AutismCRC

Sensory subtypes in children on the autism spectrum

FINAL REPORT

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The Cooperative Research Centre for Living with Autism (Autism CRC)

The Cooperative Research Centre for Living with Autism (Autism CRC) is the world's first national, cooperative research effort focused on autism. Taking a whole-of-life approach to autism focusing on diagnosis, education and adult life, Autism CRC researchers are working with end-users to provide evidence-based outcomes which can be translated into practical solutions for governments, service providers, education and health professionals, families and people on the autism spectrum.

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1. Background

Children on the autism spectrum experience a significant range of traits. A major research goal is to better understand the variation in the range of traits, to assist with identifying which practices and supports may be most effective for which children. Sensory traits including hyper-reactivity, hypo-reactivity and unusual sensory interests are behavioural characteristics of autism that may provide insights into clinically meaningful subtypes. Prior research has identified distinct sensory subtypes relating to features associated with sensory reactivity (the intensity of the behavioural response to a sensory stimulus) and sensory integration (combining information from multiple sensory stimuli). Within the autism field, however, most sensory features are measured using tools that focus on sensory reactivity only.

Sensory reactivity, also known as sensory modulation, is a feature of sensory processing functioning which involves the regulation of the intensity of behavioural responses to daily sensory input. Symptoms of sensory modulation differences now form one part of the diagnostic criteria for Autism Spectrum Disorder (1). Sensory modulation differences include: hyper-reactivity (i.e. behavioural response to sensory input is too intense e.g. extreme distress experienced with an unexpected but non-threatening sound), hypo-reactivity (i.e. behavioural response to sensory input is absent or attenuated e.g. absence of a behavioural response to a painful stimulus even when one would be expected), and unusual sensory interests (i.e. seeking or craving sensory inputs). The project provides an important extension of the work identifying sensory subtypes by investigating sensory modulation differences in participants contributing to three Autism CRC datasets; Australian Autism Biobank (AAB), Longitudinal Study of Australian Students with Autism (LASA) and the Autism Subtyping Project (ASP). This project will provide key information about the nature of sensory modulation differences in autism across early and middle childhood and their relationship with other features of autism.

1.1. Introduction

Sensory features are a core behavioural characteristic of autism. Sensory features commonly described in autism are hypo-reactivity, hyper-reactivity, and unusual sensory interests. Estimates of the prevalence of sensory features in people on the autism spectrum range from 69% to 93% (2). While some individuals on the spectrum report benefits from these characteristics, other individuals can experience significant distress and attribute functional limitations to these features (3). The autism field has only recently started to provide a major research focus on this aspect of

autism, both in terms of characterising the nature of sensory features and understanding how we can best help individuals who experience associated distress.

Children and adolescents on the autism spectrum can present with multiple sensory features and rarely present with a single sensory domain involvement. As described above, consideration also needs to be given to the intensity of response to a sensory input or sensory modulation. Children and adolescents on the autism spectrum can have difficulty regulating responses to sensations and specific stimuli and may use self-stimulation to compensate for limited sensory input or to avoid overstimulation (4).

Hand et al. (2017) (5) identified that two broad sensory symptom dimensions, sensory reactivity and multisensory integration, best explain the differences between sensory subtypes in autism. However, to date, sensory subtype research has been carried out on measures that differ in their symptom focus. Ausderau et al. (2016), using the Sensory Experiences Questionnaire (6), reported four sensory subtypes in autism ranging from mild to severe in symptom severity and including sub-foci on enhanced perceptual abilities and sensory seeking (7). Lane et al. (2014), utilising the original Short Sensory Profile, also reported a four subtype structure with differences in symptom expression from mild to severe (8). In contrast, however, these authors reported sub-foci in hyper-reactivity to tastes and smells and postural and attention differences. Furthermore, there is limited previous research exploring non-sensory factors which may be associated with sensory subtypes. Recently, the Short Sensory Profile, the most commonly used sensory measure in autism, has undergone a revision (Short Sensory Profile-2; Dunn, 2014 (9)) and is now focused more exclusively on sensory modulation differences. Simpson et al (2019) (10) have published the first study to use the Short Sensory Profile-2 (SSP-2) to identify sensory subtypes in children on the autism spectrum. Children were reported to experience differences in responses to sensory input, in particular in the area of sensitivity and avoiding. A limitation of this work is the limited sample size which meant a subtype consisting of a small number of children may not have been evident in this study. Analyses were also carried out by sensory modulation domain rather than at item level which may have further obscured important differences in patterns of sensory features.

Identification of commonly co-occurring sets of sensory symptoms in sensory subtypes, may provide insights into the mechanisms underlying sensory disruption. This knowledge is important in understanding links between sensory responses and associated behaviours and will also inform the development of supports for children with sensory symptoms across all environments including school and home.

The AAB, LASA and the ASP included the SSP-2 and therefore provides an internationally unique opportunity to understand how sensory modulation differences may manifest throughout early and middle childhood. This project leverages these resources created by the Autism CRC Early Years Program 1 and School Years Program 2. This project will also contribute to the research goal of identifying clinically meaningful subtypes of autism. Outputs of this project are consistent with the themes in Programs 1 and 2 but also are consistent with improved academic outcomes in the longer term for children with autism through an improved understanding of subtypes and thus potentially leading to matching interventions and improved outcomes for children and families.

1.1.1 Project Objectives

The main objectives for this project are:

1. To identify sensory subtypes in children and adolescents on the autism spectrum aged 3-15 years, using the item level responses from the Short Sensory Profile-2 (SSP-2)
2. To comprehensively evaluate the relationship between sensory subtypes and clinical phenotype including autism traits, cognitive level, adaptive behaviour, attention and withdrawal problems, communication competence and psychiatric co-morbidities such as anxiety using the Australian Autism Biobank, the Longitudinal Study of Australian Students with Autism and the Autism Subtyping Project datasets.

2. Research design and methods

2.1. Participants and setting

Children between the ages of 2 and 17 were recruited to the Autism CRC research programs in four Australian States: New South Wales, Queensland, Victoria and Western Australia. The research programs included the AAB, LASA and the ASP (11, 12). All participants in the AAB had received a clinically confirmed diagnosis of autism per DSM-IV or DSM-5 criteria, depending on their age at diagnosis. Participants in the LASA were included if they had a score of greater than or equal to 15 on the Social Communication Questionnaire (SCQ) (13) consistent with the autism cutoff score. Participants in the LASA who had a score on the SCQ below 15 were included if they had a confirmed clinical diagnosis by a health professional in the community. For the Autism Subtyping Project a child must be less than six years of age, with a diagnosis of autism or probable autism like traits. Only children between the ages of 3 and 15 were included in this project due to the age range in which the SSP-2 has been validated (9). No other exclusion criteria were applied.

Participants provided consent for their data to be used for future research purposes for projects deemed appropriate after consideration by the respective data access committees.

Applications were submitted to the AAB and LASA data access committee to utilise phenotype data in the proposed analyses. Participants had previously provided consent for their data to be used for future research purposes for projects deemed appropriate after consideration by the respective data access committees. A negligible risk research application was submitted to the University of New South Wales (UNSW Sydney) and reviewed by a Human Research Advisory Panel (HREAP) (HC191034).

2.2. Measures

2.2.1. Sensory features

The Short Sensory Profile – 2 (SSP-2; (9)) is a standardised questionnaire assessing the sensory profile of children (3.0-14.11 years) based on their neurological threshold to sensory input and method of self-regulation (14). The parent completed questionnaire contains 34 items distributed across the four quadrants of Dunn's Sensory Processing Framework: seeking (7 items), avoiding (9 items), sensitivity (10 items) and registration (8 items) (14). The frequency with which the child displays each item is scored on a Likert scale 1 (*almost never* = 10% or less) to 5 (*almost always* = 90% or more). The SSP-2 is utilised as an interval scale as it captures frequencies of item-wise presentation so there are defined boundaries at each Likert level.

