Uljarević, Mirko; Lane, Alison; Kelly, Amanda; Leekam, Susan; Sensory subtypes and anxiety in older children and adolescents with autism spectrum disorder, Autism Research Vol. 9, Issue 10, p. 1073-1078 (2016)

Available from: http://dx.doi.org/10.1002/aur.1602

Accessed from: http://hdl.handle.net/1959.13/1346344
Title page

Manuscript Title: Sensory subtypes and anxiety in older children and adolescents with Autism Spectrum Disorder

Mirko Uljarević1,2*, Alison Lane3,4*, Amanda Kelly2, Susan Leekam2

(1) Olga Tennison Autism Research Centre, School of Psychological Science, La Trobe University, Victoria 3086, Australia.
(2) School of Psychology, Cardiff University, 70 Park Place, Cardiff, Wales UK, CF10 3AT.
(3) School of Health Sciences, University of Newcastle, New South Wales 2308, Australia.
(4) Priority Research Centre GrowUpWell®

Running Title: Sensory Subtypes and Anxiety in Autism

Corresponding author: Dr. Mirko Uljarević, Olga Tennison Autism Research Centre, School of Psychological Science, La Trobe University, Victoria 3086, Australia. Email: M.Uljarevic@latrobe.edu.au. Telephone: +61394796762.

Funding: This research was supported by PhD funding to MU and SL from the Wales Office of Research and Development for Health and Social Care, National Institute for Social Care and Health Research and the School of Psychology, Cardiff University.

Conflict of interest statement: None declared.

* Joint First Authors
Lay Abstract

This study had two main goals: to identify sensory subtypes in older children and adolescents with Autism Spectrum Disorders (ASD), and to examine the association between sensory subtypes and anxiety levels in this group. Mothers of 57 children and adolescents with ASD, aged 11-17 years were asked to complete the Short Sensory Profile and Spence Anxiety Scales as measures of sensory features and anxiety. A statistical technique called model-based cluster analysis grouped children into sensory subtypes based on their sensory features. Three sensory subtypes were identified and named as Sensory Adaptive, Sensory Moderate and Sensory Severe. Children and adolescents from the Adaptive Subtype had significantly lower anxiety scores when compared to other two subtypes. There were no differences between subtypes based on chronological age, expressive language or severity of autism features. This is the first study to identify the existence of sensory subtypes among older children and adolescents with ASD and explore their association with anxiety levels.
Scientific Abstract

The present study aimed to identify sensory subtypes in older children and adolescents with Autism Spectrum Disorders (ASD) and examine the relationship of sensory subtypes with anxiety levels in this group. Mothers of 57 children and adolescents with ASD aged 11-17 years (Mean age = 14 years, 2.4 months, SD= 1.81) completed the Short Sensory Profile and Spence Anxiety Scales. Model-based cluster analysis was applied to sensory profile scores to identify sensory subtypes. Three sensory subtypes, Sensory Adaptive (N= 19), Sensory Moderate (N= 29) and Sensory Severe (N= 9) were identified. The results indicated that the differences between the subtypes were well characterised by the severity of sensory symptoms and were not attributable to sensory modality or varying types of sensory-related behaviours. Children and adolescents from the Adaptive Subtype had significantly lower anxiety scores when compared to other two subtypes. There were no differences between subtypes based on chronological age, expressive language or severity of autism diagnostic features as measured by the Social Communication Questionnaire (SCQ total score). This is the first study to identify the existence of sensory subtypes among older children and adolescents with ASD and explore their association with anxiety levels.

Key Words: Sensory Subtypes, Anxiety, Autism
Introduction

Many individuals with Autism Spectrum Disorder (ASD) respond to sensory stimuli in ways that are incongruent with the intensity and nature of sensory stimulation (Ben-Sasson, Hen, Fluss, Cermak, Engel-Yeger, & Gal, 2009; Leekam, Nieto, Libby, Wing, & Gould, 2007). These sensory features are described as sensory hyper-reactivity – characterized by negative reactions to innocuous stimuli; sensory hypo-reactivity – characterized by failure to respond to sensory stimuli; and unusual sensory interests – characterized by behaviours indicating a strong preference or craving for specific sensory inputs (APA, 2013). Many individuals show a mixed pattern of both sensory hyper-and hypo-reactivity and/or unusual sensory interests (Baranek et al., 2006; Lidstone, Ujlarević, et al., 2014; Tomchek & Dunn, 2007). This mixed sensory presentation represents an interesting paradox, not easily explained by current sensory models. For example, Dunn and Brown (1997) suggest the existence of four sensory behaviour patterns: sensory hyper- and hypo-reactivity, avoidance and seeking but this model does not account for variability between individuals with ASD.

