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Sensory processing in low-functioning adults with Autism Spectrum Disorder:
Distinct sensory profiles and their relationships with behavioral dysfunction

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Abstract

Sensory processing abnormalities are relatively universal in individuals with autism spectrum disorder, and can be very disabling. Surprisingly, very few studies have investigated these abnormalities in low-functioning adults with autism. The goals of the present study were (a) to characterize distinct profiles of sensory dysfunction, and (b) to understand how sensory dysfunction relates to behavioral disorders in this population. Data were collected for a representative sample of inpatients in autism care centers ($N = 148$) and a non-clinical control group. Results demonstrated that sensory dysfunction (a) is highly prevalent in low-functioning adults with ASD and differentiates at least four sub-profiles of patients, and (b) predicts specific patterns of behavioral disorders. Implications for care are discussed.

Keywords

Autism spectrum disorder (ASD); Sensory profile; Sensory processing subtypes; Intellectual disability; Inpatient care center

The amount of attention devoted to sensory processing abnormalities has grown over recent years, culminating with their inclusion as a diagnostic criterion for ASD in the fifth edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5; American Psychiatric Association 2013). Indeed, sensory symptoms in autism seem relatively universal: a recent meta-analysis of fourteen studies found sensory processing abnormalities in all samples, regardless of chronological age and severity of autism (Ben-Sasson et al. 2009). The prevalence of sensory symptoms approaches 100 % in many studies (Baker et al. 2008; Baranek et al. 2006; Crane et al. 2009; Dunn et al. 2002; Leekam et al. 2007; Tomchek and Dunn 2007). These abnormalities are observable as early as the first months of life (see Iarocci and McDonald 2006) and constitute one of the most diagnostically relevant symptoms of ASD (O'Neill and Jones 1997); they also hold a central place in first-hand accounts of adults with autism (e.g. Grandin 2009).

Sensory abnormalities in ASD are extremely polymorphous (for reviews, see Gal et al. 2007; Iarocci and McDonald 2006; O'Neill and Jones 1997). All sensory modalities seem to be affected (Degenne-Richard 2014; Gal et al. 2007; Kern et al. 2006; Tomchek and Dunn 2007): although symptoms are most generally reported for auditory (O'Connor 2012), visual and tactile stimuli, abnormalities have also been identified in the processing of olfactory, gustative and even proprioceptive information. The most agreed-upon symptoms seem to be over-responsivity, under-responsivity, and sensation seeking abnormalities (Ben-Sasson et al. 2009). Over-responsivity is defined as exaggerated reactions to a stimulus (Gal et al. 2007). Individuals with ASD may be distressed or experience pain when confronted to certain stimuli, such as lights, sounds, or innocuous tactile contacts; this over-responsivity is frequently accompanied by sensory avoiding behaviors. Conversely, under-responsivity refers to the lack of awareness of certain stimuli such as sounds or spoken language. Lastly, sensation seeking abnormalities reflect a tendency to actively seek and take interest in certain stimuli such as spinning circular

objects. Other abnormalities are also observed, such as synesthesia (O'Neill and Jones 1997) and distortion of sensory input (Jones et al. 2003).

A key feature of sensory symptoms in ASD is that they vary wildly from one individual to another (e.g. Grandin 2009; Kientz and Dunn 1997). For example, a stimulus that elicits sensory overload in a person may yield no reaction at all in a different person. To better characterize this variability, several studies have attempted to identify sub-profiles of sensory processing - i.e., specific patterns of symptoms shared by different persons. All these studies have found distinct sub-profiles of sensory processing (Ausderau et al. 2014; Baker et al. 2008; Baranek et al. 1997a; Ben-Sasson et al. 2008; Degenne-Richard 2014; Lane et al. 2010; Lane et al. 2011; Liss et al. 2006), although the precise nature of these sub-profiles varies from one study to another, owing in part to large differences in methods. Certain studies have concluded that participants may be primarily characterized as either under-responsive or over-responsive (Baker et al. 2008; see also Ausderau et al. 2014, and Liss et al. 2006). Some studies have also distinguished sub-profiles based on the overall severity of sensory symptoms, with certain profiles associated with more severe abnormalities than others (Ausderau et al. 2014; Ben-Sasson et al. 2008; Liss et al. 2006). Yet other studies have distinguished sub-profiles based on affected sensory modalities (Baranek et al. 1997a; Degenne-Richard 2014; Lane et al. 2010, 2011): for example, some authors have identified a sub-profile chiefly characterized by auditory symptoms (Baranek et al. 1997a; Lane et al. 2010, 2011).