2.2.2. Autism traits

The Autism Diagnostic Observation Schedule-2 (ADOS-2) (15) is a semi-structured, standardised diagnostic observational assessment used to confirm the DSM-5 (1) diagnosis and determine the characteristics of ASD at baseline. The ADOS-2 module administered by the trained clinician or researcher is determined by the child's age and expressive language ability.

2.2.3. Cognitive and Language Function

The Mullen Scales of Early Learning (MSEL; (16)) is a standardised measure of nonverbal and verbal development in children from birth through to sixty-eight months of age. It assesses development across key domains including language, motor, and perceptual abilities. For each domain, raw scores are obtained, and a corresponding age equivalent score calculated.

2.2.4. Adaptive function

The Vineland Adaptive Behaviour Scales Second Edition (Vineland-II; (17) (caregiver self-report form) is a standardised, norm-referenced evaluation tool for children where parents report on the child's functional level. It provides a measure of adaptive behaviour in four broad domains of communication, daily living skills, socialization, and motor skills.

2.2.5. Sleep problems

The Childhood Sleep Habits Questionnaire (CSHQ) (18) is a retrospective caregiver reported questionnaire assessing a child's sleep behaviours in a typical week. It contains 33 items which are divided into 8 subscales. Parents are asked to recall sleep behaviours occurring over a "typical" recent week. Scoring is on a 3-point scale: "usually" if the sleep behaviour occurred five to seven times/week; "sometimes" for two to four times/week; and "rarely" for zero to one time/week. Some items are reverse scored in order to keep higher scores indicative of sleep problems. Summation of all items give a total CSHQ score which ranges from 33 to 99. A total score of 41 is used as a cut-off score for clinically significant sleep problems.

2.3 Procedures and analysis

Descriptive statistics, including age and gender, were prepared for all the items and subscale of the Short Sensory Profile – 2. Corresponding frequencies were also calculated to specify sample characteristics. One-way analysis of variance (ANOVA) was used to compare participant mean score for age group responses and sex (male/female) on the mean score for each question. 95% CI was calculated at the item level. Fisher's exact test was applied for pairwise post hoc analyses to identify differences between age groups. We used the Chi-square test to determine the differences in the distribution of categorical variables between different subgroups or items. Model-based Bayesian information criterion (BIC) clustering algorithm was applied to determine if there were clinical differences in item responses. We also calculated the effect and association of clinical and behavioural attributes on the Short Sensory Profile – 2. In this regard we have considered subscale level profiles of the ADOS-2, MSEL, Vineland-II and CSHQ. Pearson correlation was applied to identify the correlation and corresponding p-values between the subscale values of SSP-2 and the clinical, behavioural and sleep subscale values. For all analyses used in this study, statistical significance was set at a p-value of 0.05.

3. Findings

Participants (n = 919) were classified based on school age cohorts: preschool (3 - 5 years; n = 352), primary school (6 - 12 years; n=524) and high school (13 - 14.11 years; n=43) (Table 1). The primary school age children comprised the largest group accounting for 57% of the sample. The male:female ratio for the preschool (278:74), primary school (407:117), and high school (33:10) groupings was within current reported expected ratios for autism (3.76:1, 3.49:1, 3.3:1) (19).

Table 1. Cohort demographics

Classificlikelation	Preschool	Primary School	High School	
Age at Assessment	Ages 3-5	Ages 6-12	13-14.11	Total
Male	278	407	33	**718**
Female	74	117	10	**201**
Total	**352**	**524**	**43**	**919**

3.1. Sensory item level distribution based on age

One-way ANOVA was conducted to compare scores on the SSP-2 between children in the preschool, primary school and high-school groups. Post hoc analyses were conducted on items reported as significantly different (Table 2). Findings are as follows:

1. There was *no significant difference between groups* on 14 items [6, 9, 10, 15, 16, 18, 21, 26, 27, 28, 29, 30, 31, and 33 (p= 0.05 to p=0.74)].
2. There was, however, *a significant difference between groups* on 20 items (Table 3). Post-hoc comparisons revealed the following:
    a. Primary school group reported a significantly higher frequency of sensory responses than the preschool group on 13 items [1, 2, 3, 5, 7, 11, 12, 19, 20, 22, 23, 24, 25].
    b. Primary school group reported a significantly higher frequency of sensory responses than both preschool and high school groups on two items [8, 14].
    c. Preschool group reported significantly different mean item scores than both the primary and older group on three items [4, 13, 34] - higher scores on 'shows distress during grooming" (Item 4), and lower scores on items "becomes tired easily, especially when standing or holding the body in one position" (Item 13) and "has a hard time finding objects in competing backgrounds" (Item 34).

d. Both preschool and primary groups indicated higher mean scores on Item 17 “has temper tantrums” than the secondary group.

3. While there was an overall difference between groups on Item 32 “jumps from one thing to another so that it interferes with activities”, post-hoc analysis revealed no inter-group difference.

Table 2. Results of a One-way ANOVA between participant age group: Preschool (3-5y), Primary (6-12y) and Older (13-14y) on mean score for each SSP-2 question. SSP-2 questions that were not significantly different between the age categories included questions 6, 9, 10, 15, 16, 18, 21, 26, 27, 28, 29, 30, 31, and 33 (p= 0.05 to p=0.74). Non-significant results are excluded from Table 3.

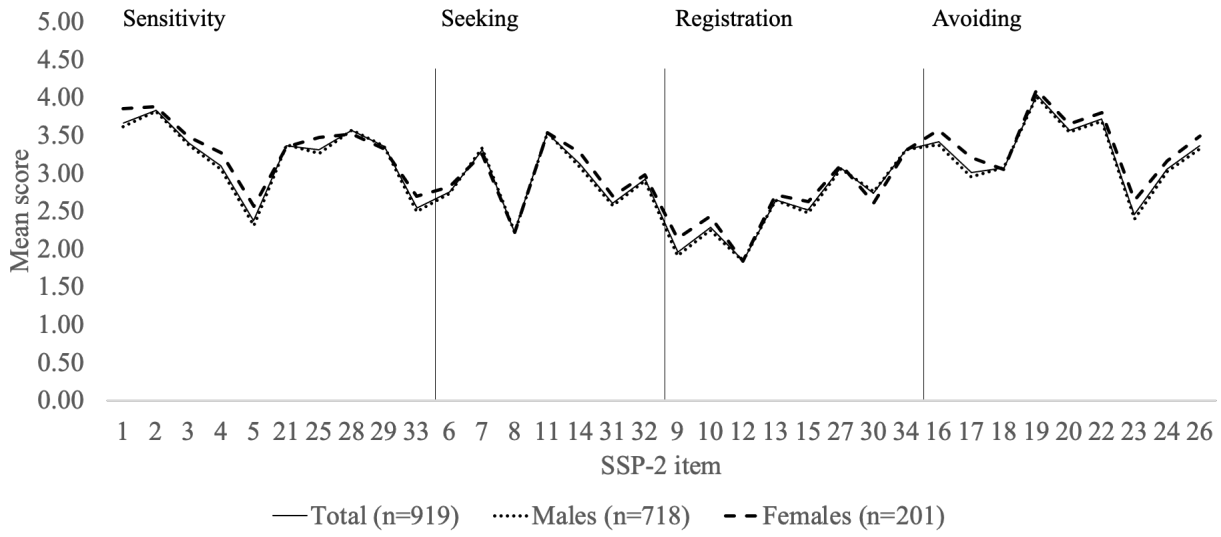
Item		Quadrant	Preschool (n=351)	Primary (n=524)	Older (n=43)	df	F	p	Difference (95% CI)		
			Mean							Preschool	Primary
1	Struggles to complete tasks when music or TV is on	Sensitivity	3.32	3.88	3.72	2, 915	19.5	<.01	**Primary**	-.557* (-0.77 to -0.35)	
									Older	-0.396 (-0.89 to 0.09)	0.161 (-0.32 to 0.64)
2	Is distracted when there is a lot of noise around	Sensitivity	3.5	4.04	3.91	2, 914	22.3	<.01	**Primary**	-.539* (-0.73 to -0.35)	
									Older	-0.404 (-0.85 to 0.04)	0.135 (-0.30 to 0.57)
3	Tunes me out or seems to ignore me	Sensitivity	3.23	3.52	3.21	2, 914	7.61	<.01	**Primary**	-.294* (-0.48 to -0.11)	
									Older	0.019 (-0.41 to 0.45)	0.314 (-0.11 to 0.74)
4	Shows distress during grooming	Sensitivity	3.51	2.88	2.42	2, 915	23.3	<.01	**Primary**	.634* (0.39 to 0.88)	
									Older	1.091* (0.52 to 1.66)	0.457 (-0.10 to 1.02)
5	Becomes anxious when standing close to others	Sensitivity	2.24	2.47	2.19	2, 915	3.43	0.03	**Primary**	-.235* (-0.46 to -0.01)	
									Older	0.05 (-0.47 to 0.57)	0.284 (-0.23 to 0.8)
7	Pursues movement to the point it interferes with daily routines	Seeking	3.19	3.42	2.91	2, 914	4.6	0.01	**Primary**	-.235* (-0.47 to 0.00)	
									Older	0.281 (-0.26 to 0.82)	0.516 (-0.02 to 1.05)
8	Rocks in chair, on floor, or while standing	Seeking	1.98	2.41	1.81	2, 915	11.1	<.01	**Primary**	-.431* (-0.67 to -0.2)	
									Older	0.166 (-0.38 to 0.71)	.597* (0.06 to 1.14)
11	Shows a strong preference for certain tastes	Seeking	3.35	3.64	3.63	2, 915	4.21	0.02	**Primary**	-.282* (-0.51 to -0.05)	
									Older	-0.275 (-0.82 to 0.27)	0.008 (-0.52 to 0.54)
12	Moves stiffly	Registration	1.69	1.94	1.86	2, 915	4.12	0.02	**Primary**	-.251* (-0.46 to -0.05)	
									Older	-0.173 (-0.66 to 0.31)	0.078 (-0.40 to 0.55)
13	Becomes tired easily, especially when standing or holding the body in one position	Registration	2.17	2.94	3.12	2, 915	29.7	<.01	**Primary**	-.766* (-1.01 to -0.52)	
									Older	-.943* (-1.51 to -0.38)	-0.177 (-0.73 to 0.38)

