

Increased prevalence of unusual sensory behaviors in infants at risk for, and teens with, Autism

Spectrum Disorder

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Abstract

The current study investigated the prevalence and pattern of unusual sensory behaviors (USBs) in teens with Autism Spectrum Disorder (ASD) and infants (3 – 36 months) at risk for ASD. From two different sites (UCSD and UConn), caregivers of infants at high ($n=32$) and low risk ($n=33$) for ASD, and teenagers with ($n=12$) and without ASD ($n=11$), completed age-appropriate Sensory Profile questionnaires (Infant/Toddler Sensory Profile, Dunn 2002; Adolescent/Adult Sensory Profile, Brown & Dunn 2002). The results show that high-risk infants and teenagers with ASD exhibit higher-than-typical prevalence of USBs. Results of our distribution analyses investigating the *direction* of sensory atypicalities (greater-than-typical vs. less-than-typical) revealed a fair degree of consistency amongst teens, however, USB patterns were more varied in high-risk infants.

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Autism Spectrum Disorder (ASD) is characterized by the presence of social communication deficits as well as restricted and repetitive behaviors (DSM-5; American Psychiatric Association, 2013). It has long been known – since ASD was first described in the 1940s (Asperger 1944; Kanner 1943) – that unusual sensory behaviors (USBs; i.e., increased prevalence of sensory atypicalities, for example, covering ears or avoiding bright rooms) are also a common feature of the disorder, with estimates ranging from 45% to 95% of individuals with ASD demonstrating some level of USB () across various perceptual domains (visual, auditory, tactile, and oral) (Baker, Lane, Angley, & Young 2008; Baranek, David, Poe, Stone, & Watson 2006; Kay 2001; Kern 2007; Tomchek & Dunn 2007). In addition, it is now widely recognized that USBs can profoundly impact one’s daily functioning. For example, teens with ASD report decreased concentration in school as a result of their sensory difficulties (Howe & Stagg 2016), and increased anxiety as they attempt to navigate crowded school hallways (Humphrey & Lewis 2008). Likewise, caregivers report that sensory processing difficulties prevent children from engaging in activities and hinders their ability to explore novel environments (Schaaf, Toth-Cohen, Johnson, Outten, & Benevides 2011). In response to this increased awareness, the most recent DSM-5 has incorporated USBs as a criterion for ASD.

Based on the high prevalence of USBs and because of their negative impact on those affected, it is crucial that we develop a more comprehensive understanding of the emergence of USBs early in development, and the manifestation of these USBs as children with ASD reach teenage years. To address this, the current study investigated USBs in young infants who are at risk for developing ASD (younger siblings of children diagnosed with ASD; Ozonoff et al.

2011), with the notion that these infants carry some of the genes associated with ASD and therefore may exhibit USBs, which may elucidate early risk factors in ASD as well as the broader autism phenotype (i.e., atypicalities seen in first-degree relatives of individuals with ASD). The current study also investigated USBs in teens with ASD, to better understand not just the prevalence of USBs, but the nature of USBs, during this stage of development. Finally, by comparing USBs between infants at risk for ASD and teens with ASD we hope to potentially elucidate developmental changes in USBs.

Currently, there is no easy way for diagnostic assessments of ASD to test for USBs, since this would require observing the child within an unwieldy number of sensory contexts. As such, data regarding the prevalence of USBs comes predominantly from questionnaires filled out by individuals with ASD or their caregivers. Many studies, including ours, have employed the “Sensory Profile” (SP, Dunn 1999) questionnaire. The SP examines the frequency of USBs by characterizing two dimensions of sensory behaviors: (1) an individual’s neurological sensitivity, which refers to the sensitivity of relatively low-level sensory systems, and can be classified as “low” vs. “high”, and (2) reactivity, which refers to how an individual responds behaviorally to sensory stimuli, and can be classified as “passive” vs. “active”. Based on this model (Dunn 1997), sensory behaviors can be classified according to four categories of sensory responsiveness – Low Registration (i.e., low sensitivity and passive response), Sensation Seeking (i.e., low sensitivity and active response), Sensory Sensitivity (i.e., high sensitivity and passive response), and Sensation Avoiding (i.e., high sensitivity and active response) (see Table 1 for examples). Additionally, questions on the Sensory Profile are separated based on the perceptual domain in which these behaviors occur (e.g., Visual, Auditory, Tactile, Vestibular, Oral). Responses are

then examined based on their deviation from reported norms, leading to a specific “sensory profile” for each individual.

It is important to point out that in individuals with ASD, USBs can manifest as either occurring at higher or lower frequencies than those seen in typically developing individuals. For example, in response to a statement such as “My child is unaware of people coming in and going out of a room” (Infant/Toddler Sensory Profile (ITSP); Dunn 2002), one would expect this to be true some of the time in a typically developing child. If a child is *always* unaware, this is considered atypical. Likewise, if a child is *never* unaware, this is also considered atypical. Additionally, note that contradictory patterns of sensory responsiveness can co-occur in the same individual, across different sensory modalities (Dunn 1997). For example, an individual might be over-responsive to certain visual stimuli and under-responsive to certain auditory stimuli.

[Table 1]

In a large meta-analysis, Ben-Sasson and colleagues (2009) reviewed data from 14 studies that used sensory questionnaires to investigate USBs in individuals with ASD. Seventy-nine percent of these studies used the Sensory Profile of Dunn (1999) or a variation of it (the Infant/Toddler Sensory Profile (ITSP) for infants, Dunn 2002, or the Short Sensory Profile, for older children, McIntosh, Miller, Shyu, & Dunn 1999), with the remaining studies using other sensory questionnaires such as the Sensory Experience Questionnaire (Baranek, David, Poe, Stone, & Watson 2006) and Sensory Sensitivity Questionnaire (Talay-Ongan & Wood 2000). They classified sensory behaviors according to three categories: (1) under-responsivity, which refers to indifference or slow response to sensory input, (2) over-responsivity, which refers to exaggerated or prolonged reaction to sensory input, and (3) sensation seeking, which refers to intense desire or interest in sensory experiences. Results indicated that, compared to typically-

developing controls, individuals with ASD exhibited a greater prevalence of all three aspects of sensory processing examined – under-responsivity, over-responsivity, and sensation seeking, with the strongest difference seen for under-responsivity. With regard to *changes* in the prevalence of USBs with age, results have been mixed. The meta-analysis of Ben-Sasson et al. (2009) suggested a steady increase in the frequency of USBs in children with ASD until 9 years of age, and then subsequently decreasing. In a longitudinal study following children with ASD from 2 to 8 years of age, McCormick, Hepburn, Young, and Rogers (2015) employed the Short Sensory Profile (McIntosh, Miller, Shyu, & Dunn 1999) and reported that USBs neither increased nor decreased with age. Using the Sensory Profile (Dunn, 1999), Lidstone et al. (2014) reported a decrease in Sensory Seeking behaviors in children with ASD between 3 and 17 years of age.