Importantly, sensory symptoms can be highly disabling for affected individuals, who tend to experience distress when confronted with abnormal sensory inputs (Leekam et al. 2007). Severe sensory symptoms are associated with impaired daily living skills (Liss et al. 2006), and are a likely cause for anxiety and depression in individuals with ASD (Pfeiffer et al. 2005). Multiple studies have also observed that the severity of sensory symptoms is predictive of the severity of behavioral disorders (Baker et al. 2008; Baranek et al. 1997b; Boyd et al. 2010;

Hilton et al. 2007; Kern et al. 2007b); likewise, different sub-profiles of sensory processing are associated with differences in behavioral disorders (Ben-Sasson et al. 2008; Lane et al. 2010; Liss et al. 2006). The understanding of sensory abnormalities therefore represents a key concern for autism care.

Despite the prevalence and clinical relevance of sensory processing abnormalities, the majority of studies have examined sensory processing in children, rather than adults with ASD (see Crane et al. 2009; Degenne-Richard et al. 2014; Tavassoli et al. 2014). The distinction is not trivial: it could well be the case that abnormalities are not stable in time and evolve throughout the lifespan (Ben-Sasson et al. 2009; Gal et al. 2007). In support of this possibility, several studies have reported that sensory processing abnormalities associated with ASD tend to decrease into adulthood (Baranek et al. 1997a; Ben-Sasson et al. 2009; Kern et al. 2006; Kern et al. 2007a; Kern et al. 2008; Leekam et al. 2007). This decrease of symptoms with aging might be due to the maturation of brain regions involved in sensory processing (Harrison and Hare 2004; O'Neill and Jones 1997), or to the fact that adults are better at coping with their sensory abnormalities (Degenne-Richard et al. 2014; O'Connor 2012). Regardless, a number of experimental results have shown that sensory symptoms persist in adults (Baranek et al. 1997a; Bontempo 2009; Crane et al. 2009; Harrison and Hare 2004; Leekam et al. 2007; Tavassoli et al. 2014), suggesting that it is clinically relevant to investigate sensory processing abnormalities in this population.

Among the few studies that included adults with ASD, most were limited to high-functioning adults with ASD and assessed their sensory symptoms using a self-report method (Bontempo 2009; Crane et al. 2009; Tavassoli et al. 2014). It is difficult to generalize the conclusions of these studies to low-functioning adults, since sensory symptoms appear to differ as a function of the overall level of functioning (Freeman et al. 1981; Leekam et al. 2007). Other studies did include low-functioning adults with ASD, but they worked with mixed

samples combining participants with or without intellectual disability, adults with different pathologies, or adults and children (e.g. Kern et al. 2006; Kern, Garver et al. 2007a, b; Kern et al. 2008), making their results difficult to interpret.

To our knowledge, only two studies have specifically assessed sensory processing abnormalities in low-functioning adults with ASD. In one study, Leekam and colleagues (Leekam et al. 2007; Study 2) found that 94 % of low-functioning adults with ASD demonstrated sensory symptoms, and that all sensory modalities were affected in at least one-fourth of participants. However, the small sample size ($N = 35$) and the limited age range (participants were aged 10–35 years; the mean age was 17.19 years) in this study make it difficult to generalize its results. In an unpublished study, Degenne-Richard (2014) also found evidence that sensory symptoms persist in adults with ASD: all participants demonstrated at least some abnormalities, and for each sensory modality, at least 60 % of participants were impaired. Although the sample size and the age range in this study were more representative of low-functioning adults in general, the author's use of a specifically-developed tool (the Evaluation Sensorielle de l'Adulte avec Autisme [Sensory Assessment of Adult with Autism]) again makes generalization difficult.