Item		Quadrant	Preschool (n=351)	Primary (n=524)	Older (n=43)	df	F	p	Difference (95% CI)		
			Mean							Preschool	Primary
14	Drapes self over furniture or on other people	Seeking	2.84	3.33	2.72	2, 916	14	<.01	**Primary**	-.496* (-0.73 to -0.26)	
									Older	0.117 (-0.43 to 0.67)	.613* (0.07 to 1.15)
17	Has temper tantrums	Avoiding	3.11	2.99	2.44	2, 916	4.81	<.01	**Primary**	0.117 (-0.10 to 0.33)	
									Older	.663* (0.15 to 1.17)	.547* (0.05 to 1.05)
19	Needs positive support to return to challenging situations	Avoiding	3.8	4.19	3.98	2, 916	13.2	<.01	**Primary**	-.389* (-0.57 to -0.21)	
									Older	-0.178 (-0.60 to 0.24)	0.21 (-0.20 to 0.62)
20	Has strong emotional outbursts when unable to complete a task	Avoiding	3.36	3.72	3.28	2, 916	9.05	<.01	**Primary**	-.365* (-0.58 to -0.15)	
									Older	0.079 (-0.42 to 0.58)	0.444 (-0.05 to 0.94)
22	Gets frustrated easily	Avoiding	3.52	3.84	3.6	2, 915	8.31	<.01	**Primary**	-.321* (-0.51 to -0.13)	
									Older	-0.085 (-0.52 to 0.35)	0.237 (-0.19 to 0.67)
23	Has fears that interfere with daily routines	Avoiding	2.14	2.64	2.56	2, 915	13.6	<.01	**Primary**	-.502* (-0.73 to -0.28)	
									Older	-0.416 (-0.95 to 0.12)	0.086 (-0.44 to 0.61)
24	Is distressed by changes in plans, routines, or expectations	Avoiding	2.7	3.28	3.09	2, 915	19.9	<.01	**Primary**	-.581* (-0.80 to -0.36)	
									Older	-0.391 (-0.90 to 0.12)	0.19 (-0.31 to 0.69)
25	Needs more protection from life than same-aged children	Sensitivity	3.14	3.42	3.35	2, 914	3.75	0.02	**Primary**	-.279* (-0.52 to -0.04)	
									Older	-0.209 (-0.77 to 0.35)	0.07 (-0.48 to 0.62)
32	Jumps from one thing to another so that it interferes with activities	Seeking	3.06	2.85	2.58	2, 914	3.88	0.02	**Primary**	0.212 (-0.01 to 0.43)	
									Older	0.481 (-0.04 to 1.00)	0.269 (-0.24 to 0.78)
34	Has a hard time finding objects in competing backgrounds	Registration	2.91	3.56	3.51	2, 914	20.7	<.01	**Primary**	-.654* (-0.90 to -0.41)	
									Older	-.603* (-1.17 to -0.04)	0.051 (-0.50 to 0.61)

3.2. Sensory item level distribution based on sex

Figure 1 shows the SSP-2 item level responses, grouped by quadrants, for males, females and the total group. In general, item scores for males and females were similar across the SSP-2. However, females tended to report more sensory features overall. A one-way ANOVA was conducted to compare item scores on the SSP-2 by sex (males/females). There was a significant difference between groups on six items. Females were reported to demonstrate more frequent responses on Item 1; Struggles to complete tasks when music or TV is on (p = .021), Item 5; Becomes anxious when standing close to others (p = .024), Item 9; Loses balance unexpectedly when walking on uneven surface (p = .023), Item 16; Can be stubborn and uncooperative (p = .035), Item 17; Has temper tantrums (p = .016) and Item 23; Has fears that interfere with daily routines (p = .033).