Other studies have restricted their investigation of USBs in individuals with ASD to the teen years. In one study, De la Marche, Steyaert, and Noens (2012) obtained self-reported sensory behaviors in 80 teens with ASD using the Adolescent/Adult Sensory Profile (AASP; Brown & Dunn 2002). In comparison to controls, teens with ASD indicated atypically low frequencies of Sensation Seeking behaviors, and atypically high frequencies of Sensation Avoiding behaviors. Since atypicalities can manifest as either atypically low or high frequencies of behaviors, these results in teens suggest consistency in the *direction* of the atypicalities. The authors concluded that these combined (directional) effects suggest different ways in which teens with ASD seek to minimize sensory input. In another study of 25 teens, Stewart et al. (2015) corroborated the findings of atypically low frequencies of Sensation Seeking behaviors in ASD, however, unlike De la Marche et al. (2012), this study also reported atypically high frequencies of Low Registration behaviors. Finally, in a study of 14 teens with ASD, Howe and Stagg (2016)

reported that all participants exhibited atypical frequencies of behaviors in at least one quadrant on the AASP, with 86% of individuals indicating atypicalities in two or more quadrants. When examining the Howe and Stagg (2016) data more closely, some consistencies in the *direction* of the atypicalities can be seen. Specifically, the majority of atypical responses were atypically *high* frequencies of Low Registration, Sensory Sensitivity, and Sensation Avoiding behaviors, and atypically *low* frequencies of Sensation Seeking behaviors. In sum, despite some discrepancies in the exact nature and developmental course of USBs in ASD, there is consensus across studies that they occur early in childhood and continue into adolescence.

But what about USBs in the first few months of life? Since ASD is not diagnosed reliably before 24 months of age (Cox et al. 1999; Charman & Baird 2002; Lord 1995; Lord et al. 2006; Moore & Goodson 2003; Stone et al. 1999; although see Osterling, Dawson, & Munson 2002 for evidence indicating reliable diagnosis as early as 12 months), it is difficult to discover the symptoms of ASD in early infancy. One approach to this challenge has been to study early videos of children later diagnosed with ASD and/or rely on retrospective reports of early life from their parents. However, this approach is potentially limited by lack of experimental control and/or parental bias. A second approach involves examining infant siblings of children diagnosed with ASD. These siblings are referred to as “high-risk” infants, as they have a greater likelihood of developing ASD (~ 20%, Ozonoff et al. 2011) than that seen in the general population (~1.5%, Center for Disease Control and Prevention 2014). Using this prospective approach, high-risk infants are compared to “low-risk” controls (defined as infants without a family history of ASD) on a particular behavior early in development. These data can then be examined in two ways. Researchers may wait until ASD can reliably be diagnosed (between 24 and 36 months), and then examine differences between infants who developed ASD versus those

who did not develop ASD. Results using this approach indicate the presence of early social, communication, and language deficits in infants who later developed ASD (e.g., Jones, Gilga, Bedford, Charman, & Johnson 2014, Rogers 2009, and Zwaigenbaum et al. 2009). Alternatively, researchers may focus on differences that are seen between high-risk and low-risk infants, regardless of a potential future ASD diagnosis. This method provides information about the “broader autism phenotype”, i.e., behavioral markers of ASD that are seen in unaffected relatives of those with ASD. Using this method, results indicate that high-risk siblings who do not develop ASD nevertheless demonstrate sub-clinical levels of ASD symptomology as well as other delays in development (e.g., Charman et al. 2016; Messinger et al. 2013).

Most relevant to the current study, there have been multiple studies demonstrating the presence of USBs in high-risk infants using observational measures, parent interviews, or parent questionnaires. In a study that used a semi-structured interview, Sacrey et al. (2015) asked caregivers about developmental concerns across a variety of domains. Parents of high-risk infants who later developed ASD reported more sensory concerns than those of high-risk infants who did not develop ASD and low-risk infants. In another study that used an in-lab object exploration task, Ozonoff et al. (2008) reported that, on average, high-risk infants who went on to develop ASD exhibited more visual exploration of objects than high-risk infants who did not develop ASD, high-risk infants who developed a developmental delay, and low-risk control infants. Similarly, Kaur, Srinivasan, and Bhat (2015) reported that high-risk infants exhibited greater visual exploration of objects at 6 and 9 months and greater oral exploration of objects at 15 months compared to low-risk controls. Using both observation and parent-report in case studies of nine high-risk infants who later went on to develop ASD, Bryson et al. (2007) reported that all nine displayed unusual sensory or motor behaviors during their first three years of life.

There is also evidence that some of these behaviors constitute a broader autism phenotype of ASD, seen even in high-risk infants who do not develop ASD. Specifically, during administration of the Autism Observational Scale for Infants (Bryson, Zwaigenbaum, McDermott, Rombough, & Brian 2007) (at 18 months), Bryson et al. (2007) reported that both high-risk infants who did and did not go on to develop ASD put their hands to their ears more often than low-risk infants, suggestive of a type of auditory sensory avoidance in this sample.

To date, only two studies have compared USBs between high and low-risk infants by using the Infant/Toddler Sensory Profile (ITSP), which is the questionnaire used in the current study. Mulligan and White (2012) tested 13 high-risk infants between 11- and 13-months of age. As opposed to comparing their data with established norms on the ITSP, they compared their high-risk cohort to a low-risk cohort. This “local” control group of low-risk infants is preferable, since it can provide a better control for demographics such as geographical region and SES, which might also influence outcomes on the ITSP. In this study, high-risk infants (outcome diagnosis was unknown) were reported to exhibit atypically low frequencies of Sensation Seeking behaviors and atypical auditory processing (although the direction of this atypicality was not specified). In another study, Germani et al. (2014) compared 59 high-risk and 31 low-risk infants at 24 months, and found that high-risk infants who later developed ASD differed from both the high-risk group that did not develop ASD and the low-risk group. Specifically, high-risk infants who later developed ASD were reported to exhibit atypically high frequencies of both Low Registration behaviors and auditory behaviors. Additionally, there was a non-significant trend for group differences in Sensation Seeking, with the high-risk infants who developed ASD exhibiting atypically high frequencies of these behaviors.

One limitation of both of these previous studies using the ITSP is that they collected data at only a single age (roughly 1 year, and 2 years, respectively). To get a more comprehensive look at USBs in infancy, the first aim of the study was to investigate the frequency and direction (i.e., atypically high or atypically low frequency of sensory behaviors) of USBs in infants at high risk for ASD during the first three years of life by having parents complete the ITSP (Dunn 2002) at multiple time points. While it is unknown which, if any, of our sample of high-risk infants later developed ASD, we hoped to gain information about the presence of sensory behaviors in the broader autism phenotype. For comparison, the second aim of the study was to investigate USBs in teens with ASD using the AASP (Brown & Dunn 2002), asking whether the frequency and direction of USBs in high-risk infants differs from that seen in teens with ASD.

