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Sensory Atypicalities in Dyads of Children with Autism Spectrum Disorder (ASD) and Their Parents

Magdalena Glod ¹, Deborah M. Riby ², Emma Honey ³ and Jacqui Rodgers ¹

¹ Institute of Neuroscience, Newcastle University, UK
² Department of Psychology, Durham University, UK
³ School of Psychology, Newcastle University, UK

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Corresponding Author

Dr Jacqui Rodgers

Email: jacqui.rogers@ncl.ac.uk

Telephone: +44 (0) 191 208 7562

Fax: +44 (0) 191 208 7520
Lay Abstract

**Introduction:** Sensory atypicalities are a common characteristic of autism spectrum disorder (ASD). To date, the relationship between sensory atypicalities in pairs of children with ASD and their parents has not been investigated. Exploring these relationships can advantage our understanding of contribution of familial factors towards children’s sensory profiles and sensory atypicalities to parental broader autism phenotype (a tendency to exhibit milder traits of ASD).

**Methods:** Parents of 44 children with ASD and 30 typically developing (TD) children, aged between 3 and 14 years, participated. Information about children’s sensory experiences was collected through parent report using the Sensory Profile questionnaire (Dunn, 1999). Information about parental sensory experiences was collected via self-report using the Adolescent/Adult Sensory Profile (Brown & Dunn, 2002).

**Results:** Parents of children with ASD had significantly higher scores than parents of TD children in relation to low registration, over responsivity and taste/smell sensory processing. Significant correlations were found between parents and children in ASD families but not TD pairs for sensation avoiding and auditory, visual and vestibular sensory processing.

**Discussion:** The findings suggest that there are similarities in sensory processing profiles between parents and their children in both ASD and TD dyads, however, familial sensory processing factors are likely to contribute towards the broader autism phenotype. Further work is needed to explore genetic and environmental influences on the developmental pathways of the sensory atypicalities in ASD.
Scientific Abstract

**Introduction:** Sensory atypicalities are a common feature of autism spectrum disorder (ASD). To date, the relationship between sensory atypicalities in dyads of children with ASD and their parents has not been investigated. Exploring these relationships can contribute to an understanding of how phenotypic profiles may be inherited, and the extent to which familial factors might contribute towards children’s sensory profiles and constitute an aspect of the broader autism phenotype.

**Methods:** Parents of 44 children with ASD and 30 typically developing (TD) children, aged between 3 and 14 years, participated. Information about children’s sensory experiences was collected through parent report using the Sensory Profile questionnaire (Dunn, 1999). Information about parental sensory experiences was collected via self-report using the Adolescent/Adult Sensory Profile (Brown & Dunn, 2002).

**Results:** Parents of children with ASD had significantly higher scores than parents of TD children in relation to low registration, over responsivity and taste/smell sensory processing. Similar levels of agreement were obtained within ASD and TD parent-child dyads on a number of sensory atypicalities; nevertheless significant correlations were found between parents and children in ASD families but not TD dyads for sensation avoiding and auditory, visual and vestibular sensory processing.

**Discussion:** The findings suggest that there are similarities in sensory processing profiles between parents and their children in both ASD and TD dyads. Familial sensory processing factors are likely to contribute towards the broader autism phenotype. Further work is needed to explore genetic and environmental influences on the developmental pathways of the sensory atypicalities in ASD.
Key words

sensory atypicality, parent-child dyads, autism spectrum disorder, broader autism phenotype
Introduction

Effective reception, integration and processing of sensory input, within our own bodies and from the external environment, enables us to transform sensory information into signals that we can respond to in an adaptive manner (John & Mervis, 2010). Sensory input is used to create our individual sensory maps of the body and the environment (Dunn, 1998), and this process is essential for everyday functioning and learning. Sometimes, sensory processing can however be disrupted. Sensory problems are common among individuals with neurodevelopmental disorders; including attention deficit hyperactivity disorder (ADHD), Fragile X syndrome, Williams syndrome (Ermer & Dunn, 1998; Rogers, Hepburn, & Wehner, 2003), and autism spectrum disorder (ASD; Ben-Sasson et al., 2009). Although the presence of sensory difficulties in ASD was reported in the very first descriptions of the disorder (Asperger, 1944/1991; Kanner, 1943), unusual sensory responses were included in the diagnostic criteria for the disorder only very recently (American Psychiatric Association, 2013). They are defined as “hyper- or hypo-reactivity to sensory input or unusual interests in sensory aspects of the environment (e.g., apparent indifference to pain/temperature, adverse response to specific sounds or textures, excessive smelling or touching of objects, visual fascination with lights or movement)” (American Psychiatric Association, 2013; p.50). Sensory characteristics, hence, alongside impairments in social communication and the presence of restricted and repetitive interests and behaviours, became part of a diagnostic feature.