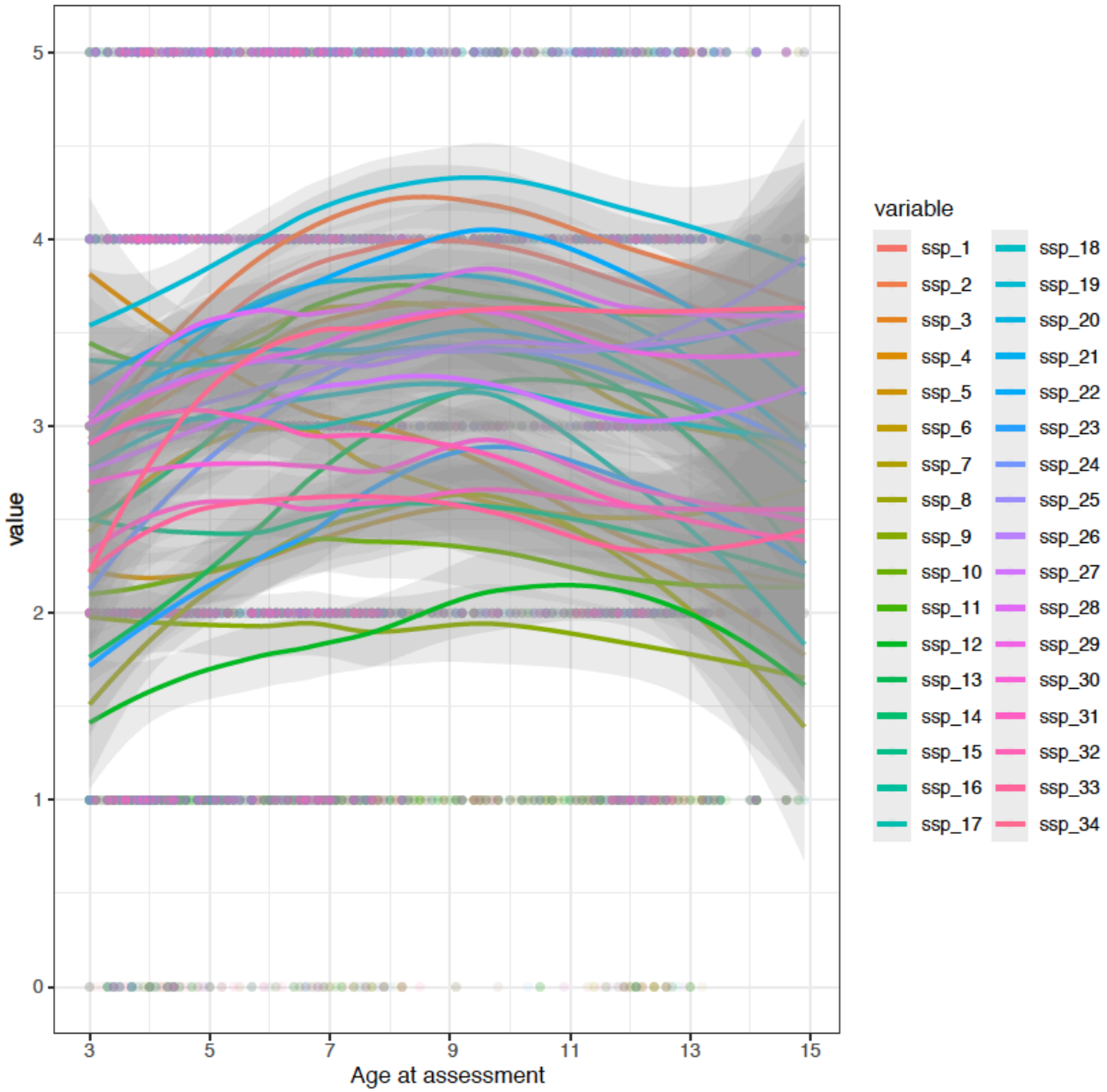
Figure 1. Sensory item level distribution based on sex



3.3. Sensory item level distribution based on age and sex.

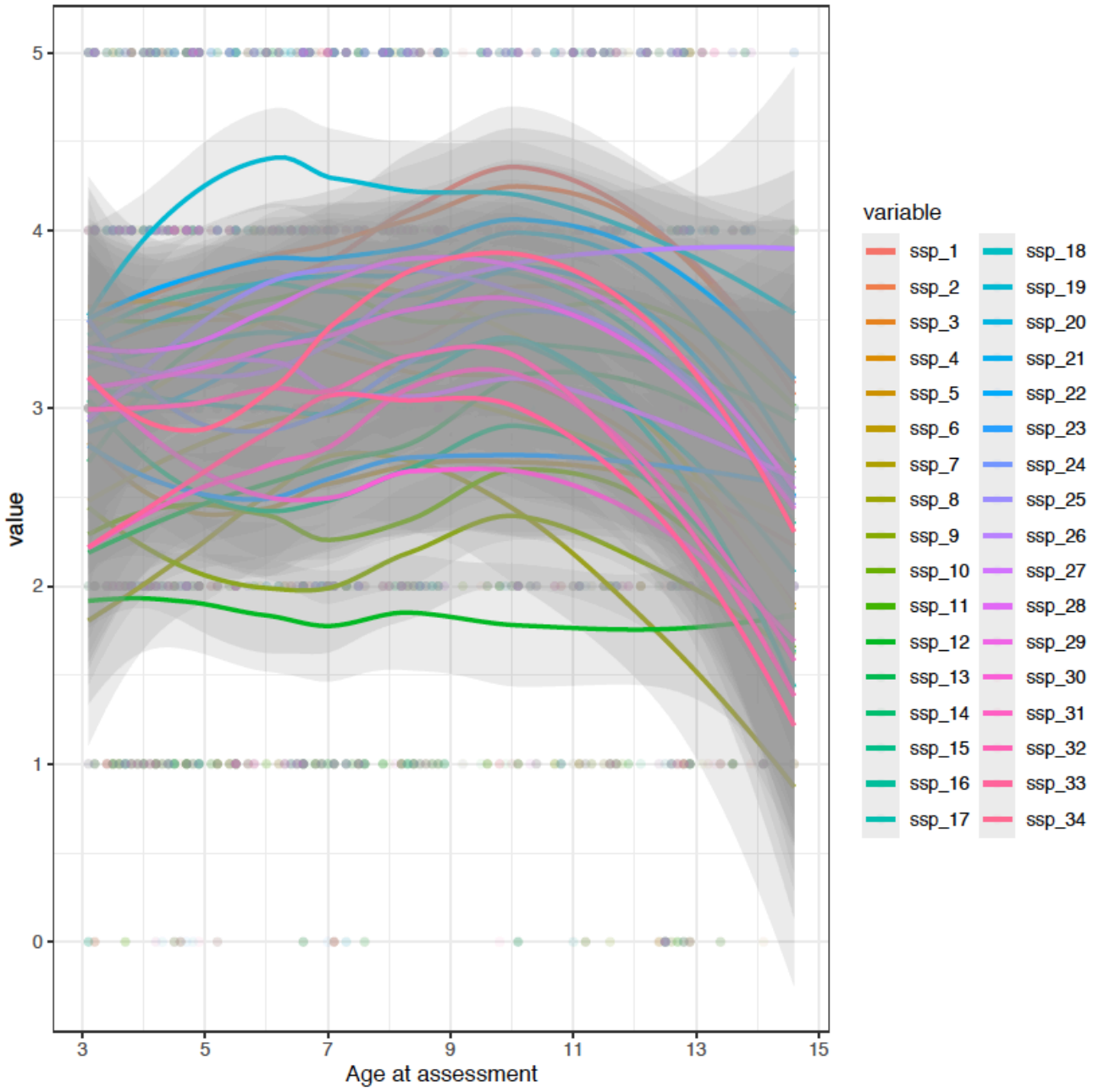
The pattern of most items scores for male participants were increased from 3 to 9 years of age and then decreased as participants approached 15 years of age (Figure 2). There were 10 items that did not exhibit this trend:

Figure 2. Sensory item level distribution of male participants based on age and sex



The pattern of most items scores for female participants were increased from 3 to 10 years of age and then decreased sharply as participants approached 15 years of age (Figure 3). There were 4 items that did not exhibit this trend:

Figure 3. Sensory item level distribution of female participants based on age and sex



3.4. Cluster analysis

Initial model-based cluster analysis generated six clusters (see Figure 3). The proportion of individuals identified in each cluster is displayed in Table 3. We analysed the differences between cluster membership by SSP-2 item (see Table 4). Overall, Clusters 1 (n = 102) and 3 (n = 296) were characterised by individuals who demonstrated adaptive or only mildly elevated responses to sensory input. In this cluster, only one item mean was observed to exceed 3.5 (Cluster 3, Item 19) indicating the vast majority of reported sensory responses were in the typical range and unlikely to be clinically significant. However, individuals in Cluster 4 (n= 138) demonstrated clinically significant responses to most SSP-2 items the majority of the time, with item means ranging from 3.0-4.85 (out of 5). Individual item means were then examined across clusters to determine those with the highest and lowest responses and to identify potential patterns of difference. Visual inspection revealed that Items 1, 2, 19, 22 and 28 were scored more frequently across all the clusters but varied predictably in their level of frequency across clusters.

