects models applied to sdIBI data highlighted significant main effects of group ($F(2, 42) = 3.23, p = .05$) and condition ($F(1, 2042) = 4.65, p = .03$). Children with WS showed the highest nonspecific event-related increases of variability of beat-to-beat interval, while TD children showed the lowest variability ($M_{WS} = 64.16, M_{ASD} = 53.43, M_{TD} = 40.36$, all pair-wise comparisons p -values $< .05$). Finally, vocal stimuli ($M = 52.97$) elicited greater changes of sdIBI than music ($M = 45.76$), regardless of their affective valence.

Focused analyses targeting the vocal condition revealed a two-way interaction between group and trial number linked to event-related sdIBI measures ($F(2, 994) = 3.79, p = .02$), suggesting group-specific differences in reactivity and habituation trends to vocalizations regardless of their emotional valence.

Focused analyses within the musical domain indicated a significant effect of group on EDA weighted change scores ($F(2, 42) = 6.47, p = .004$). Whereas both ASD and TD groups showed decreases in EDA, WS group exhibited positive EDA change scores, reflecting increased arousal to music ($M_{WS} = .12, M_{ASD} = -.20, M_{TD} = -.46$; all p -values $< .05$) (Fig. 1). Group by trial number interaction was significant ($F(2, 999) = 6.73, p = .001$), providing support to the between-group differences in reactivity and habituation to music.

SRS

The data are displayed in Table 2. A one-way ANOVA conducted for the data from WS and ASD groups indicated only one significant result for social motivation ($F(1, 27) = 9.22, p = .005$), with children with WS demonstrating less impaired behavior (all other p -values $> .10$). The data from TD participants were not included in these analyses due to each of them scoring within the normative range across all subtests.

Relations Between ANS Functioning and SRS

Pearson correlations (two-tailed) were applied between event-related EDA change scores and SRS data to elucidate associations between autonomic sensitivity to vocal and musical emotion and social functioning. For WS, all correlations failed to reach significance (all p -values $> .082$). For ASD, lower EDA response to sad vocalizations (and to vocalizations in general) was associated with more typical (i.e., lower) total ($r(17) = .57, p = .017$), social awareness ($r(17) = .59, p = .012$), and social motivation ($r(17) = .52, p = .034$) scores on the SRS. For TD children, more typical (i.e., lower) SRS total scores were associated with a lower EDA response to fearful vocal ($r(20) = .47, p = .039$) and sad musical ($r(20) = .51, p = .021$) stimuli. Further, more typical social cognition scores were associated with lower EDA response to fearful vocal ($r(20) = .53, p = .017$), fearful musical ($r(20) = .59, p = .006$), and sad musical ($r(20) = .49, p = .029$) stimuli, and more typical Social motivation scores were associated with lower EDA response to sad music ($r(20) = .46, p = .039$). Lower EDA

response to music in general was associated with more typical social functioning (SRS total score) ($r(20) = .50, p = .027$), in addition to the social cognition subscale ($r(20) = .57, p = .008$) for the TD children.

DISCUSSION

Current results failed to support our hypotheses, with no significant associations between autonomic sensitivity and social functioning for children with WS, which may reflect the lack of power resulting from the small sample size of this group. Alternatively, autonomic responsivity to emotion may not linearly correspond to social functioning across the social–motivational and social–communicative domains in WS, which may also reflect heterogeneity (Järvinen & Bellugi, 2013; Little et al., 2013). For children with ASD, in contrast to the hypothesis, lower autonomic reactivity to vocalizations was associated with more normal pro-social functioning (social awareness and social motivation). This contrasts with the pattern observed for TD children, for whom lower autonomic response to music was related to higher social functioning, and specifically, to social cognitive/communicative functions. The overall result that lower autonomic responses were linked to more typical social functioning in ASD (and TD) is consistent with previous evidence that more typical arousal patterns (i.e., not hypo- or hyperarousal) to social stimuli in such individuals are linked to less impaired social behavior (Mathersul, McDonald, & Rushby, 2013). The current results suggest that when arousal level is comfortable, individuals with ASD are more likely to be motivated to socially interact with others.