That conceptualisation of hyper- and hypo-reactivity to sensory input could be related to the presence of high or low levels of nervous system reactivity (neurological thresholds) proposed by Dunn (Dunn, 1997). She distinguished four patterns of sensory processing: Low Registration, Sensation Seeking, Sensory Sensitivity and Sensation Avoiding, which refer to
an interaction between neurological threshold (high or low) and behavioural response (passive or active). As described by Dunn (2006) low registration indicates the degree to which an individual misses sensory input. Sensation seeking refers to the degree to which a person attempts to gain sensory input. Sensory sensitivity refers to level of detection of sensory input by an individual and sensation avoiding relates to the degree to which someone will attempt to remove themselves from sensory input.

Sensory processing characteristics effect people with ASD in a number of specific ways. Differences in sensory processing have been associated with other characteristics of ASD, such as communication and social impairments (Watson et al. 2011), repetitive behaviours (Boyd et al. 2009), over focusing of attention (Liss et al., 2006), insistence on sameness and anxiety (Uljarevic et al., 2015) and have also been associated with the presence of enhanced attention to detail (Happe & Frith, 2006) and absolute pitch (Miller, 1999). Hence, differences in sensory processing can present significant challenges across a wide range of daily life for a child with ASD, including attention, ability to learn, emotion regulation and effective management of interpersonal relationships with both peers and family members.

It is known that there is a hereditable component to ASD (Silverman et al., 2002) as shown by twin studies (Bailey et al., 1995; for the review see Ronald & Hoekstra, 2011). Interestingly, some unaffected relatives of individuals with ASD, including parents have been reported to have a number of autism-related traits, and subclinical atypicalities in social and communication skills (Gerdts & Bernier, 2011), including language skills (Ruser et al., 2007) and memory (Baron-Cohen & Hammer, 1997). This phenomenon of increased likelihood of autism-related traits in some family members of individuals with ASD (Bernier et al., 2012), known as the broader autism phenotype (BAP), has rarely been investigated in relation to sensory atypicalities.
Only one study to date (Uljarevic et al. 2014) has examined sensory processing in parents of individuals with ASD. The authors reported elevated levels of sensory atypicalities in mothers of children and adolescents with ASD, with 98% of mothers of children with ASD having sensory processing scores within an atypical range on the Adolescent/Adult Sensory Profile (AASP; Brown & Dunn, 2002) compared to a normative sample. In a similar study De la Marche, Steyaert and Noens (2012) assessed sensory processing in adolescent siblings of individuals with ASD and reported that non-affected siblings shared some aspects of an atypical sensory processing profiles with their affected sibling. In addition, data from baby siblings of children with ASD show that sensory processing differences, in particular difficulties with auditory processing and lowered registration of sensory stimulation, were more common in high-risk siblings subsequently diagnosed with ASD than in typically developing infants (Germani et al., 2014; Loh et al., 2007; Mulligan & White, 2012). These findings suggest that behavioural responses to sensory input may serve as an early risk marker of ASD, particularly in high-risk infants.

The relationships between sensory processing profiles in ASD families may not be unique to the disorder. A level of sensory heritability (perceptual sensitivity) and sensory over-responsivity in relation to both tactile and auditory processing has also been reported in the general population in monozygotic and dizygotic typically developing twins (Goldsmith, Buss, & Lemery, 1997; Van Hulle, Schmidt, & Goldsmith, 2012). Taking these findings together the limited evidence to date suggests that parents of children with ASD may also present with atypicalities in their sensory processing profiles. Surprisingly, the relationship between sensory atypicalities in matched dyads of children with ASD, and developing typically children and their parents has not been investigated.