Method

Subjects. The sample consisted of 12 teens with ASD (3 females, 25.0%; M=15.5 years, SD=1.48) and 11 typically developing teens (TD; 5 females, 45.5%; M=16.8 years, SD= 1.91; Table 1). [Table 1] Teens with ASD were diagnosed with ASD (Autistic Disorder, Asperger's Syndrome, or PDD-NOS) by a licensed clinical psychologist or medical doctor not associated with this research based on DSM-IV-TR criteria (American Psychological Association 2004), and confirmed in the laboratory during their visit using the Autism Diagnostic Observation Schedule (ADOS; Lord et al. 2000) by a trained, research reliable clinician not associated with this research. These teens had no known specific neurological or genetic conditions (e.g., Fragile X syndrome, Rett Syndrome) and were recruited from community resources in San Diego, the San Diego Unified School District, and from past participants who had completed studies in the University of California, San Diego lab. Teen subjects and their parents provided written, informed consent.

Infant subjects were recruited from two testing sites (University of California, San Diego (UCSD) and University of Connecticut-Storrs (UConn)). Subjects consisted of 32 (6 female, 18.8%) high-risk infants and for comparison, 33 (13 female, 39.4%) low-risk infants between the ages of 3 and 36 months (Table 2). [Table 2] Infants were classified as high-risk if they had an older sibling with a diagnosis of ASD (Autistic Disorder, Asperger's Syndrome, or Pervasive Developmental Disorder Not Otherwise Specified, PDD-NOS), confirmed through the administration of the ADOS (Lord et al. 2000) and the Autism Diagnostic Interview-Revised (ADI-R; Lord et al. 1994) by a trained, research reliable clinician not associated with this research. Classification of high-risk infants was based on risk status, and outcome data regarding future diagnosis were not available. Infants were classified as low-risk if they had no immediate or extended family members with ASD. All low-risk infants also had an older sibling to better match the fact that high-risk infants, by definition, have an older sibling. Infants came in at multiple time points between 3 and 36 months of age as part of a broader longitudinal study examining differences between high- and low-risk infants. At each visit, they were given a cognitive assessment and parents were asked to fill out the ITSP (see below for exact time points). The study was approved by the Institutional Review Board at UCSD and UConn. Parents provided written, informed consent.

Cognitive Assessment. Cognitive assessments were performed to ensure that any differences observed between groups were not due to differences in cognitive ability. The cognitive performance of teens was assessed using the Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler 1999), which rendered a “verbal IQ”, “performance IQ”, and a “full scale IQ” (see Table 2). Two typically developing teens did not complete the WASI, but

were included in future analyses.¹ The cognitive ability of infants was based on the Early Learning Composite score of the Mullen Scales of Early Learning (MSEL; Mullen 1995), an overall measure of development (see Table 3). Trained experimenters/clinicians administered both measures. Although we conducted the MSEL each time an infant came in for the ITSP (see below for exact time points), because the number and months at which infants came in differed somewhat across infants and research sites, we decided to look at MSEL scores at months where we had the most data for both the high- and low-risk groups. To this end, we focused on data from 6, 8-9 and 14-15 months. All infants contributed data to at least one of these time points, with 22 high-risk (68.8%) and 18 low-risk (54.5%) infants contributing data to all three time points.

Sensory Profile Questionnaires. For *teens*, we used the Adolescent/Adult Sensory Profile (AASP; Brown & Dunn 2002), which is a self-report questionnaire of sensory processing in individuals aged 11 years and older. The internal consistency coefficients of the AASP range from 0.64-0.78 for the quadrant scores. During a one-time visit to the lab, teen subjects indicated how often they exhibit certain behaviors related to sensory experiences using a one through five scale, ranging from “almost never” (score of 1) to “almost always” (score of 5). Coding of the AASP is described below.

For *infants*, we used the Infant/Toddler Sensory Profile (ITSP; Dunn 2002), which is a parent-report questionnaire of sensory behaviors in children from birth to 36 months of age. Two different versions of the ITSP were used, dependent on if the child was between birth and 6 months of age, or between 7 and 36 months of age. Questions vary slightly between the two versions to reflect appropriate developmental activities and milestones. The reliability of the

¹ Our results showed very similar effects whether these two teens were included vs. excluded, and we therefore decided to include them. Including them was also a conservative decision since the results were somewhat more significant when the two subjects were excluded.

ITSP ranges from 0.74 to 0.86. Internal consistency coefficients range from 0.17-0.83 in the birth to 6 month age range, and from 0.42-0.86 in the 7-36 month age range. Parents filled out the ITSP during each visit to the lab. At UCSD, they were asked to come to the lab at 3, 6, 8, 10, 12, 14, 18, 24, and 36 months of age, whereas at UConn, parents were asked to come to the lab at 3, 6, 9, 12, 15, 18, and 24 months. Parents indicated how often their child exhibits certain behaviors related to sensory experiences using a one through five scale, ranging from “almost always” (score of 1) to “almost never” (score of 5). Note that the scoring of the ITSP is the opposite of the AASP scoring, with higher scores on the ITSP indicative of atypically low frequency of a behavior, and lower scores indicative of atypically high frequency of a behavior. Thus, for presentation purposes and to be consistent with the scoring of the AASP, we reverse scored the ITSP data, such that a higher score referred to atypically high frequency of behaviors and a lower score referred to atypically low frequency of behaviors. This reverse scoring system for the ITSP is used throughout the paper.

Coding of the Questionnaires. The AASP used in teens examines four different “quadrants” of sensory processing: Low Registration, Sensation Seeking, Sensory Sensitivity, and Sensation Avoiding. For the different “quadrants”, the AASP has been normed, such that each subject (for each quadrant) received one of five possible scores (“much less than others”, “less than others”, “typical”, “more than others”, and “much more than others”), which we assigned numerically as -2, -1, 0, 1, and 2. Scores of -2 and -1 were categorized as “atypically low frequencies”, scores of 1 and 2 were categorized as “atypically high frequencies”, and 0 was categorized as “typical”. As the AASP does not categorize responses according to individual “perceptual domains” (such as auditory, visual, tactile, etc.), the perceptual domain analysis was not performed.

Like the AASP, the ITSP examines four different “quadrants” of sensory processing: Low Registration, Sensation Seeking, Sensory Sensitivity, and Sensation Avoiding. Separately, responses are categorized into different “perceptual domains”: Auditory, Visual, Tactile, Vestibular, and Oral, which were used for the perceptual domain analyses. Like the AASP for teens, the ITSP has been normed (for both the “quadrants”, and the “perceptual domains”), such that scores of -2 and -1 were categorized as “atypically low frequencies”, scores of 1 and 2 were categorized as “atypically high frequencies”, and 0 was categorized as “typical”.

Categorization. Originally we aimed to collect data from all 65 infants at all time points (9 for UCSD and 7 for UConn) with analysis completed separately for each time point. This proved to be problematic, for three reasons. *First*, the two research sites (UCSD and UConn) had independently been collecting ITSP data before we decided to combine our samples. Unfortunately, the time points were not exactly the same between sites. *Second*, as in any longitudinal study in infants, getting consistent compliance of parents is a challenge, and therefore some time points were missing for infants. A *third* related point is that some infants started the ITSP study later than others, and so, for those infants, we were missing earlier time points. As a result, we had insufficient numbers of infants at each time point to conduct a systematic study of the effects of age on our measure. We therefore used an alternative method to characterize sensory atypicalities across age, which assessed whether an infant showed evidence of a sensory atypicality at any time.