The differential pattern of associations for the ASD and TD groups may suggest that as ASD inherently is a disorder of social function, autonomic responsivity to social information is crucial. In TD, reactivity to music was associated with improved social functioning, and this may reflect relative cognitive demands/loads associated with the vocal versus musical stimuli, specifically, as the processing of musical as compared to vocal emotion appeared more challenging. It is important to emphasize that social functioning of the TD group was unimpaired, and thus the current results describe patterns within normative functioning in this group, unlike in ASD. Children with ASD demonstrated more robust responses to vocalizations than music, and responses to vocalizations were also stronger than those in TD. ASD is commonly associated with hyperarousal (e.g., Bal et al., 2010; Hirstein, Iversen, & Ramachandran, 2001), and the present findings indicating an association between lower autonomic function and more normal social reciprocity are consistent with earlier studies (e.g., Mathersul et al., 2013). It is interesting that lower autonomic arousal was specifically related to higher social motivation and awareness in ASD, and not to social cognitive/communicative functions, which may be due to pro-social skills developing in close connection with psychophysiological feedback from social experiences and situations.

No between-group differences in behavioral emotion identification were found for either condition when IQ differences between groups were controlled for. The extant literature on auditory emotion recognition is mixed in both WS and ASD. It is also possible that the present failure to detect such differences may reflect Type II error and/or a lack of power due to the smaller sample size of the WS group.

SRS data were broadly consistent with previous studies (Klein-Tasman et al., 2011; Riby et al., 2014; van der Fluit et al., 2012) indicating relatively preserved social motivation in children with WS combined with severe impairments in social cognitive domains (cf. Tager-Flusberg & Sullivan, 2000). WS and ASD groups were associated with similar general degree of social dysfunction. However, while ASD was associated with an even profile of deficit across the targeted domains, children with WS displayed less uniform pattern with relatively less affected pro-social than social–cognitive functions (cf. Klein-Tasman et al., 2011). This evidence thus contributes to the growing literature that highlights similarities between WS and ASD in social–emotional functions (e.g., Klein-Tasman et al., 2007, 2011).

Despite a lack of differences in emotion identification between groups, ANS evidence revealed interesting between-group differences, which may reflect specific features of the social profiles of the clinical groups. Cardiac measures showed that children with WS demonstrated the highest variation of all groups in heart periods (sdIBI) in response to vocalizations, suggesting reduced arousal since cardiac vagal reactivity is diminished for emotionally arousing stimuli, while the TD group displayed the opposite pattern. This result is consistent with our earlier counterpart study with adults with WS and TD (Järvinen et al., 2012). As the polyvagal theory postulates that vagal influence plays a crucial role in social processes (Porges, 2007), the extent between-group variability in response patterns may reflect their characteristic degree of social impairment.

For EDA measures, the groups demonstrated divergent patterns of autonomic responsivity across emotion categories: whereas TD children consistently exhibited negative modulations in EDA change scores, less systematic response patterns characterized those with WS and ASD, with the exception of the responsivity pattern of children with ASD for sad stimuli, which closely resembled that observed for TD. For happy and fearful stimuli, the WS group was the most reactive of all groups, while those with ASD showed slow and variable responses. Habituation for sad stimuli in both the ASD and TD groups was significant. As the EDA signal is thought to mirror sympathetic branch activity of the ANS, this is consistent with the general observation that sadness represents a low arousal emotion, specifically contrasting with high-arousal emotions of happiness and fear (Lang, Bradley, & Cuthbert, 1995). Children with WS demonstrated a different pattern, characterized by fluctuating increases and decreases in arousal to these stimuli over time, which may suggest unusual autonomic sensitivity and a nonfamiliarizing pattern to emotional auditory stimuli.

Second, general EDA-based results for vocalizations indicated that whereas both clinical groups demonstrated increasing levels and fluctuating patterns of arousal with a lack of habituation, TD children were characterized by lower and gradually diminishing arousal with increased exposure to stimuli (habituation). Children with WS and ASD showed stronger responses to vocalizations than TD participants, which likely index their atypical social behavior. Specifically for ASD, we additionally found that lower, i.e., more normal, autonomic reactivity to vocalizations was associated with more typical social functioning. This may be supported by altered neurobiology. The social impairment of ASD has been linked to atypical neural responsivity to complex speechlike information (Boddaert et al., 2003, 2004) and vocalizations (Gervais et al., 2004; Lloyd-Fox et al., 2013). Such individuals also demonstrate deficits in processing speech prosody evident behaviorally

(Järvinen-Pasley et al., 2008b; Peppé et al., 2007) and neurobiologically (Kujala, Lepistö, Nieminen-von Wendt, Näätänen, & Näätänen, 2005). A recent study examining perceptual auditory processing across vocalizations and music reported more abnormal behavioral and electrophysiological processing of speech than music in ASD (DePape, Hall, Tillmann, & Trainor, 2012), highlighting the primarily social nature of their impairment.