Investigation of similarities and differences in sensory processing in parent-child dyads in neurodevelopmental disorders will inform our understanding of how phenotypic profiles may
be inherited within families. The concordance in sensory profiles between individual parent and child dyads in ASD families has never been examined. Therefore, the aim of this study was to explore the profiles of sensory processing in child-parent dyads within ASD families in comparison to TD dyads. We hypothesised that (1) parents of children with ASD would present with more sensory atypicalities than parents of typically developing children and (2) sensory processing patterns in child-parent dyads would be more similar in ASD families than in typically developing families.

Methods

Participants

Forty-four parents (38 mothers and 6 fathers) of children with ASD and thirty parents (25 mothers and 5 fathers) of typically developing (TD) children were recruited. All children with ASD had previously been diagnosed with ASD based on a multidisciplinary team assessment following the guidelines of the UK National Autism Plan for Children (Le Couteur, 2003). Additionally, for the children with ASD data from the Social Responsiveness Scale, Second Edition (SRS-2; Constantino & Gruber, 2012) were available for all children of an appropriate developmental age, with the exception of four (due to a large amount of missing data), with the scores falling between the mild to moderate (n=4; total raw score ranging from 58 to 80, mean=70, SD=9.38) and severe range (n=31; total raw score ranging from 88 to 171, mean=116.9, SD=23.73). Children for whom the SRS-2 total score could not be calculated, did not differ on gender, age and any sensory variable compared to children for whom the SRS-2 data were available. All TD children obtained scores within the normal range (0-13; mean=6.70, SD=3.73) on the Strengths and Difficulties Questionnaire (SDQ; Goodman, 1997).
Children with ASD were recruited via ASD-UK (www.ASD-UK.com), a major UK family research database of children with ASD (Warnell et al., 2015). Families whose children met the study criteria were initially sent information about the study by email or letter, and reminders were sent to non-responders. In order to ascertain whether ability plays a role in sensory atypicalities presentation, children across the ability range were recruited, so the sample included those with and without comorbid intellectual disability (ID) as reported by parents. Twenty-three children in the ASD sample also had an intellectual disability (ID). TD children were recruited through local schools, a University research volunteers’ database and word of mouth.

Measures

The Sensory Profile (SP; Dunn, 1999) is a caregiver questionnaire that measures a child’s sensory processing abilities. The questionnaire consists of 125 items, rated on a five-point Likert scale, ranging from always (1) to never (5). Children can be classified as fitting into one of the four general sensory processing quadrants: low registration, sensation seeking, sensory sensitivity and sensation avoiding. Scores on sensory processing within sensory modalities (such as tactile, visual, auditory) scores can also be obtained.

The SP is commonly used with 3 to 10 year olds, however it has been used with older ASD participants (in Kern et al., 2007 the oldest participant for whom the SP was completed was 43 years old). Cronbach’s alpha, as reported in the manual, ranged from .47 to .91 across different subscales and the tool is reported to have a good convergent and discriminant validity (SP; Dunn, 1999).
The Adolescent/Adult Sensory Profile (AASP; Brown & Dunn, 2002) is a self-report questionnaire designed for individuals between 11 and 65 years old evaluating their responses to everyday sensory events. In this 60-item questionnaire, 15 questions are related to each of the four sensory quadrants—low registration, sensation seeking, sensory sensitivity, and sensation avoiding. Scores for taste/smell, movement, visual, touch and auditory processing can also be calculated (to be consistent with the SP domains, we refer to taste/smell sensory processing using oral sensory processing term, and to movement sensory processing, using vestibular sensory processing term). As in the SP, each statement is rated on a five-point Likert scale; however, the rating system is reversed, ranging from almost never (1) to almost always (5). Some individuals may have atypical scores in more than one sensory quadrant. The internal consistency of the measure is good with alpha values ranged from .63 to .77, as reported in the measure manual, for the various quadrant scores. Evidence of good convergent and discriminant validity was also provided (AASP; Brown & Dunn, 2002).

The Social Responsiveness Scale – Second Edition (Constantino & Gruber, 2012) is a 65-item parent-report four-point Likert-like rating scale of autistic trait that covers unusual interpersonal behaviours, communication or repetitive/stereotyped behaviours. The SRS-2 describes a degree of autistic social impairment and the severity of autistic symptoms. It is reported to have good psychometric properties (Bruni, 2014).