In summary, there is a dearth of empirical information on the sensory symptoms of low-functioning adults with ASD, despite the fact that this population is especially vulnerable due to the few coping mechanisms afforded by their low autonomy. The overarching goal of the present study was to remedy this lack of empirical information. Our objectives were twofold: (1) characterizing sub-profiles of sensory abnormalities in low-functioning adults with ASD; and (2) examining how profiles of sensory abnormalities relate to behavioral dysfunction in this population.

Method

Participants

A sample of 148 low-functioning adults with ASD¹ (44 females and 104 males; mean age = 32.98 years, SD = 8.82, range = 19–59 years) was included in the study. All participants lived in French inpatient care centers (either Foyers d'Accueil Médicalisé or Maisons d'Accueil Spécialisé); a total of 20 care centers were visited for the study, and five to eight inpatients per center were included in the protocol. All patients were diagnosed with a form of ASD, in reference to the revised version of the fourth edition of the DSM (DSM-IV TR; American Psychiatric Association 2000). More specifically, 70 % of participants (n = 103) were diagnosed with autistic disorder (299.00 DSM-IV TR) and 30 % of participants (n = 45) were diagnosed with atypical autism (299.80 DSM-IV TR). Patients with other diagnoses from the family of pervasive developmental disorders (such as Asperger's disorder, Rett's disorder, and childhood disintegrative disorder) were not included in the protocol. The original diagnoses of autism were made by psychiatrists and motivated the initial admission in care centers; patients were then classified into one of the categories of the DSM-IV TR by the investigators at the time of the study using available information. All patients suffered from profound to severe intellectual disability (318.1 or 318.2 DSM-IV TR) and low autonomy, motivating their permanent residence in a care center. Most participants had no access to spoken language (n = 86) or mainly demonstrated echolalia (n = 16); some participants were able to formulate a few words (n = 20) or simple sentences (n = 26). Almost half the participants suffered from epilepsy (n = 72), and most were under psychoactive medication (n = 124). Exclusion criteria were the

¹ This sample size is in line with past research analyzing subprofiles of sensory dysfunction in participants with ASD: not counting the national survey reported by Ausderau et al. 2014, the average sample size was $N = 99$ in the studies cited here (range = 22–170; Baker et al. 2008; Baranek et al. 1997a; Ben-Sasson et al. 2008; Degenne-Richard 2014; Lane et al. 2010, 2011; Liss et al. 2006). For the relationship between sensory dysfunction and behavioral disorders, most effect sizes reported in prior studies ranged between $R^2 = 0.05$ and $R^2 = 0.20$ (Ben-Sasson et al. 2008; Lane et al. 2010; Liss et al. 2006); a power analysis indicated that a sample size of $N = 152$ was required to detect an effect size of $R^2 = 0.05$ with 0.80 power.

presence of major vision loss, hearing loss or movement disorder; uncertainty on the diagnosis of ASD; and continuous residence in the same care center for less than 6 months.

A matched sample including 148 non-clinical control participants was recruited in the community (44 females and 104 males; mean age = 31.51 years, SD = 10.95, range = 19–62 years; mean age not significantly different from the ASD group, $p = 0.227$). They participated in the study on a voluntary basis, and received no benefit or compensation. Exclusion criteria were a native language other than French; the presence of acute sensory problems (e.g., hearing loss or color blindness); history of neurological disorders; psychoactive medication; intellectual disability; and history of developmental disorders².