Third, TD children demonstrated decreasing levels of arousal with slow habituation over time for music, similar to vocalizations, while children with ASD showed irregular pattern of responsivity, with higher general arousal. By contrast, WS group demonstrated a unique nonlinear pattern characterized by an initial increase in arousal relative to the ASD and TD groups, reflecting decreased habituation. This is consistent with an earlier finding indicating a lack of habituation to facial expressions in mature individuals with WS (Järvinen et al., 2012). Taken together, the pattern of EDA-based results suggest that in TD, responses as well as overall arousal levels were similar across both experimental conditions, reflecting a general ANS response pattern that was independent of the degree of social relevance of the auditory stimuli.

Finally, increased arousal in children with WS to music as indexed by EDA was found, while the ASD and TD groups were associated with decreased arousal. This is interesting in light of previous evidence suggesting atypically strong music-evoked emotional reactions in WS (Dykens et al., 2005; Hopyan et al., 2001; Levitin et al., 2004). Rewarding aspects of music listening and experience of subjective pleasure (termed “chills”) have been linked to increased arousal (Salimpoor, Benovoy, Longo, Cooperstock, & Zatorre, 2009). While the results may reflect a similar phenomenon in the WS group, larger-scale studies are needed.

In conclusion, the present exploratory study identified both syndrome-specific and syndrome-general features in profiles of autonomic reactivity to vocal and musical affect and social functioning in WS and ASD. Further studies are needed to clarify the link between autonomic reactivity in individuals with WS and ASD and their social difficulties, as well as problem behaviors. It may also be valuable to consider the history of social skills intervention and/or music therapy of participants, which are relatively commonly administered in developmental disorders. Such studies would elucidate the interesting question of whether behavioral interventions may play a role in reorganizing psychobiological functions.

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FIGURE 1.
Group differences in electrodermal weighted responses to music (asterisks denote pairwise comparisons where $p < 0.05$).

Table 1.

Mean Correct Emotion Identification Performance for Fearful, Happy, and Sad Stimuli Across the Vocal and Musical Conditions for Children With WS, ASD, and TD

Stimulus Type	WS (<i>n</i> = 12)		ASD (<i>n</i> = 17)		TD (<i>n</i> = 20)	
	Mean	% Correct (SD)	Mean	% Correct (SD)	Mean	% Correct (SD)
Vocal fearful	84.38	(25.07)	91.18	(24.51)	98.75	(3.85)
Vocal happy	91.67	(17.94)	99.27	(3.03)	99.27	(2.80)
Vocal sad	85.42	(14.92)	93.38	(7.80)	96.25	(7.14)
Musical fearful	62.50	(38.06)	77.21	(27.68)	91.88	(10.94)
Musical happy	82.29	(27.93)	94.85	(9.94)	99.38	(2.80)
Musical sad	59.38	(29.74)	66.18	(27.87)	85.00	(14.96)

Table 2.

Mean SRS T-Scores Across Participant Groups

SRS Domain	WS (<i>n</i> = 12)		ASD (<i>n</i> = 17)		TD (<i>n</i> = 20)	
	Mean	T-Score (SD)	Mean	T-Score (SD)	Mean	T-Score (SD)
Social awareness	65.17	(11.1)	68.53	(98.)	50.75	(8.4)
Social cognition	81.25	(6.8)	74.71	(11.7)	47.20	(6.5)
Social communication	74.00	(13.2)	75.60	(9.6)	47.25	(7.1)
Social motivation	58.08	(12.4)	72.00	(12.0)	48.70	(8.4)
Autistic mannerisms	88.92	(15.2)	83.71	(13.6)	48.05	(7.0)
Total score	77.50	(10.8)	79.88	(9.4)	47.75	(7.2)

Note: Higher T-scores reflect greater deficits in the domain.