Specifically, infants were considered as exhibiting a sensory atypicality if they were categorized as atypical at *any* time point, that is, if they had a non-0 score at any time point. This was done for each quadrant (in the quadrant analysis) and each perceptual domain (in the perceptual domain analysis). As such, each infant was categorized as either “typical” or

“atypical”. We then further classified infants who were atypical into either “atypically low” or “atypically high”, as follows. We averaged their atypical scores for each quadrant and perceptual domain (i.e., -2, -1, +1, +2), leading to a final score for each quadrant and perceptual domain ranging from -2 to 2. For an infant who exhibited a sensory atypicality, their final categorization was “atypically low” if their average was less than 0, or “atypically high” if their average was greater than 0. For 6 instances, the average turned out to be 0. To be conservative, we excluded the individual data point from these subjects in analyses that categorized subjects into one of three categories (atypically low, typical, and atypically high), and only in the quadrant or perceptual domain in which the 0 average occurred. However, these data points were included in analyses that categorized subjects into one of two categories (atypical and typical), see below.

Analyses. Our analyses addressed two questions. *First*, we asked whether the overall prevalence of sensory atypicalities differed between high- and low-risk infants, and between TD teens and teens with ASD. For this analysis, we used two-category data (typical vs. atypical). For teens, the analysis was performed using a Pearson’s Chi-Square analysis. As it was not expected that a high number of TD teens or low-risk infants would display sensory atypicalities, a Fisher’s Exact test correction was used to control for the low expected count. As all significant results were consistent between the two tests, only the Fisher’s Exact test results were reported. For infants, the analysis was performed using a Monte Carlo simulation, to address the fact that there were unequal number of time points between high- and low-risk infants (Table 2). *Second*, we asked whether the distribution of data (i.e., direction) across three categories (atypically low, typical, and atypically high) differed between high- and low-risk infants and between TD teens and teens with ASD. We addressed this using Chi-square and Fisher’s Exact test. We described each analysis, in turn, below.

Overall Prevalence of Atypicalities. To ask whether prevalence of sensory atypicalities differed between high- and low-risk infants, we not only compared our data between groups but also further analyzed the data using a Monte Carlo simulation. This is because there was an unequal number of subject time points between high- and low-risk infants, with more time points for the high-risk infants than low-risk infants (Table 3). This difference meant that there were more opportunities for high-risk infants to be categorized as “atypical”, as any one time point with an atypical score would categorize the infant as atypical for that quadrant or perceptual domain, which could skew the results towards showing a greater prevalence of sensory atypicalities in high-risk infants.

We addressed this problem by employing a Monte Carlo simulation, which asked whether the observed difference between groups in prevalence of sensory atypicalities was greater than that which would be predicted by chance. As a first step, each subject, at each time point, was assigned one of two values: “typical” (assigned a “0”) or “atypical” (assigned a “1”), with the latter disregarding whether the atypicality was “atypically low” versus “atypically high”, as described above. This yielded a table of 0s and 1s for time points and subjects, separately for high- and low-risk infants. We then calculated the prevalence of “atypical” infants, separately for the high- and low-risk groups. We next computed the difference in prevalence between subject groups (i.e., high-risk prevalence – low-risk prevalence). We refer to this as the “observed” group difference in prevalence of atypicalities.

Then, the Monte Carlo simulation was performed by taking all the 0s and 1s in the table and randomly reassigning them, across the time points, subjects, and across group assignment. This simulation was repeated 10,000 times. For each simulation, we calculated the difference in prevalence between subject groups, leading to a distribution of differences that occur by chance.

As a final step, we determined if the “observed” group difference was a value greater than 95% of the simulated values from the Monte Carlo. If so, this provides evidence of a significant difference in prevalence of sensory atypicalities between groups.

Distribution of Data Across Three Categories. In this analysis, we asked whether the *distribution* of subjects across the three categories (i.e., atypically low, typical, or atypically high) differed between groups (between TD teens and teens with ASD, and between high- and low-risk infants). This analysis provided information over and beyond that provided by the “overall prevalence” analysis that divided subjects into “typical” vs. “atypical” (see above). Specifically, the distribution analysis allowed us to determine the *direction* of the atypicalities, i.e., whether the majority of atypicalities were low versus high frequencies of behaviors. The analysis was performed for each quadrant (both teens and infants) and perceptual domain (infants only), using Pearson’s Chi-Square analysis. As it was not expected that a high number of TD teens or low-risk infants would display atypicalities, a Fisher’s Exact test correction was used to control for the low expected count. As all significant results were consistent between the two tests, only the Fisher’s Exact test results were reported.²

Results

Cognitive Assessment. For *teens*, there were no significant differences on the WASI (Table 2) between TD teens and teens with ASD. For *infants*, there was no significant difference between groups based on the Early Learning Composite (ELC) score at all three time points tested (Table 3).

² Note that we could not perform a Monte Carlo simulation for this distribution analysis, since there was no obvious single “observed” group difference value to test in the model (as there was when we computed group difference in “prevalence of atypicalities”, above). We do not, however, expect the unequal time points between high- and low-risk infants to affect the three-category analysis.

Overall Prevalence of Atypicalities. For *teens*, the results of a Fisher's Exact test indicated differences between TD teens and teens with ASD in some (but not all) quadrants (Figure 1). Specifically, the prevalence of atypicalities was greater in ASD teens for Sensation Seeking (66.6% vs. 18.2%, $p = .026$), and marginally significantly greater for Sensation Avoiding (58.3% vs. 18.2%, $p = .060$). As the AASP does not categorize responses according to perceptual domain, the perceptual analysis was not performed.

[Figure 1]

For *infants*, the results of the Monte Carlo simulation indicated that the "observed" group difference in prevalence of atypicalities was substantially (and significantly) higher than the "simulated" group difference in prevalence of atypicalities, for some (but not all) quadrants and some perceptual domains (Figures 2 & 3). In the *Low Registration* quadrant, the observed difference in prevalence was 35.7% (high-risk = 78.1%, low-risk = 42.4%), which was greater than 97.3% of the (10,000) "simulated" values, translating to a significant p value of 0.027. In the *Tactile* domain, the observed difference in prevalence was 27.0% (high-risk = 90.6%, low-risk = 63.6%), which was greater than 95.5% of the (10,000) "simulated" values, translating to a significant p value of 0.04. In the *Vestibular* domain, the observed difference in prevalence was 51.4% (high-risk = 93.8%, low-risk = 42.4%), which was greater than 99.7% of the (10,000) "simulated" values, translating to a significant p value of 0.003. In sum, a significantly increased prevalence of atypicalities was observed for high-risk infants in the Low Registration quadrant, and the Tactile and Vestibular domains.