It has been suggested that a fruitful approach to understanding individual variability in both type and severity of sensory features is to go beyond describing specific sensory behaviours and instead identify subgroups of individuals with ASD with similar patterns of sensory features (Lane et al., 2010; Lane, Molloy, & Bishop, 2014). Lane et al. (2014) used model-based cluster analysis to examine parent-reported sensory features in 228 young children with ASD (aged 2-10 years). Their analysis yielded four sensory subtypes distinguishable from each other on the basis of the severity of sensory symptoms, hyper and hypo-reactivity, sensory modality and sensory interests replicating the results of previous studies (Lane et al, 2010; Lane et al, 2011). Using different statistical approaches, several other studies have also reported the existence of sensory-based subgroups of ASD children (Ausderau et al., 2014; Ben-Sasson et al., 2008; Liss, Saulnier, Fein, & Kinsbourne, 2006). All
subtyping studies to date have concentrated on young children with ASD. ASD is a lifelong condition, and sensory features tend to persist over time (Jones et al., 2003; Leekam et al., 2007; Talay-Ongan & Wood, 2000), it is therefore important to establish if systematic variation in sensory symptoms exists in older ASD groups.

Sensory features have previously been associated with increased anxiety in both ASD (Green et al., 2012; Lidstone, Uljarević et al., 2014; Wigham et al., 2014) and typically developing (Aron, Aron, & Jagiellowicz, 2012) populations. Furthermore, a study by Ben Sasson et al. (2008) found that toddlers belonging to a low frequency sensory cluster had significantly lower levels of anxiety symptoms when compared to sensory clusters characterised by more severe symptoms. However, there is less evidence about the associations between specific sensory subtypes and anxiety in a group of older children and adolescents.

The present study had two main aims: (1) to identify sensory subgroups of older children and adolescents with ASD based on distinct patterns of sensory features within individuals (sensory subtypes) and (2) examine the association between identified subtypes and anxiety levels in this age group. In keeping with the previous literature, we hypothesised that anxiety levels would be higher in individuals with the most severe sensory symptoms.

**Methods**

**Participants**

Parents of 57 children and adolescents with ASD aged 11-17 years (Mean age = 14 years. 2.4 months, SD= 1.81) from two different parts of the United Kingdom (South Wales, N = 23, and Ireland, N= 34) took part. There was no significant difference in age between two samples \((t(55)=.219, p = .828)\). All children had a community multidisciplinary team assessment leading to a best estimate clinical diagnosis of an ASD.

**Procedure and measures**
Ethical approval for the study was provided by Research Ethics Committees of Cardiff University. Both samples were recruited through local schools and parent support groups. Most parents (84%) completed and returned the questionnaires by post.

*The Short Sensory Profile (SSP; Dunn, 1999).* The SSP is a parental report of children’s sensory symptoms. Seven subscales are identified: tactile sensitivity, taste/smell sensitivity, movement sensitivity, visual/auditory sensitivity, underresponsive/seeks sensation, auditory filtering, and low energy/weak with lower scores indicating more impairment. Although both short and full sensory profile were originally designed for children aged 3-10 years, they have been also used in older children, adolescent and adults (Kern et al., 2006; Kern, Garver, Carmody, Andrews, Trivedi, & Mehta, 2007; Kern et al., 2007). In our sample all SSP subscales had good to excellent internal consistency (Cronbach’s alphas ranging from .766 to .939).

*The Spence Anxiety Scale (SCAS; Nauta et al., 2004).* The SCAS is a questionnaire designed to measure anxiety symptoms in children and adolescents available in both parent and self-report versions. For this study we chose the parental version. For each item, parents indicate the frequency of particular anxiety symptom on a 4-point scale. SCAS-P has good psychometric properties (Nauta et al., 2004) and good construct validity in ASD sample (Russell & Sofronoff, 2005). Total score was converted into standardized T scores and scores of ≥ 60 were used as a clinical cut point (see [www.scaswebsite.com](http://www.scaswebsite.com)).

*The Social Communication Questionnaire (SCQ; Rutter et al., 2003).* The SCQ is a 40-item, parent-report screening questionnaire for ASD derived from the revised version of the Autism Diagnostic Interview (ADI-R; LeCouteur et al., 1989). SCQ data were only available for South Wales sample.

A short questionnaire based on language items extracted from the Diagnostic Interview for Social and Communication Disorders (DISCO; Wing et al., 2002) and previously used in
questionnaire studies (Honey, Leekam, Turner & McConachie, 2007; Lidstone, Uljarević, 2014) provided an estimate of expressive language level.