- Item 1, Struggles to complete tasks when music or TV is on (Sensitivity)
- Item 2, Is distracted when there is a lot of noise around (Sensitivity)
- Item 19, Needs positive support to return to challenging situations (Avoiding)
- Item 22, Gets frustrated easily (Avoiding)
- Item 28, Struggles to pay attention (Sensitivity)

Clusters 1 and 3 showed the lowest means (indicating adaptive responses), Clusters 2, 5 and 6 displayed mid-range means (indicating elevated responses) and cluster 4 showed the highest means (indicating very elevated responses). One-way analysis of variance confirmed that mean scores for these items between clusters were significantly different from each other with cluster 2 showing difficulties in avoiding behaviours related to emotion regulation and attention (see Table 4). This suggests that cluster differences are associated with differential sensory symptoms and their impact on daily activities.

Figure 3. Cluster analysis

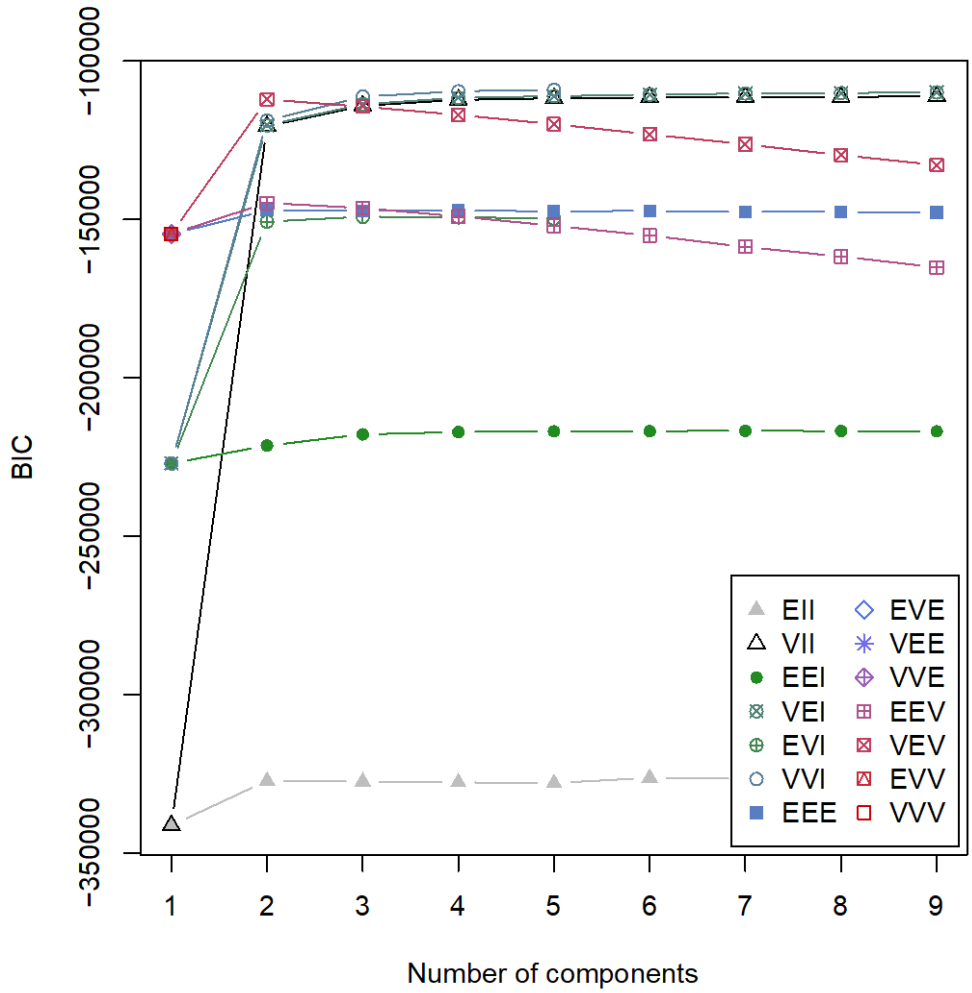


Table 3. Number of participants by cluster membership

Cluster #	N	Mean Age (SD)	Male	Female	Comment
Cluster 1	**102**	6.80 (3.05)	90	12	Generalised adaptive (all responses reported less than 50% of the time)
Cluster 2	**136**	7.58 (2.63)	110	26	Elevated avoiding and sensitivity responses (emotional regulation and attention items)
Cluster 3	**296**	6.92 (3.02)	225	71	Adaptive Text (slightly elevated avoiding and sensitivity responses)
Cluster 4	**138**	7.67 (2.69)	99	39	Elevated; Generalised
Cluster 5	**198**	7.15 (2.92)	156	42	Elevated across domains but not as severe as Cluster 4
Cluster 6	**6**	5.13 (1.43)	4	2	Elevated seeking and sensitivity
Total	**876**	6.80 (3.05)	684	192	Text

Table 4. Cluster means by SSP-2 items.

	Question	Quadrant	Cluster mean						df	F	p
			1 (N=102)	2 (N=136)	3 (N=296)	4 (N=138)	5 (N=198)	6 (N=6)			
1	Struggles to complete tasks when music or TV is on	Sensitivity	2.49	4.25	3.27	4.71	3.7	3.5	5, 868	58.36	p<.01
2*	Is distracted when there is a lot of noise around	Sensitivity	2.74	4.39	3.4	4.83	3.92	4.33	5, 868	67.96	p<.01
3	Tunes me out or seems to ignore me	Sensitivity	1.85	3.9	3.11	4.4	3.6	4.17	5, 867	111.63	p<.01
4	Shows distress during grooming	Sensitivity	2.12	3.33	2.75	3.99	3.48	3.6	5, 868	27.57	p<.01
5	Becomes anxious when standing close to others	Sensitivity	1.28	2.8	1.82	3.48	2.72	2.33	5, 866	58.85	p<.01
6	Touches people and objects more than same-aged children	Seeking	1.51	2.61	2.23	3.86	3.49	4	5, 865	60.46	p<.01
7	Pursues movement to the point it interferes with daily routines	Seeking	1.75	3.68	2.75	4.33	4.04	4.2	5, 867	90.001	p<.01
8	Rocks in chair, on floor, or while standing	Seeking	1.16	2.46	1.5	3.43	2.89	2.17	5, 866	73.86	p<.01
9	Loses balance unexpectedly when walking on uneven surface	Registration	0.97	1.35	1.33	3.43	2.89	0.8	5, 867	169.07	p<.01
10	Bumps into things, failing to notice objects or people in the way	Registration	1.11	1.7	1.59	3.78	3.31	2.67	5, 867	191.7871	p<.01
11	Shows a strong preference for certain tastes	Seeking	2.38	3.82	3.24	4.26	3.8	4.2	5, 866	29.05	p<.01
12	Moves stiffly	Registration	0.93	1.71	1.34	3.08	2.26	2.17	5, 869	69.58	p<.01
13	Becomes tired easily, especially when standing or holding the body in one position	Registration	1.4	2.54	2.05	4.12	3.18	1.83	5, 867	72.61	p<.01
14	Drapes self over furniture or on other people	Seeking	1.72	3.12	2.6	4.35	3.83	3.17	5, 870	78.09	p<.01
15	Seems accident prone	Registration	1.3	2.05	1.86	3.96	3.43	2.5	5, 866	148.71	p<.01
16	Can be stubborn and uncooperative	Avoiding	2.16	4.27	2.91	4.42	3.56	3.5	5, 870	103.08	p<.01
17	Has temper tantrums	Avoiding	1.79	4.08	2.38	4.28	3.09	2.5	5, 870	123.91	p<.01