The Strengths and Difficulties Questionnaire (SDQ; Goodman, 1997) is a 25-items caregiver-report of children of 4-16 years old that screens whether the child has any emotional, conduct, hyperactivity/inattention, and peer relationship problems or displays prosocial behaviour. The
SDQ has been widely used in large epidemiological studies and is well adapted for studies of the general population (Goodman & Goodman, 2009).

Procedure

Questionnaire packs including an information sheet, consent form, the Sensory Profile (Dunn, 1999), Adolescent/Adult Sensory Profile (Brown & Dunn, 2002), Social Responsiveness Scale – Second Edition (Constantino & Gruber, 2012; parents of ASD children only), and the Strengths and Difficulties Questionnaire (Goodman, 1997; parents of TD children only) were sent to parents who had agreed to participate in the study.

Favourable ethical opinion was granted by Newcastle University Faculty of Medical Sciences Ethics Committee.

Data analysis

After initial data entry, parents were contacted again and asked to provide missing information, if relevant. Some parents did not respond resulting in 1.27% of the SP and 0.09% of the AASP item scores missing. There were no patterns within missing data. Missing values were treated as missing at random and replaced by the mean of the non-missing subscale items when less than 20% of the data within the subscale were missing. Descriptive statistics, inferential and Intraclass Correlation Coefficient (ICC) analyses were subsequently undertaken on the complete dataset for both quadrant scores and sensory processing modalities scores. Intraclass Correlation Coefficients were used to quantify the agreement between parent-child pairs and establish consistency between the sensory processing measurements for the pairs.
Results

Descriptive statistics of participant characteristics are presented in Table 1.

(Insert Table 1 about here)

Sensory quadrants

There were no significant differences in the sensory scores between mothers and fathers in each group and between ASD children with ID and without ID. Further analyses were performed on all parents together (irrespective of gender) and all ASD participants together (irrespective of ability level).

First, one way ANOVA analyses were performed to compare group means on the sensory scores. Parents of children with ASD had significantly higher scores than parents of TD children in the Registration, Sensitivity and Avoiding quadrants ($F_{(1,72)}=4.08, p=.047$ $F_{(1,72)}=8.72, p=.004$ and $F_{(1,72)}=6.36, p=0.014$ respectively), with a higher score indicative of more atypicality. There were no other differences between the parent groups (see Table 2).

(Insert Table 2 about here)

Subsequently, Intraclass Correlation Coefficient analyses (ICC; two-way mixed, consistency) were undertaken. Due to directional differences in the Likert scale scoring of the SP and AASP (e.g. score 1 is interpreted as ‘always’ in the SP and refers to ‘almost never’ in the AASP), the Z scores of sensory quadrants were calculated (and reversed for the parental data) to estimate the level of agreement for sensory quadrants between parent-child dyads. The ICC results are shown in Table 3. Significant agreement was obtained between parents and their children in both groups on low registration and sensory sensitivity scores. There were no significant correlations between parent-child sensation seeking scores in either group. A significant
association was found between parental and child scores on *sensation avoiding* within ASD dyads, however, that correlation was non-significant within TD dyads.

(Insert Table 3 about here)

Sensory processing modalities

There were significant differences on the sensory processing modality scores between mothers and fathers in each group (ASD group: taste/smell: $t(42)=-1.997$, $p=.05$, movement: $t(42)=-1.401$, $p=.17$, visual: $t(42)=-.645$, $p=.52$, touch: $t(42)=-.035$, $p=.97$, auditory: $t(42)=2.338$, $p=.02$; TD group: taste/smell: $t(28)=-.106$, $p=.92$, movement: $t(28)=-2.345$, $p=.03$, visual: $t(28)=-2.206$, $p=.04$, touch: $t(28)=-1.582$, $p=.12$, auditory: $t(28)=1.873$, $p=.07$). Further analyses were therefore performed only on mothers.