**Analysis Plan**

Statistical analyses were conducted using the Statistical Package for the Social Sciences (SPSS v 21.0, New York, New York, USA) and R (v 3.0.3, Vienna, Austria). Model-based cluster analysis (mclust: Zhong & Ghosh, 2003) was applied to SSP z-scores to identify sensory subtypes replicating the method reported previously by Lane and colleagues (Lane et al., 2010, 2011, 2014). Model-based cluster analysis determines the best model by comparing the fit to the data of a number of possible models using the Bayesian Information Criteria (BIC) where a high BIC indicates that an optimal balance between the fit of the model to the data and the complexity of the model has been achieved. One-way analysis of variance (ANOVA) with Tukey post-hoc analyses was used to determine the sensory, age, expressive language, autism severity and anxiety differences between the clusters.

**Results**

**Aim 1: Sensory subtypes**

Three sensory clusters were identified based on optimisation of the BIC (Figure 1). Significant differences between the clusters are noted between all SSP subscales (see Table 1) and Figure 2 shows the scatterplots of all pairwise comparisons between clusters on each sensory subscale. Overall, individuals classified in Sensory Adaptive Subtype (n=19; circle icons) demonstrate scores close to or above the normative mean on all sensory subscales indicating adaptive sensory function. Individuals classified in Sensory Severe Subtype (n=9; square icons) demonstrate scores on average two to six standard deviations below the mean indicating severe sensory symptoms. Finally, individuals in Sensory Moderate Subtype 3 (n=29; triangle icons) display sensory scores one to three standard deviations below the normative mean suggestive of moderate level sensory symptoms.
There were no differences between subtypes based on chronological age, expressive language or SCQ total score (Table 1).

**Aim 2: Sensory subtypes and anxiety**

44.6% of the entire sample scored above the indicative clinical cut-off for anxiety. When frequency of anxiety was explored for each sensory subtype, 26.3% of children in the Sensory Adaptive, 66.7% in the Sensory Severe and 50% in the Sensory Moderate subtype had scores indicative of anxiety.

Children and adolescents from the Sensory Adaptive had significantly lower anxiety scores than each of other two subtypes. Sensory Severe had higher anxiety score than Sensory Moderate subtype but difference did not reach statistical significance (see Table 1).

**Discussion**

This study has demonstrated the existence and nature of individual variability between older children and adolescents with ASD as a function of sensory features, further highlighting the potential of the use of sensory features as a means to identify clinically meaningful subgroups in individuals with ASD. Specifically, three sensory subtypes were identified in our study that differed on the basis of the severity of sensory symptoms. Individuals in the Sensory Adaptive subtype did not show clinically significant sensory features, a similar pattern as identified in previous work with younger children (Lane et al, 2014). Sensory Severe, and Sensory Moderate subtypes differ from one another based on the severity of sensory symptoms. This finding
contrasts with findings in young children, which suggest sensory subtypes on the basis of sensory hyper- and hypo-reactivity, sensory seeking, sensory modality, multisensory integration, in addition to sensory symptom severity (Lane et al, 2014; Ausderau et al, 2014). No obvious differences between the sensory subtypes in our study can be attributed to sensory modality or varying types of sensory-related behaviours. It is possible that over time, the more specific patterns of sensory features identified in toddlers and younger children tend to restructure according to sensory severity continuum, however, this hypothesis remains to be explored in a longitudinal design with a particular emphasis on potential mediators and moderators of this change. Further, a definitive interpretation of the patterns of sensory features in this study is limited by the item sampling frame of the SSP. It is possible that not all relevant sensory features in older childhood and adolescence have been captured by the SSP which is limited in its inclusion of sensory perception, hypo-reactivity and multisensory items. Importantly, in keeping with findings of Lane et al. (2014), we found no discernible differences between sensory subtypes in terms of chronological age, expressive language and severity of ASD traits.

Children and adolescents in the Sensory Severe subtype had the highest anxiety scores, although the difference with Sensory Moderate subtype did not reach statistical significance. This provides some support to our initial hypothesis that individuals with more severe sensory symptoms will also experience greater anxiety. Previous studies of both ASD (Green et al., 2012; Mazurek et al., 2012) and non-ASD populations have shown a strong association between anxiety and sensory symptoms. Ben Sasson et al. (2008), utilising a different clustering approach, showed that ASD toddlers with the most severe sensory features showed highest anxiety.

Despite convergence of evidence linking sensory features and anxiety, further research is needed. It is not known whether sensory features serve as a causal factor for the development
of anxiety (Green & Ben-Sasson, 2010), whether both might be a consequence of common underlying pathology such as amygdala dysregulation, and how sensory features relate to cognitive processes and traits such as for example Intolerance of Uncertainty (IU), known to serve as main risk factors for development and maintenance of anxiety in non-ASD populations (Carleton et al., 2010). Although recent research in population of mothers of children with ASD, found that IU mediated the relationship between sensory features and anxiety (Uljarević, Carrington, & Leekam, 2015), these findings remain to be validated in ASD population.