Question		Quadrant	Cluster mean						df	F	p
			1 (N=102)	2 (N=136)	3 (N=296)	4 (N=138)	5 (N=198)	6 (N=6)			
18	Resists eye contact from me or others	Avoiding	1.75	3.62	2.77	3.96	3.19	3.83	5, 869	62.50	p<.01
19*	Needs positive support to return to challenging situations	Avoiding	2.62	4.76	3.69	4.8	4.24	4	5, 868	99.43	p<.01
20	Has strong emotional outbursts when unable to complete a task	Avoiding	2.13	4.66	2.98	4.61	3.76	3.6	5, 866	117.56	p<.01
21	Struggles to interpret body language or facial expressions	Sensitivity	1.89	3.99	3.04	4.2	3.59	2.67	5, 867	68.35	p<.01
22	Gets frustrated easily	Avoiding	2.28	4.62	3.21	4.72	3.88	3.5	5, 869	146.42	p<.01
23	Has fears that interfere with daily routines	Avoiding	1.28	3.1	1.87	3.59	2.66	1.83	5, 869	64.65	p<.01
24	Is distressed by changes in plans, routines or expectations	Avoiding	1.71	3.86	2.43	4.23	3.29	3.2	5, 866	96.37	p<.01
25	Needs more protection from life than same-aged children	Sensitivity	1.83	3.71	2.94	4.38	3.57	3.4	5, 866	54.30	p<.01
26	Interacts or participates in groups less than same-aged children	Avoiding	2.14	3.54	3.22	4.09	3.53	3	5, 864	28.85	p<.01
27	Misses eye contact with me during everyday interactions	Registration	1.52	3.6	2.81	3.93	3.23	3.4	5, 867	67.01	p<.01
28	Struggles to pay attention	Sensitivity	2.02	4.17	3.09	4.61	3.87	4.17	5, 869	146.72	p<.01
29	Looks away from tasks to notice all actions in the room	Sensitivity	2.02	3.74	2.91	4.47	3.66	3	5, 868	82.98	p<.01
30	Seems oblivious with an active environment	Registration	1.5	2.9	2.43	3.75	3	3	5, 866	52.87	p<.01
31	Watches everyone when they move around the room	Seeking	1.75	2.82	2.26	3.49	2.75	2	5, 866	31.83	p<.01
32	Jumps from one thing to another so that it interferes with activities	Seeking	1.65	3.22	2.41	3.96	3.45	4.25	5, 868	67.80	p<.01
33	Gets lost easily	Sensitivity	1.15	2.58	2.07	4.04	2.88	3	5, 864	70.96	p<.01
34	Has a hard time finding objects in competing backgrounds	Registration	1.92	3.53	2.88	4.57	3.59	4	5, 866	55.42	p<.01

3.5. Clinical characteristics

We calculated the association of the quadrants of the SSP-2 and clinical characteristics including autism symptoms, cognitive and language function, adaptive behaviour and sleep problems.

The restricted and repetitive behaviour domain of the ADOS-2 was significantly correlated with the avoiding, registration and sensory quadrants of the SSP-2 (Table 5). There were no correlations between the comparison score, which indicates level of autism-related symptoms, and the SSP-2 quadrants or the sensory and behavioural sections.

Table 5. Correlations between SSP-2 and autism symptoms

		Seeking	Avoiding	Sensitivity	Registration	Sensory	Behavioural
ADOS-2 Social Affect	Pearson R	-0.009	-0.076	0.052	-0.018	-0.055	0.013
	p-value	0.835	0.072	0.22	0.669	0.193	0.765
ADOS-2 RRB	Pearson R	-0.07	-0.114	-0.019	-0.106	-0.116	-0.058
	p-value	0.11	**0.009**	0.667	**0.015**	**0.008**	0.189
Comparison score	Pearson R	0.016	0.012	0.056	0.038	-0.011	0.061
	p-value	0.702	0.774	0.186	0.374	0.79	0.147

There were no significant correlations between the SSP-2 quadrants or the sensory and behavioural sections with the MSEL subscales (Table 6).

Table 6. Correlations between SSP-2 and cognitive and language function

Mullen Subscale		Seeking	Avoiding	Sensitivity	Registration	Sensory	Behavioural
Visual Reception	Pearson R	0.029	0.096	-0.119	0.012	0.018	-0.012
	p-value	0.675	0.162	0.083	0.864	0.796	0.864
Fine Motor	Pearson R	-0.003	0.056	-0.131	-0.02	-0.01	-0.044
	p-value	0.967	0.42	0.056	0.774	0.879	0.519
Receptive Language	Pearson R	-0.02	0.116	-0.115	0.03	0.001	0.001
	p-value	0.778	0.092	0.096	0.667	0.99	0.986
Expressive Language	Pearson R	0.008	0.123	-0.104	0.031	0.025	0.006
	p-value	0.909	0.074	0.135	0.654	0.724	0.935

All of the Vineland-II subdomains were significantly correlated with the SSP-2 quadrants and the sensory and behavioural sections except the fine motor subdomain and the avoiding quadrant (Table 7).

Table 7. Correlations between SSP-2 and adaptive behaviour

Vineland-II Subdomain		Seeking	Avoiding	Sensitivity	Registration	Sensory	Behavioural
Receptive	Pearson R	-0.292	-0.239	-0.428	-0.324	-0.321	-0.379
	p-value	<0.001	<0.001	<0.001	<0.001	<0.001	<0.001
Expressive	Pearson R	-0.085	-0.044	-0.22	-0.105	-0.085	-0.158
	p-value	0.038	0.286	<0.001	0.01	0.037	<0.001
Written	Pearson R	-0.231	-0.134	-0.273	-0.215	-0.227	-0.239
	p-value	<0.001	0.001	<0.001	<0.001	<0.001	<0.001
Personal	Pearson R	-0.204	-0.119	-0.267	-0.275	-0.251	-0.227
	p-value	<0.001	0.004	<0.001	<0.001	<0.001	<0.001
Domestic	Pearson R	-0.193	-0.183	-0.306	-0.272	-0.248	-0.276
	p-value	<0.001	<0.001	<0.001	<0.001	<0.001	<0.001
Community	Pearson R	-0.235	-0.156	-0.326	-0.278	-0.238	-0.3
	p-value	<0.001	<0.001	<0.001	<0.001	<0.001	<0.001
Interpersonal	Pearson R	-0.194	-0.221	-0.343	-0.286	-0.246	-0.321
	p-value	<0.001	<0.001	<0.001	<0.001	<0.001	<0.001
Play	Pearson R	-0.195	-0.231	-0.365	-0.271	-0.239	-0.337
	p-value	<0.001	<0.001	<0.001	<0.001	<0.001	<0.001
Coping	Pearson R	-0.253	-0.304	-0.393	-0.299	-0.29	-0.386
	p-value	<0.001	<0.001	<0.001	<0.001	<0.001	<0.001
Gross Motor	Pearson R	-0.171	-0.122	-0.212	-0.269	-0.199	-0.215
	p-value	0.001	0.023	<0.001	<0.001	<0.001	<0.001
Fine Motor	Pearson R	-0.168	-0.061	-0.225	-0.194	-0.155	-0.189
	p-value	0.002	0.26	<0.001	<0.001	0.004	<0.001
Adaptive Behaviour Composite	Pearson R	-0.282	-0.237	-0.413	-0.332	-0.306	-0.379
	p-value	<0.001	<0.001	<0.001	<0.001	<0.001	<0.001
Internalising	Pearson R	0.376	0.564	0.505	0.402	0.46	0.546
	p-value	<0.001	<0.001	<0.001	<0.001	<0.001	<0.001
Externalising	Pearson R	0.426	0.612	0.439	0.37	0.45	0.55
	p-value	<0.001	<0.001	<0.001	<0.001	<0.001	<0.001
Maladaptive Behaviour Index	Pearson R	0.515	0.685	0.598	0.497	0.567	0.679
	p-value	<0.001	<0.001	<0.001	<0.001	<0.001	<0.001

There were a number of the CSHQ subscales significantly associated with the SSP-2 quadrants and the sensory and behavioural sections (Table 8).