First, one way ANOVA analyses were performed to compare group means on the sensory processing modality scores. Children with ASD had significantly lower scores ($p<.001$) than TD children in all modalities, with a lower score indicative of more atypicality. Mothers of children with ASD had significantly higher scores than mothers of TD children in the taste/smell modality ($F(1,62)=5.69$, $p=.020$), indicating more atypicality. There were no other differences between the mothers’ groups (see Table 2).

Intraclass Correlation Coefficient analyses (ICC; two-way mixed, consistency) showed that significant agreement was obtained between mothers and their children in both groups on touch processing scores. A significant association was found between parental and child scores on
auditory, visual and vestibular sensory processing within ASD dyads, however, those correlations were non-significant within TD dyads (see Table 3).

Discussion

This is the first study exploring sensory processing atypicalities in dyads of children with ASD and their parents, compared to typically developing children. Parents of children with ASD showed significantly more over responsivity sensory atypicalities, with higher scores on sensory sensitivity and sensation avoiding and more low registration difficulties compared to parents of TD children. Also mothers of children with ASD showed more taste/smell sensory processing related difficulties than mothers of TD children. The effect sizes between the groups ranged from small to medium. A similar level of agreement was obtained within ASD and TD parent-child dyads on sensory atypicalities, showing that to a degree sensory processing might be universally heritable within families, irrespective of ASD status. However a significant association between parent and child quadrant scores on sensation avoiding, and sensory processing scores on auditory, visual and vestibular processing were found in ASD families only.

In this study parents of children with ASD showed atypical sensory processing on three sensory quadrants (low registration, sensory sensitivity and sensation avoiding) in comparison to parents of typically developing children. These data are in contrast to the Uljarevic et al. (2014) study, where parent group differences were found for all sensory quadrants. However, in the current study, TD parent data were obtained directly from a control group and inferential analyses were performed. In Uljarevic et al. (2014) sensory scores of parents of children with
ASD were compared to the original American normative sample (Brown & Dunn, 2002). Further work on psychometric properties of the tool and replication of this study are required.

With regards to the results on sensory quadrants, our findings might suggest a genetic contribution for *sensory sensitivity*, in parent-child dyads. Interestingly, a similar level of agreement was found between parent and child data for both the ASD and TD groups, on the *sensitivity* quadrant suggesting that that aspect of sensory processing might be heritable, irrespective of ASD status. We did not find agreement between parent and child scores on the *sensation seeking* quadrant in either group. De la Marche et al. (2012) reported that both adolescents with ASD and their siblings had reduced *sensation seeking* and argued that sensory seeking atypicalities might be a candidate endophenotype. In this study, ASD participants showed more difficulties related to *sensation seeking* than their TD peers. Also in contrast to the familial relationship reported by De la Marche et al. (2012) we found no significant difference between parents of children with ASD and parents of typically developing children on that quadrant. This might suggest that *sensation seeking* atypicalities are not heritable, but may be more related to the presence of sensory atypicalities common for individuals with ASD or inherent in the other aspects of the disorder. The sensory processing differences in the ASD participants between the studies could also be explained by age discrepancies in the samples as younger individuals with ASD are reported to show more sensory atypicalities than adolescents (for review, see Ben-Sasson et al., 2009).

Although support for the familiality of *sensation seeking* was not found, agreement between parent and child scores on the *sensation avoiding* quadrant was found for the ASD dyads only, which suggests that this aspect of the atypical sensory processing profile may be heritable solely within ASD families. This phenomenon needs further investigation.
As in previous studies (Kern et al., 2006; Kientz and Dunn, 1997), we found that children with ASD had more sensory processing difficulties across different modalities than typically developing children. Goldsmith et al. (2006) investigated heritability of auditory and tactile defensiveness in twin study of the general population. They found that tactile defensiveness demonstrated greater heritability than auditory defensiveness. Our study supports that, as a similar level of agreement was found between parent and child data for both the ASD and TD groups on the tactile sensory processing quadrant. However, the findings also showed that for auditory, visual and vestibular sensory processing an agreement was found between child and parent scores, suggesting that for these aspects of sensory processing familial factors might play a role only within ASD families.