[Figure 2]

[Figure 3]

Distribution of Data Across Three Categories. For this analysis, we asked whether the *distribution* of categories (atypically low, typical, and atypically high) differed between groups. This analysis additionally allowed us to investigate whether the direction of the atypicalities were more skewed towards atypically low, or atypically high, frequencies of sensory behaviors. For *teens*, there were group differences in this distribution analysis in three of the four quadrants, two of which also showed increased overall prevalence of atypicalities (see above) (Figure 1). Specifically, we observed a group difference in the Sensation Seeking quadrant ($p = .033$), which appeared to be driven by a high percentage of teens with ASD (58.3%) indicating atypically *low* frequencies of sensory behaviors, which was not seen in typically developing teens (9.10%). Further examination showed that all but one teen with ASD who endorsed an atypicality endorsed atypically low frequency of behavior, meaning that the direction of atypicality was quite homogeneous across subjects. For the Sensation Avoiding quadrant, we observed a group difference (which was marginally significant, $p = .084$), which appeared to be driven by a high percentage of teens with ASD (50.0%) indicating atypically *high* frequencies of sensory behaviors, which was not seen in typically developing teens (9.10%). Further examination showed that all but one teen with ASD who endorsed an atypicality endorsed atypically high frequency of behavior, meaning that the direction of atypicality was quite homogeneous across subjects with ASD.

Although Low Registration did not reveal group differences in the overall prevalence of atypicalities (above), in this distribution analysis, we observed a group difference ($p = .024$), which appeared to be driven by a high percentage of teens with ASD (58.3%) endorsing atypically *high* frequencies of sensory behaviors, which was not seen in typically developing teens (9.10%). Further examination showed that *all* teens with ASD who endorsed an atypicality

endorsed atypically high frequency of behavior, meaning that the direction of atypicality was quite homogeneous across subjects with ASD.

For *infants*, we observed significant group differences in the distribution analysis in five of the nine categories (four quadrants and five perceptual domains) (Figures 2 & 3). Three of these significant group differences in distribution were also revealed as group differences in the “overall prevalence of atypicalities” analysis, above. Specifically, first, we observed a group difference in the Low Registration quadrant ($p = .013$), which appeared to be driven by a low percentage of high-risk infants (21.9%) exhibiting typical frequencies of sensory behaviors, with roughly equal percentages of atypically high and atypically low sensory behaviors, while the low-risk group showed a high percentage of infants exhibiting typical frequencies (57.6%). Second, we observed a group difference in the Tactile domain ($p < .0001$), which appeared to be driven by a high percentage of high-risk infants (81.3%) exhibiting atypically *high* frequencies of sensory behaviors, which was not seen in low-risk infants (21.2%). Third, we observed a group difference in the Vestibular domain ($p = .0001$), which appeared to be driven by a high percentage of high-risk infants (73.3%) exhibiting atypically *low* frequencies of sensory behaviors, which was not seen in low-risk infants (30.3%).

We also observed group differences in the distribution analysis in conditions that did not yield group differences in “overall prevalence of atypicalities”, above. Specifically, for the Sensation Avoiding quadrant and the Auditory domain, we observed a group difference ($p = .012$ & $p = .004$, respectively), which in both cases, appeared to be driven by a high percentage of high-risk infants (35.5% & 59.4%, for Sensation Avoiding and Auditory, respectively) exhibiting atypically *high* frequencies of sensory behaviors, which was not seen in low-risk infants (6.10% & 21.2%, for Sensation Avoiding and Auditory, respectively).

Further examination showed that for the Auditory, Tactile, and Vestibular perceptual domains, the direction of the atypicality (atypically low or atypically high) was quite homogenous across high-risk subjects. Specifically, for both the Auditory and Tactile domains, all but three high-risk infants who exhibited an atypicality exhibited atypically high frequency of behavior. For the Vestibular domain, all but five high-risk infants who exhibited an atypicality exhibited atypically low frequency of behavior. Thus, the direction of the atypicality was quite homogenous in the perceptual domains. In contrast, the Low Registration and Sensation Avoiding quadrants did not show homogeneity in the direction of the atypicality, with similar percentages of high-risk infants exhibiting atypically low and atypically high frequency of behavior.

Discussion

The purpose of this study was to examine USBs in teens with ASD and infants at high risk for developing ASD, with USB defined as atypical (with low or high) frequencies of sensory behaviors. In line with previous studies (De la Marche et al. 2012; Howe and Stagg 2016; Stewart et al. 2015), our results in teens with ASD indicated significantly increased overall prevalence of USBs in the Sensation Seeking quadrant (and marginally significantly increased for Sensation Avoiding). Interestingly, although our sample size was relatively small ($n = 12$), there was a large degree of homogeneity in the direction of the atypicality across subjects (as seen in previous studies, see *Introduction*). Specifically, for Low Registration, all teens who showed a USB endorsed atypically *high* frequency of behavior, for example, “not noticing when people enter the room or when their name was being called”. Likewise, for Sensation Seeking, all but one teen who showed a USB endorsed atypically *low* frequency of behavior, for example, “attending events with loud music or engaging in physical activity”. For Sensation Avoiding, all

but one teen who showed a USB endorsed atypically *high* frequency of behavior, for example, “staying away from crowds or moving away when others get close”. In line with the data from De la Marche, et al., (2012), the combined effects we observed for the Sensation Seeking and Sensation Avoiding quadrants suggest different ways in which teens with ASD seek to minimize sensory input.³

Patterns of USBs in high-risk infants. While results from previous studies suggest that high-risk infants may have USBs, to date there are very few direct tests. One way to examine this question is to use the ITSP. Prior to the current study, there were two studies that used the ITSP in high-risk infants, but only over limited age ranges (11-13 months, Mulligan and White 2012; 24 months, Germani et al. 2014). The current study sought to improve upon past research by examining USBs at multiple time points across the first three years of life. In line with extant research in this area involving both parent-report and observational measures (see *Introduction*), we found increased prevalence of USBs in high-risk infants, specifically, in the Low Registration quadrant and the Tactile and Vestibular domains. Additional differences were found in the *distribution* of USBs (i.e., atypically low, typical, and atypically high) in the Sensation Avoiding quadrant and the Auditory domain.

There are some notable consistencies and inconsistencies across the now three studies (including our own) that have used the ITSP in high-risk infants. While Mulligan and White (2012) found that 72% of high-risk infants (11 to 13 months) exhibited USBs in the Sensation

³ As we mention in the *Introduction*, it is interesting and important to point out that, due to the nature of the quadrant analysis combining data across all perceptual domains, it is possible to get seemingly contradictory results across the different quadrants, for example, atypically high frequencies of *both* Sensation Avoiding and Sensation Seeking. If this had occurred in the data (although it did not), it would presumably be driven by a different pattern of behaviors occurring in the different perceptual domains. For instance, an individual (or group of subjects) could exhibit high frequencies of Sensation Avoiding behaviors in the auditory domain and high frequencies of Sensation Seeking behaviors in the visual domain (Dunn, 1997).