Table 8. Correlations between SSP-2 and sleep problems

CSHQ Subscale		Seeking	Avoiding	Sensitivity	Registration	Sensory	Behavioural
Bedtime resistance	Pearson R	0.045	-0.014	0.046	0.025	0.042	0.014
	p-value	0.283	0.749	0.275	0.561	0.318	0.749
Sleep onset delay	Pearson R	0.133	0.103	0.137	0.086	0.138	0.116
	p-value	**0.002**	**0.014**	**0.001**	**0.042**	**0.001**	**0.006**
Sleep duration	Pearson R	-0.018	-0.035	-0.041	-0.035	-0.036	-0.034
	p-value	0.672	0.409	0.333	0.402	0.399	0.425
Sleep anxiety	Pearson R	0.134	0.115	0.117	0.085	0.141	0.11
	p-value	**0.001**	**0.006**	**0.006**	**0.044**	**0.001**	**0.009**
Night wakings	Pearson R	0.103	0.096	0.125	0.072	0.095	0.115
	p-value	**0.015**	**0.023**	**0.003**	0.089	**0.025**	**0.007**
Parasomnias	Pearson R	0.146	0.183	0.176	0.145	0.158	0.195
	p-value	**0.001**	**<0.001**	**<0.001**	**0.001**	**<0.001**	**<0.001**
Sleep Disordered Breathing	Pearson R	0.081	0.074	0.1	0.072	0.091	0.091
	p-value	0.057	0.082	**0.018**	0.088	**0.031**	**0.032**
Daytime sleepiness	Pearson R	0.059	0.169	0.137	0.091	0.11	0.139
	p-value	0.165	**<0.001**	**0.001**	**0.032**	**0.009**	**0.001**

4. Discussion

The purpose of this study was to further examine the pattern of sensory processing difficulties in young people on the autism spectrum, with a focus on the sensory modulation differences identified in the DSM-5. This is the first study to examine these features across the full age range covered by the SSP-2. We observed the following:

1. Sensory modulation differences for this group peaked in the primary school age group (6-12 years)
2. Females and males presented with overall similar sensory modulation differences. However, females demonstrated heighted responses on six items: Struggles to complete tasks when music or TV is on; Becomes anxious when standing close to others; when

walking on uneven surface; Can be stubborn and uncooperative; Has temper tantrums; Has fears that interfere with daily routines.

3. Cluster analysis identified six cluster groupings which varied from each other in terms of the severity of their reported difficulties on items related to emotion regulation and attention, and age.

We discuss each of these findings below.

4.1. Age

There was an observable increase in sensory modulation differences in the primary school age group with a flattening out of the profile in the older group. The items with more frequent responses were predominantly in the hypersensitivity domain (70%), which included over half the items on the SSP-2 sensitivity and avoiding domains. This increase in frequency of hypersensitivity responses in primary school age children has been previously reported (see meta-analysis (20)). However, in contrast to Ben-Sasson et al's (20) findings, this peak did not decrease after 9 years of age. Previous research has focused on either a narrow age range, a broader definition of sensory symptoms, or a small sample size spread across a broader age range (10, 20, 21). The large sample size and the participant age range of the current study provides a unique opportunity to ascertain the sensory response in children on autism spectrum which may not have been detectable in previous studies. Trends were further confirmed in the cluster analysis, where cluster groupings were differentiated by age. Specifically, the cluster demonstrating the highest frequency of sensory responsivity difficulties included a large proportion of the primary school age cohort (Cluster 4, *N* = 99, mean age = 7.67 years).

4.2. Sex differences

Item level analysis identified no qualitative difference between males and females, with the profiles following a similar response pattern across items. However, there were quantitative differences on six items, with females demonstrating a higher frequency of sensory differences on these items. The majority of these items (83%) were in the hypersensitivity domains. Four of these items may also indicate difficulties with emotional-regulation (Item 1 "*becomes anxious when standing close to others*", Item 16 "*Can be stubborn and uncooperative*", Item 17 "*Has temper tantrums*", Item 23 "*Has fears that interfere with daily routines*"). Females on the autism spectrum aged between 9 and 12 years old have been reported to display significantly more anxious arousal behaviours than their male peers (22). This may provide some explanation for

the differences observed on these items, rather than specific differences in sensory responsivity per se. These sex differences in sensory responses differ to previous research that has examined subscale and domain scores on similar sensory measures (23, 24). However, Bitsika et al. (23) did identify sex differences at the item level using the Sensory Profile, with males reporting more differences than females on the item "moves stiffly", but this was based on a small sample size. To date, the research exploring sex-based differences on sensory profiles in children on the autism spectrum is limited. Further the use of item-level analysis in this study is expected to identify differences in items of clinical relevance that are not distinguished at a subscale or domain level using current measures. This is highlighted by the results of the current study.

4.3. Cluster discussion

Our findings provide evidence that differences in sensory responses that manifest as emotion regulation and/or attention difficulties may discriminate between subgroups of young people on the autism spectrum. This is a new finding in the area of sensory subtypes. Previous subtype studies have identified differences between sensory subtypes based on patterns of hyper- and hypo-responsivity (7), frequency of sensory responses (7, 8, 10) and sensory modality (7). Our study differed from previous work in that it focused exclusively on sensory features associated with sensory modulation function and sampled the entirety of the age domain of the SSP-2. Despite these differences, our findings were supportive of prior research in that:

1. we identified a mild/adaptive subtype that demonstrated very few significant sensory responses (Clusters 1 and 3), and
2. the frequency of elevated sensory responses was a key factor in discriminating the cluster groupings.

Our study, however, revealed a specific subset of five items which appeared to discriminate between clusters. These items appear to relate to emotion regulation and attention difficulties. The authors of the SSP-2 ascribe these behaviours to contrasting patterns of sensory hyper-reactivity (sensitivity or avoiding). Our findings appear to support the existence of subgroups of young people on the autism spectrum based on the severity of their sensory hyper-reactivity. Participants classified in Clusters 1 and 3 experienced low levels of sensory hyper-reactivity, whereas those in Cluster 4 experienced high levels. Members of Clusters 2, 4 and 6 experienced mid-range levels of sensory hyper-reactivity. These quantitative rather than qualitative differences in sensory profiles were also reported by Tillman et al. (24).

However, previous authors using the original Short Sensory Profile (25) to identify sensory subtypes suggested that behaviours related to attention difficulties (Items 1, 2 and 28) may be associated with altered sensory integration (5, 8). A critique of the work in sensory subtypes in autism to date, is that patterns of sensory responsivity vary between published studies due to the lack of a consensus guideline regarding the definition and scope of sensory responses. Our study attempted to address this concern by using a sensory instrument with a more narrowly focused domain, namely sensory modulation. Our findings suggest that further work is needed to identify the source and nature of sensory differences in autism.

Children on the autism spectrum, irrespective of their overall sensory profile, all endorsed more difficulties on one item (Item 2; “Is distracted when there is lot of noise around”). This finding is consistent with Tillman et al. (24) who reported that despite quantitative differences between their three subgroups (mild, moderate, severe), all groups showed consistent difficulties with sensitivity to noise (“Is distracted when there is a lot of noise around”). More frequently reported differences on sensitivity to noise does not appear to be unique to autism. Simpson et al. (26) reported sensitivity to noise was elevated in children with Developmental Language Disorder. This suggests that there may be sensory profiles that extend across neurodevelopmental conditions.

4.4. Clinical characteristics

Correlation coefficients between sensory modulation domain (sensitivity, avoiding, registration and seeking) and autism symptoms severity, cognition and language were small indicating no evidence for systematic relationships between these variables. There were several moderate-strong relationships observed, however, between sensory modulation domain and adaptive behaviour, specifically, the Adaptive Behaviour Composite, Internalising, Externalising and Maladaptive Behaviour Index. These findings support past reports of the close association of sensory modulation difficulties with functional limitations in daily life. While several statistically significant relationships were observed between sensory modulation domain and child sleep habits, no co-efficient exceeded 0.2 indicating that these relationships are unlikely to be clinically significant. All findings regarding the association of sensory modulation differences and clinical characteristics require closer examination with attention being paid to sensory subtype, child age and child sex for possible variations to these initial trends.

5. Limitations

5.1. Sample size

This study included a large sample size across the age range measured by the SSP-2. However, the children were predominantly between the ages of 3-12 years. The relatively small sample size of adolescents may have resulted in less sensitivity to sensory differences in this group.