Further research is also needed to determine the role of sensory features in the broader phenotype of parents (Uljarević, Prior, & Leekam, 2014) and other family members (de la Marche, Steyaert, & Noens, 2012) and its relation to anxiety (Uljarević, Carrington, & Leekam, 2015). This may have potential implications for clinical interventions not only in the management of children’s behaviour challenges but also in the provision of supportive strategies for parental stress.

Our findings should be treated as preliminary at this stage due to the sample size and replication in larger sample is needed. Furthermore, a reliance on single parent reported measure of sensory features and the lack of alternative, self-report and third-party informant measures presents an additional limitation that, although by no means specific to this study, should be taken into account when results are interpreted.

Summary

This is the first study to identify the existence and nature of the sensory variability between older children and adolescents with ASD and explore their differences in terms of anxiety levels.
References


Table 1. Child characteristics by subtype

<table>
<thead>
<tr>
<th></th>
<th>Sensory Adaptive (N= 19)</th>
<th>Sensory Severe (N= 9)</th>
<th>Sensory Moderate (N= 29)</th>
<th>Test Statistics</th>
<th>Cronbach’s Alpha</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Mean (SD)</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Tactile Sensitivity</td>
<td>29.79 (4.32)</td>
<td>14.11 (3.52)</td>
<td>25.97 (5.12)</td>
<td>35.22***; .54</td>
<td>.84</td>
</tr>
<tr>
<td>Taste/smell Sensitivity</td>
<td>15.21 (5.02)</td>
<td>7.33 (5.32)</td>
<td>12.83 (4.56)</td>
<td>8.11***; .2</td>
<td>.90</td>
</tr>
<tr>
<td>Movement Sensitivity</td>
<td>13.95 (1.78)</td>
<td>4 (1.73)</td>
<td>11.83 (2.88)</td>
<td>53.54***; .65</td>
<td>.89</td>
</tr>
<tr>
<td>Visual/auditory Sensitivity</td>
<td>21.26 (2.83)</td>
<td>12.78 (5.54)</td>
<td>17.21 (3.03)</td>
<td>19.43***; .39</td>
<td>.77</td>
</tr>
<tr>
<td>Underresponsive/seeks Sensation</td>
<td>31.84 (3.08)</td>
<td>21.44 (6.35)</td>
<td>22.07 (4)</td>
<td>35.79*** .55</td>
<td>.79</td>
</tr>
<tr>
<td>Auditory Filtering</td>
<td>22.21 (4.17)</td>
<td>12 (3.12)</td>
<td>16.9 (3.95)</td>
<td>22.68***; .43</td>
<td>.80</td>
</tr>
<tr>
<td>Low Energy/Weak</td>
<td>24.79 (6.34)</td>
<td>12.11 (5.75)</td>
<td>21.55 (7.42)</td>
<td>10.57***; .25</td>
<td>.94</td>
</tr>
<tr>
<td>Anxiety Score</td>
<td>50.63 (10.4)</td>
<td>65.22 (8.39)</td>
<td>57.82 (4.95)</td>
<td>7.51*** .19</td>
<td>.91</td>
</tr>
<tr>
<td>Chronological Age</td>
<td>14.65 (1.83)</td>
<td>14 (2.45)</td>
<td>13.96 (1.58)</td>
<td>.87; 0</td>
<td>NA</td>
</tr>
<tr>
<td>Expressive Language</td>
<td>4.72 (1.93)</td>
<td>5 (1.32)</td>
<td>4.86 (1.59)</td>
<td>.88; 0</td>
<td>NA</td>
</tr>
<tr>
<td>SCQ Total Score</td>
<td>25 (3.67)</td>
<td>26 (6.97)</td>
<td>23.14 (3.37)</td>
<td>.89; 0</td>
<td>.81</td>
</tr>
</tbody>
</table>
Legend: \(a,b,c\) clusters with different lettering in the superscript are significantly different by Tukey post-hoc comparisons. Numbers in superscript indicate effect sizes for post-hoc comparisons, number next to Subtype (S) 1 refers to S 1 vs S 2, next to S 2 to S 2 vs S 3 and next to S 3 to S 3 vs S 1 comparison.

*= p<.05; **= p<.01; ***= p<.001
Figure 1. The Bayesian Information Criteria (BIC) values for each Gaussian model (each represented by different icon) considered in the analysis. Each Gaussian model used in this analysis is identified in the figure legend by a combination of the letters E, V, and I. The sequence of the letters indicates specific variance and geometric features of each model. Please refer to Fraley and Raftery (2007) for a detailed description of the methodology.
Figure 2. Scatterplots of pairwise comparisons between clusters on each sensory subscale

Legend: “0” represents the normative mean for performance (i.e., typical performance) on the sensory domain. Values above and below “0” indicate the number of standard deviations from the normative mean. Values above 0 and between 0 and −1 indicate no sensory impairment; between −1 and −2 indicate mild sensory impairment; below −2 indicate clinically significant sensory impairment. Each icon represents a single case.