Seeking quadrant, and that overall, high-risk infants displayed atypically low frequencies of Sensation Seeking behaviors, our results indicated no difference between high-risk and low-risk infants in this quadrant. Mulligan and White (2012) explain their finding as potentially related to a lower capacity or motivation for infants to explore their environment. As we examined USBs in a broader age range of infants (3 to 36 months), our sample may have included children with more developed skills that allowed for increased capacity and motivation to explore, leading to a lack of observable differences in the Sensation Seeking quadrant. In contrast to this inconsistency, our data are quite compatible with that of Mulligan and White (2012) and Germani et al. (2014) in finding differences in the Auditory domain (and see Bryson et al. 2007). However, we additionally observed increased prevalence of USBs in the Tactile and Vestibular domains. Differences across studies in the domains in which USBs are present may be due to the different ages included in the analyses across studies. Future research that can sample ages more discreetly will be required to test this possibility directly.

Similar to data from teens with ASD, our data from high-risk infants demonstrate relative homogeneity in some of the quadrants/perceptual domains. Specifically, for the Auditory and Tactile domains, all but 3 infants who showed a USB exhibited atypically high frequency of behavior. For the Vestibular domain, all but 5 infants who showed a USB exhibited atypically low frequency of behavior. In contrast, no homogeneity was observed in the direction of atypicalities in the Low Registration and Sensation Avoiding quadrants.⁴ Note that although the two previous studies that used the ITSP (Germani et al. (2014) and Mulligan and White (2012)) also reported a skewedness in the direction of atypicality (i.e., high-risk infants demonstrated atypically high frequency of sensory behaviors in the Low Registration and Auditory domains

⁴ Being that the perceptual domains are comprised of questions drawn from the various quadrants, it is difficult to speak to the exact pattern of behaviors that were seen in the perceptual domains for our high-risk sample.

and atypically low frequency of Sensory Seeking behaviors), they did not examine if the distribution of USBs differed from what was seen in low-risk infants. Additionally, as they did not report the percentage of infants who indicated atypically low, typical, and atypically high frequencies of USBs for all quadrants and perceptual domains, it is difficult to draw conclusions regarding the potential homogeneity present in their samples.

It is important to note that it is unknown which, if any, of the infants from our sample will develop ASD. As such, we cannot conclude if USBs are predictive of a later ASD diagnosis. However, due to the large effect size seen in our data, we do not think that the presence of USBs in high-risk infants was being driven solely by the subset of infants who may later develop ASD. Instead, we believe that our data provide evidence for the presence of USBs in the broader autism phenotype. Future studies should obtain outcome data to differentiate between those who do and do not develop ASD, as it is possible that those who develop ASD show an even higher prevalence of USBs which may shed insight on early risk factors for developing ASD.

Comparing USBs across development. Given that the broader autism phenotype (seen in high-risk infants) can be compared to individuals with ASD, a comparison between the high-risk infants and the teens with ASD in the current study might provide insight into the development of USBs in ASD. First, we should note that while the questions obviously differ between the ITSP (for infants) and the AASP (for teens/adults), a comparison between the two age groups is nonetheless reasonable, since the Sensory Profile was created to tap into the same four quadrants (Low Registration, Sensation Avoiding, Sensory Sensitivity, Sensation Seeking) regardless of age. In our comparison, we found that both teens with ASD and high-risk infants demonstrated atypical distribution of USBs (in comparison to their respective control groups) in Low Registration and Sensation Avoiding (trending in teens) quadrants, yet not in the other two

quadrants (Sensation Seeking and Sensory Sensitivity). As such, these data suggest that the specific areas of sensory processing difficulties are somewhat consistent from infancy to adolescence. An interesting difference between infancy and teens is that, while the *direction* of atypicality was fairly homogeneous in teens for the quadrant analyses, the same was not observed for infants in the quadrant analyses. This suggests that some infants shift the direction of their atypicality as they get older. For example, in Sensation Avoiding, an infant might start out atypically high, e.g., frequently trying to escape from noisy environments, but later switch to atypically low, e.g., rarely trying to escape a noisy environment. However, we acknowledge that a comparison between teens with ASD and high-risk infants is limited by the fact that the high-risk infants do not have a diagnosis of ASD. Future studies that track individuals longitudinally will be the required to examine this possibility. (NOTE: As the AASP does not separately examine the different perceptual domains, it is not possible to compare infants and teens along this dimension).

Limitations. As with most questionnaires, the AASP and ITSP are subjective in nature. Perhaps more importantly, parents' reports on the ITSP may be biased based on their previous experience. Specifically, for a parent of a child with ASD who exhibits USBs, this bias could go in one of two (opposite) directions. On the one hand, due to their awareness of USBs, the parent may be more inclined to notice the presence of a USB in the younger (at risk) sibling. On the other hand, if the parent is making a judgment *relative* to the older child with a USB, they may underestimate the degree to which the younger (at risk) sibling exhibits USBs. Given that the current study found an increased prevalence of USBs in high-risk infants, we cannot rule out the possibility that greater awareness of USBs in parents of high-risk infants resulted in an inflation of the reporting of atypicalities in their infants. However, if this were the case, we would expect

to find increased prevalence of USBs in high-risk infants in all dimensions, which we did not find. In the future, it would be interesting to examine whether USBs are reported to be more frequent in high-risk infants who do, versus do not, have an older sibling who exhibits USBs, which might speak to the potential for parents' previous experiences to bias their reports. Since we did not collect ITSP data from the older sibling, the current study cannot address this possibility.

Another limitation of the current study is the small sample size of our teen group, which makes it a bit difficult to generalize to teens with ASD more broadly. However, even with our small numbers, we found increased prevalence of USBs in teens with ASD, in line with previous studies. A final limitation of the current study is that we combined each infant's data across a large age range (3 to 36 months), asking whether they showed an atypicality at *any* time point (because we had inconsistent time points amongst our high- and low-risk infants preventing a systematic investigation of sensory atypicalities at any one time point). As such, we cannot know when USBs start and/or possibly end during early development. To address this limitation future studies should analyze data at multiple ages throughout development.

Clinical implications. The current findings suggest that we ought to screen for the presence of USBs in high-risk infants as early as possible, starting treatment regardless of whether or not the child develops ASD. Additionally, as our findings indicate that sensory atypicalities of high-risk infants vary across infants, it will be important to customize treatment plans based on the needs of individual children with the help of pediatric occupational, physical, and speech therapists. Parents and caregivers should be made aware of accommodations that could be implemented in the home, as well as taught gradual desensitization strategies to use with their child for the affected perceptual domains (tactile, vestibular, auditory, etc.). For

example, specific types of sensory environments can be incorporated into children's everyday play activities to facilitate habituation to sensory stimuli and/or create appropriate sensory experiences (e.g., Schaaf & Case-Smith, 2014) with future studies further evaluating the therapeutic effects of such early intervention approaches.

Ethical approval: "All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards."

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Figure Captions

Figure 1. The distribution of USBs in teens, by group, for each quadrant.

Figure 2. The distribution of USBs in infants, by group, for each quadrant.

Figure 3. The distribution of USBs in infants, by group, for each perceptual domain.