While our data might support the notion that sensory atypicalities may form part of the broader autism phenotype we cannot rule out the role of the environment on the development of atypical sensory profiles. There is a strong evidence that fearful behaviours can be modelled by parents and in turn increase fear in children (de Rosnay et al., 2006; Gerull & Rapee, 2002). It has been shown that parents who experience anxiety think about their children’s environments as threatening and are more likely to interpret ambiguous situations, including those child-related, as possibly distressing (Gallagher & Cartwright-Hatton, 2009). According to Rachman’s three pathways to fear (Rachman, 1977), anxiogenic learning experiences can be provided by the parents by verbal threat information, negative vicarious learning and direct aversive conditioning experiences. It is possible that the same process takes place in the intergenerational transmission of sensory-related anxieties. Parents may react to or describe certain sensory situations as threatening, modelling to their children how distressing sensory experiences can be, resulting in the attribution of fear or distress to those stimuli by the child. However, this intergenerational transmission might also occur in the opposite direction, from the child to the parent. It is possible that some parents of children with ASD become more
avoidant of certain sensory events because of their child’s often aversive, anxious and avoidant response to those sensory stimuli and this this pattern is subsequently reinforced. It has been suggested that parents of children with ASD may use an escape-avoidance coping style to deal with stressful situations more often than parents of typically developing children (Dabrowska & Pisula, 2010). It has been also shown that those mothers who were more anxious compared to nonanxious mothers, expected their children to perform more poorly on a number of experimental tasks (Creswell, Apetroaia, Murray, & Cooper, 2012), hence their perception of their children performance was biased. It is then possible that parental anxiety or stress could have influenced parental reporting of children's sensory problems.

In order to assess whether increased levels of sensation avoidant behaviours are a consequence of genotype or learnt coping strategies, longitudinal studies are needed. To establish whether auditory, visual and vestibular sensory processing atypicalities constitute a part of the broader autism phenotype, a replication study is required.

The present study has a number of limitations. Two different baseline tools were used in the children’s evaluation of autistic symptoms and emotional and behavioural difficulties (SRS-2 and SDQ). Although the measures were appropriate for the samples, using only the SRS-2 would allow for more direct comparison of some of the behavioural features between the groups. A small sample size restricted further investigation of the level of agreement between parent and child sensory profile scores for young children and adolescents with ASD separately. There is evidence suggesting that patterns of sensory processing change with development in individuals with ASD (Ben-Sasson et al., 2009) and it is unknown which aspects of sensory profiles would be shared between parents and their young or adolescent children with ASD. Also the data were obtained only from parental reports and no direct
measures of sensory processing were applied. Moreover, children with ASD without co-morbid ID were not asked to complete the SP questionnaire themselves, which could enrich our understanding of sensory processing in individuals with ASD. Information on sensory quadrant scores from mothers and fathers were combined, and presented for mothers only on sensory processing modalities. It has been suggested that females present more sensory atypicalities than males (Goldsmith, Van Hulle, Arneson, Schreiber, & Gernsbacher, 2006) and further investigation of whether a similar pattern can be found in parents of children with ASD is needed, requesting recruitment of fathers of children with ASD. Although a control group of parents of TD children was recruited to the study, including the children and parents of children with other neurodevelopmental disorders would benefit our understanding of the specificity of these findings to ASD. Last, but not least, in this preliminary study investigating sensory processing patterns in parent-child dyads, a measure of parental broader autism phenotype traits was not used. Elevated BAP features in parents could not only possibly indicate parents with atypical sensory processing, but also impact parental ability to report on their children’s sensory experiences. It is likely that highly sensitive parents might have been biased toward perceiving similar traits in their children, and equally, parents who are less sensitive might have been reporting their children as less bothered by everyday sensory input. Further studies investigating sensory processing in parents of children with ASD would benefit from including a BAP measure.

In conclusion, sensory profiles were similar for parent-child dyads across both groups, however children with ASD and their parents shared more sensory avoidant behaviours, and auditory, visual and vestibular sensory processing atypicalities compared to TD dyads. Some sensory characteristics might therefore need to be included into the broader autism phenotype features, alongside well-established social communication skills and personality traits (Gerdts &
Bernier, 2011). It is also possible that attitudes towards sensory experiences are transmitted inter-generationally. Further investigation of whether sensation avoiding, auditory, visual and vestibular atypicalities in parents of children with ASD have genetic or environmental origin, or are a result of interaction between the two, is needed.

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