5.2. Item level response

The item level responses provided a unique picture of age and sex differences, as well as highlighted differences between clusters which may not have been as apparent if analyses were conducted at a domain or subscale level. However, these profiles were obtained using responses on only one measure, and there may have been other variables that were not measured which further explain these profiles. For example, Cluster 2 was characterised by behaviours that appeared to be related to emotional regulation and attention. The inclusion of additional measures of emotional regulation and attention would provide more detail on the relationship between these factors and the response to sensory stimuli.

5.3. Parent reported measure

The SSP-2 is a parent reported measure and therefore is solely based on the parental observations of their child's behaviours rather than a subjective experience of the individual. However, sensory responsivity is an individual's subjective experience to sensory stimuli. Collecting data from multiple informants, including the young person on the autism spectrum themselves may provide a better understanding of the individual's sensory experience (21).

6. Implications for research and practice

Sensory subtyping conducted on the SSP-2 revealed differences that were largely linked to severity gradient in terms of sensory responsivity, however, we also found a subtype characterised predominantly by emotional regulation and attentional difficulties (e.g. cluster 2). The remaining subtypes could not be discriminated based on the nature/type of sensory modulation but were characterised by patterns of sensory modulation where the difference was more linked to degree rather than type of sensory responsivity.

The long term objective is to utilise the results of these analyses to identify homogeneous subgroups and to inform whether interventions for young children on the autism spectrum based on their sensory profile optimise outcomes in school participation, symptom reduction and independent daily living.

7. Key recommendations

The findings of this project on the sensory subtypes will help develop tailored practices that can support individuals who experience distress from different sensory experiences. For example, the sensory subtype with predominant emotion regulation and attention difficulties as found in cluster 2 will be important to recognise in clinical practice. Such 'personalised' approaches will be crucial in the comprehensive assessment and management of each individual on the spectrum, creating support that is tailored to their life and experience. This principle is a core part of the Autism CRC philosophy, and is also reflected in surveys from around the world that define community research priorities. For example, the recently released consultation report of the Australian Autism Research Council included an important section about how the built environment can be modified to meet the sensory needs of individuals on the spectrum. In this regard, the current findings are a critical step in the direction of increasing the evidence base and knowledge about how service providers and our community can best adapt to meet the sensory needs of those on the spectrum.

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Appendix A

Participant Consent (parent/guardian and child)

The Australian Autism Biobank Study

Declaration by Parent/Guardian

- I have read the Participant Information Sheet or someone has read it to me in a language that I understand.
- I understand the purposes, procedures and risks of the research described in the project.
- I have had an opportunity to ask questions and I am satisfied with the answers I have received.
- I freely agree to the child participating in this research project as described and understand that I am free to withdraw them at any time during the project without affecting their future health care.
- I understand that I will be given a signed copy of this document to keep.

Please indicate your consent for the following components of the study for your child / children:

I consent to the collection of a blood sample via venepuncture from which plasma, white blood cells, RNA and DNA will be extracted and stored for gene and biochemical studies	Yes ☐ No ☐
I consent to the collection, storage and analysis of a stool sample	Yes ☐ No ☐
I consent to the collection, storage and analysis of a urine sample	Yes ☐ No ☐
I consent to the collection, storage and analysis of a hair sample	Yes ☐ No ☐
I consent to the collection, storage and analysis of a saliva sample	Yes ☐ No ☐
I would like to be contacted in the future about participating in further follow-up studies	Yes ☐ No ☐

I consent to my child / children's samples being used for autism gene discovery, other biochemical research, and for research projects deemed appropriate after consideration by "The Australian Autism Biobank" Biobank Access Committee. I understand that the sample will not be used for purposes other than this. I also understand that the samples may be shared with other researchers for these purposes (and only these purposes), provided that names or addresses are not used.

I understand that through participation in this study, personal and sensitive information (including health information, and possibly information regarding ethnic or racial origin) will be collected, stored (indefinitely) and used as I would reasonably expect in accordance with The Australian Autism Biobank information sheet provided. This includes to record my child's participation, to undertake research related to autism (within and possibly outside Australia), process results and, if necessary, to contact me. I understand that I have a right to access any personal information held about my child (including to ensure its accuracy), or to complain about the handling of personal information, and that to do so I can contact any listed individual or institution on the information sheet provided.

I agree that research data gathered from the results of this study may be published and shared with other researchers, provided that names and addresses are not used.

Parent / Guardian Consent

Name of parent / guardian (please print):
_____

Signature of parent / guardian:
_____
_

Name of child / children (please print):
_____

_____
_____

_____
_____

_____
_____

_____
_____

Date: _____ **Email:**
_____

Address:
_____
_____

Phone: _____ **Mobile:**
_____

Child Assent

Name of child / children (please print)	Signature of child / children	Date

Appendix B

Longitudinal Study of Australian Students with Autism (LASA)

PARENT/CAREGIVER CONSENT FORM

By signing below, I confirm that I have read and understood the Parent/Caregiver Information Sheet and in particular have noted that:

- My involvement in this research will include completion of a questionnaire about my child with ASD and my family;
- I have had any questions answered to my satisfaction;
- I understand the risks involved;
- I understand that the Participant Coordinator will be in regular contact with me initially and then every six months and is available to contact if I have any additional questions;
- I understand that I will receive regular newsletters about the project and I will be given an annual aggregate summary of the outcomes;
- I understand that my participation in this research is voluntary and the information collected will not in any way impact on my child's relationship with their educator or medical or allied health professionals;
- I understand that I am free to withdraw at any time, without explanation or penalty;
- I understand that I can contact the Manager, Research Ethics, at Griffith University Human Research Ethics Committee on 3735 4375 (or research-ethics@griffith.edu.au) if I have any concerns about the ethical conduct of the project;
- I give permission for a member of the research team to contact me in regard to obtaining from me copies of any reports I may have in regard to my child's development or educational outcomes that are relevant to the study, e.g. previous assessments or school reports. Please tick "yes" or "No":

  ☐ Yes

  ☐ No

- I give permission for a member of the research team to contact my child's school in order to invite my child's teacher and the school principal to participate in this research study. In contacting the teacher and principal, I give my consent for you to also request my child's school records. Please tick "yes" or "No":

  ☐ Yes

  ☐ No

- I give permission for a member of the research team to contact me in regard to other research projects linked to the longitudinal study that may be of interest for me and/or my child to participate in, please tick "yes" or "No":

  ☐ Yes

  ☐ No

- I agree to participate in the LASA research project

Name	
Date	
Child's Name	
Child's school	

The following option will be provided in the online consent form::

To proceed to the online parent questionnaire package, please tick "yes", Otherwise, please tick "No".

Yes No

Who is conducting this research?

Autism Centre of Excellence[1],Griffith University: Prof Jacqueline Roberts (Project Leader)

Robyn Garland (Project Coordinator)

Speech Pathology, Griffith University: Dr David Trembath; Dr Marleen Westerveld

Faculty of Education, Queensland University of Technology: Associate Professor Sue Walker

Autism CRC Ltd, Wojciech Nadachowski

Research team contacts:

Robyn Garland (Project Coordinator): ☎3735 6869, ✉ r.garland@griffith.edu.au

Partner organisations	**Logos**
Mater Hospital, Brisbane	
Royal Children's Hospital, Victoria	
Children's Hospital at Westmead	
Department of Education, Training and Employment (DETE,), Queensland	
AEIOU, Queensland	
Autism Spectrum Australia (ASPECT), NSW	

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Our values

Inclusion

Working together with those with the lived experience of autism in all we do

Innovation

New solutions for long term challenges

Evidence

Guided by evidence-based research and peer review

Independence

Maintaining autonomy and integrity

Cooperation

Bringing benefits to our partners; capturing opportunities they cannot capture alone

Australian Government
Department of Industry, Science, Energy and Resources

AusIndustry
Cooperative Research Centres Program