Figure 1 top

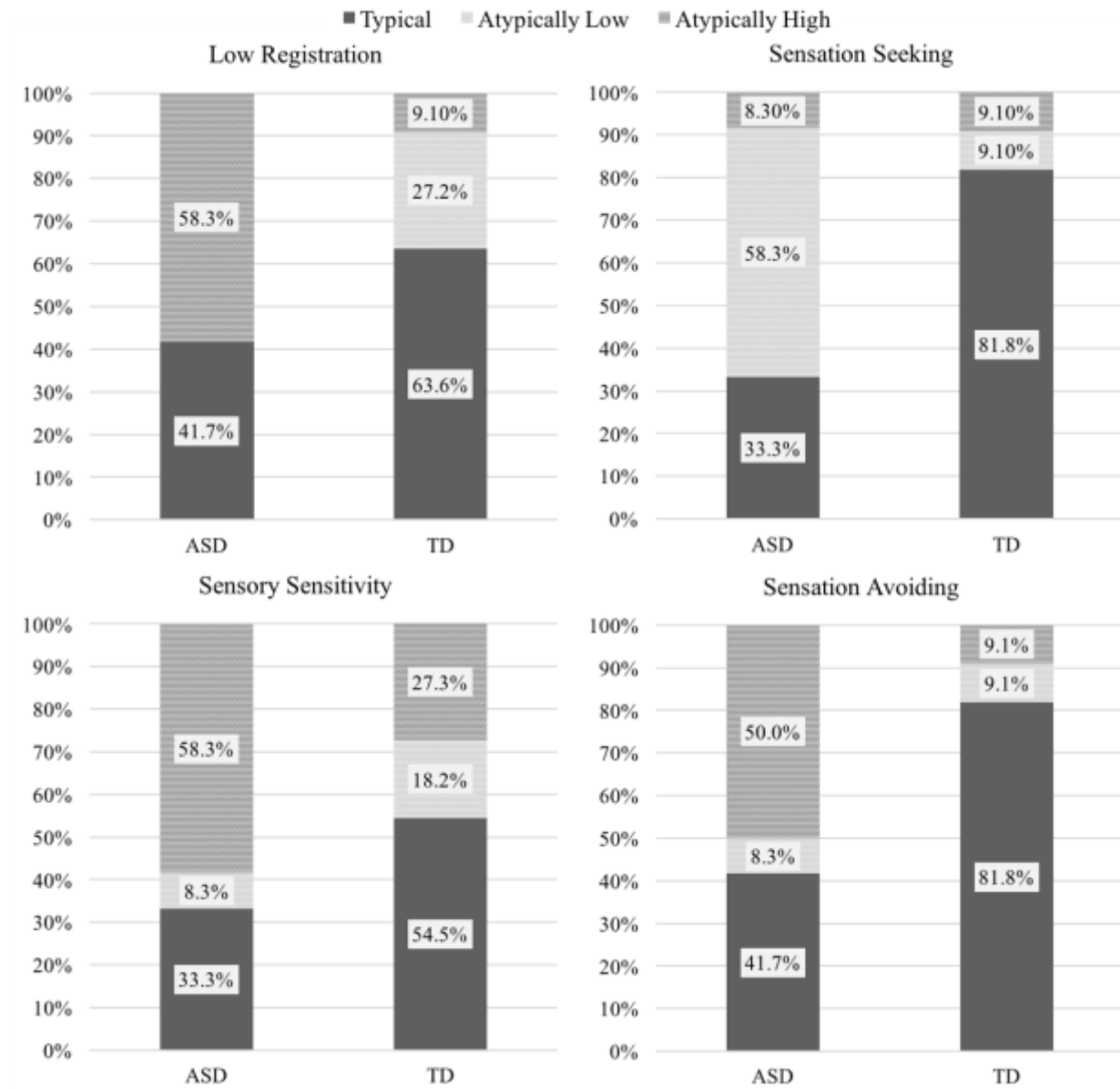


Figure 2 top

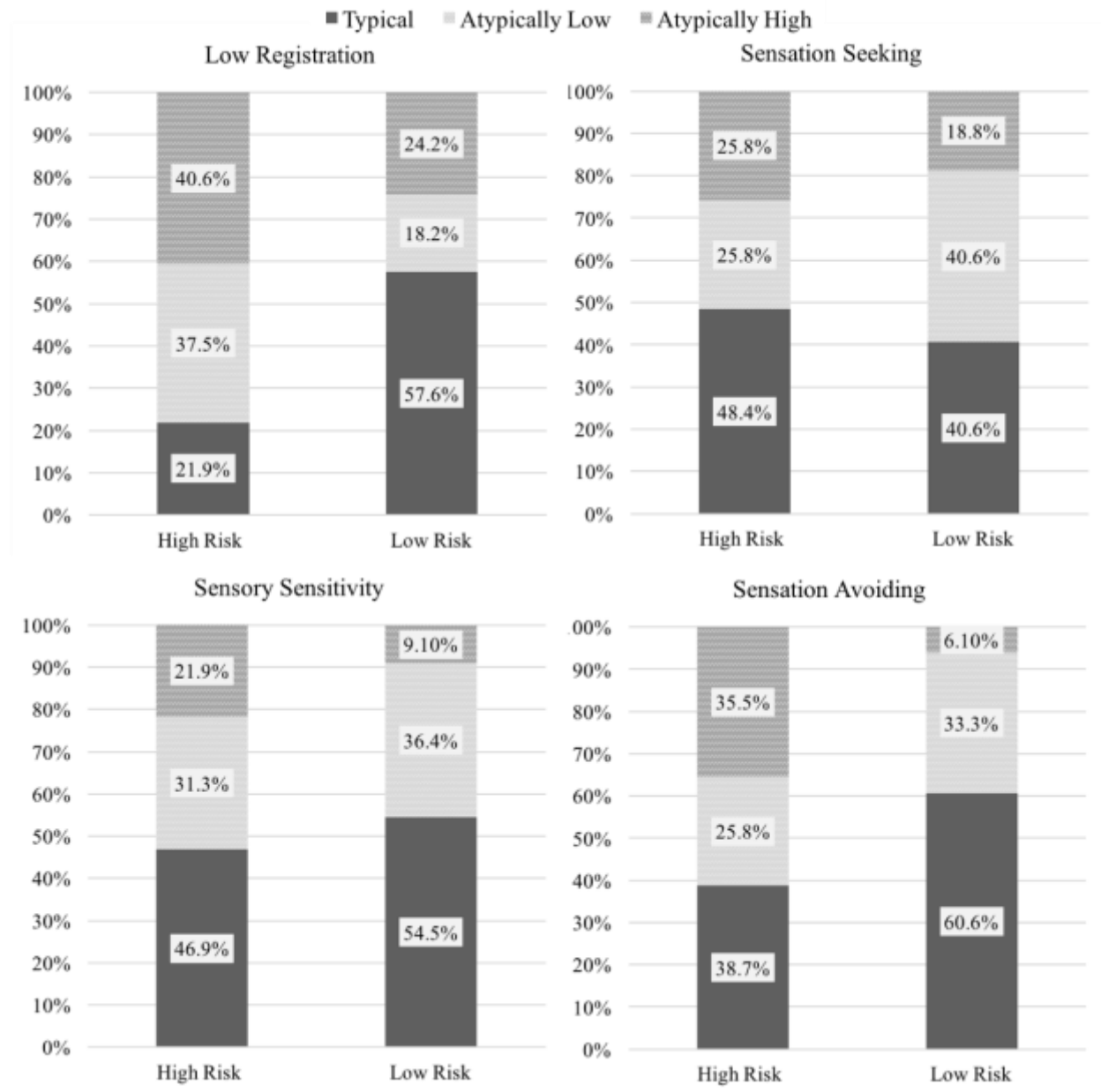


Figure 3 top

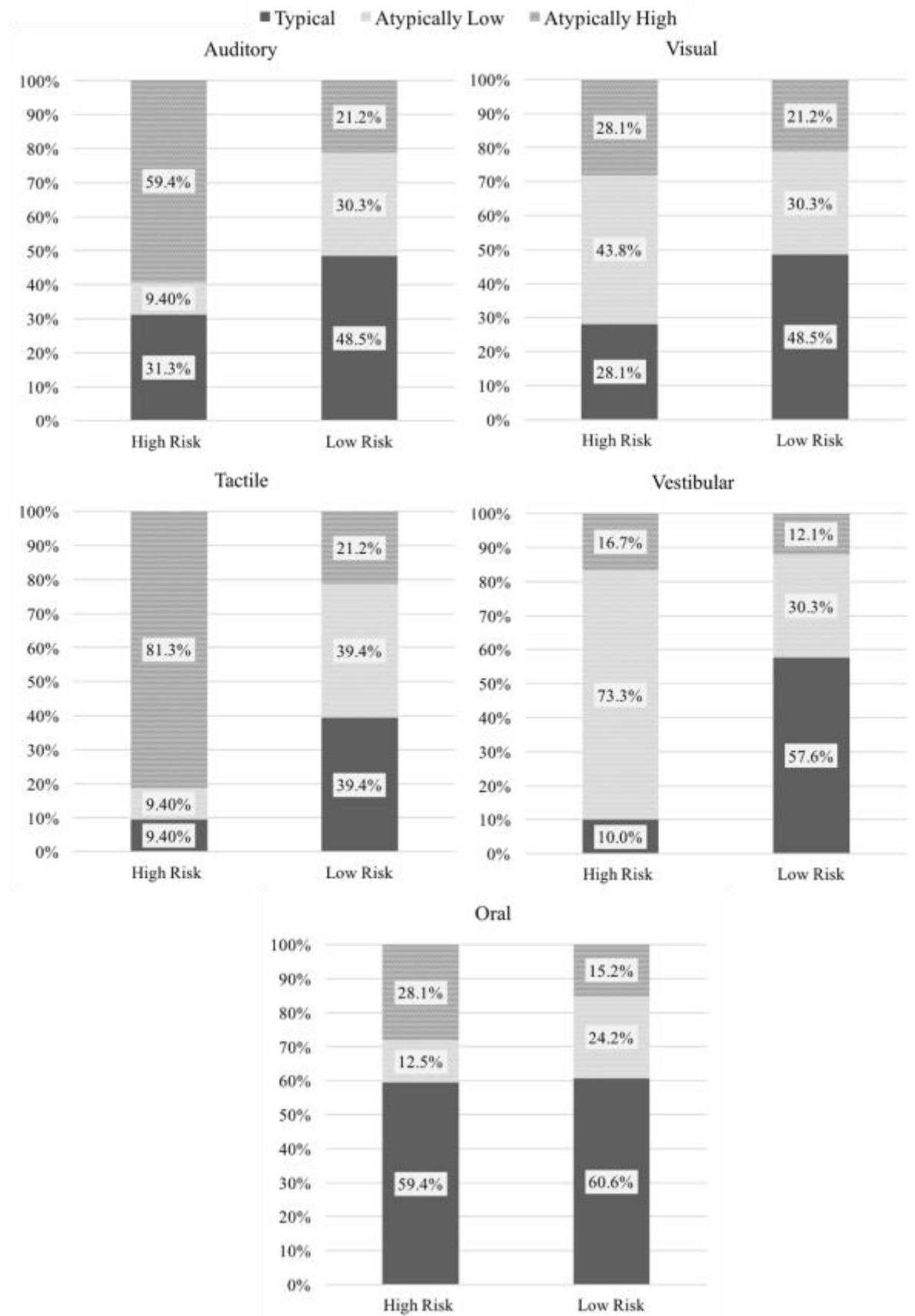


Table 1: Model of Sensory Processing. Examples taken from the Infant/Toddler Sensory Profile (Dunn 2002).

		Reaction	
		Active	Passive
Neurological Sensitivity	High	Sensation Avoiding <i>e.g. My child avoids contact with rough or cold surfaces (for example, squirms, aches, cries)</i>	Sensory Sensitivity <i>e.g. My child becomes agitated when having nails trimmed</i>
	Low	Sensation Seeking <i>e.g. My child enjoys looking at shiny objects</i>	Low Registration <i>e.g. My child takes a long time to respond, even to familiar voices</i>

Table 2: Adolescent Demographic and WASI Data

	ASD (<i>n</i> = 12)		TD (<i>n</i> = 11)		Statistics		
	Count	Percentage	Count	Percentage			
Gender	9 Male 3 Female	75.0 25.0	6 Male 5 Female	54.5 45.5	Fisher's Exact: <i>p</i> = .4		
	Mean	SD	Range	Mean	SD	Range	
Age (years:months)	15:5	1:48	13:7-17:9	16:8	1:91	13:3-19:5	F(1,21) = 3.5, <i>p</i> = .07
Verbal IQ	97.5	22.1	55-127	113.0	16.1	92-138	F(1,19) = 3.15, <i>p</i> = .09
Performance IQ	102.7	15.1	82-129	111.1	9.6	93-124	F(1,19) = 2.15, <i>p</i> = .16
Full Scale IQ	100.4	19.1	67-132	113.7	13.3	99-132	F(1,19) = 3.15, <i>p</i> = .09

Table 3: Infant Demographic and MSEL Data

	High-Risk (<i>n</i> = 32)			Low-Risk (<i>n</i> = 33)			Statistics
	Count	Percentage		Count	Percentage		
Gender	26 Male 6 Female	81.3 18.8		20 Male 13 Female	60.6 39.4		Fisher's Exact: <i>p</i> = .1
Visits	Mean	SD	Range	Mean	SD	Range	
	3.81	1.73	2-7	2.58	.94	2-6	F(1,64) = 12.94, <i>p</i> <.001
6 mo MSEL:	<i>n</i> = 27			<i>n</i> = 28			
Early Learning Composite	104	8.55	92-124	106	12.0	85-130	F(1,54) = .45, <i>p</i> = .51
8-9 mo MSEL:	<i>n</i> = 29			<i>n</i> = 30			
Early Learning Composite	106	12.4	83-132	109	15.2	83-139	F(1,58) = 1.1, <i>p</i> = .31
14-15mo MSEL:	<i>n</i> = 26			<i>n</i> = 23			
Early Learning Composite	102	12.5	74-122	108	12.8	74-127	F(1,48) = 2.2, <i>p</i> = .14

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