Materials

Dunn's Sensory Profile

Sensory processing abnormalities were assessed with the adult form of Dunn's Adolescent/Adult Sensory Profile questionnaire (AASP; Brown et al. 2001; Brown and Dunn 2002), in its French-speaking version (Brown and Dunn 2006). This scale was chosen both because it has been used in a large share of studies on sensory processing in ASD (Ben-Sasson et al. 2009), and because it provides a relatively exhaustive assessment of sensory abnormalities, with items related to visual, auditory, tactile, taste/smell and kinesthetic modalities. The scale comprises four subscales assessing different dimensions of sensory processing: low sensory registration (under-sensitivity), sensory sensitivity (over-sensitivity), sensation seeking and sensation avoiding behaviors. The scale comprises 60 items; each item describes a sensory symptom (e.g., "I am uncomfortable wearing certain fabrics"). Participants rate the frequency of this symptom on a scale from 1 (almost never) to 5 (almost always), and

² The main purpose of the control group was to obtain a baseline for the analysis of sensory sub-profiles: for example, it was of interest to characterize ASD patients as "over-sensitive" or "under-sensitive" when compared to a neutral reference point. For this reason, a non-clinical control group was most appropriate to the purpose of this study. Past studies have often based analyses of sensory profiles on non-clinical normative data (e.g. Lane et al. 2010).

the scores are then summed for all items within a domain. This procedure yields one score per subscale. Normative cut scores can be used to categorize participants: for example, a score more than one standard deviation above the mean is labeled as "more than most people" and a score more than two standard deviations above the mean is labeled as "much more than most people".

The questionnaire was modified in two ways for this study. Firstly, a number of items were reworded to better fit a sample of adults permanently living in a care center: for example, "I leave or move to another section when I smell a strong odor in a store (for example, bath products, candles, perfume)" was changed to "I leave or move to another place when I smell a strong odor in a room (for example, smell of cooking food, soap, perfume)". Secondly, all items were rephrased to change the questionnaire from self-report to other-report for the ASD sample (e.g., "I only eat familiar food" was changed to "The person only eats familiar food"). The other-report method is routinely used in studies on adults with ASD (e.g. Degenne-Richard 2014; Harrison and Hare 2004; Leekam et al. 2007).

Behavioral Dysexecutive Syndrome Inventory (BDSI)

The BDSI (Godefroy and GREFEX 2008; Godefroy et al. 2010) is a 96-items scale designed to assess dysexecutive behaviors in adults using the other-report method. The scale evaluates twelve domains of executive dysfunction, such as hypoactivity, irritability, and stereotyped behaviors. Each domain comprises eight items, with each item reflecting a different behavior. The informant rates the frequency and the severity of each behavior on a Likert scale. These two scores are then multiplied and the results are summed across all items, so as to yield one total score per domain. The present study assessed all but three domains of the scale (9. anosognosia, 10. confabulations, and 12. disorders of sexual, eating and urinary behavior).

Echelle Pour l'Observation des Comportements d'Adultes avec Autisme (EPOCAA)

The EPOCAA (Recordon-Gaboriaud and Granier-Deferre 2011) is a 198-items other-report scale specifically designed to assess behavioral dysfunction in adults with ASD. The

scale evaluates thirteen areas of behavioral dysfunction, such as inappropriate social interactions, sensitivity to change, and self-aggression. Each subscale comprises a variable number of items, with each item reflecting a different behavior. The informant rates the frequency of each behavior on a Likert scale, and the results are then summed across all items to yield one total score per domain of dysfunction.

Procedure

The data for the ASD sample were collected in the context of a nation-wide survey of the features of care centers for adults with ASD (Longu  p  e et al. in press). In the course of this study, a set of 20 French inpatient care centers were visited to assess the conditions of care, behavioral disorders and sensory profiles of the inpatients. The care centers were selected to be as representative as possible: they covered a large geographical area, included both urban and rural centers, and they had been operating for 2–29 years. The two types of care centers routinely housing low-functioning adults with ASD in France were represented in approximately equal proportion. Features of the patient sample included in the present study were very similar to prior research (in terms of gender, age and level of intellectual disability; see Degenne-Richard 2014), confirming representativeness of the sample. For each participant, the Sensory Profile, the BDSI and the EPOCAA were completed by the health or social worker who had the most frequent interactions with this person (e.g., a psychologist, a physician...).

To ensure that all informants had the same understanding of the questions, the scales were filled out with the assistance of one of the investigators. In total, completing the protocol took approximately 2 hours and a half per participant in the ASD sample. Participants in the control group were recruited by word of mouth in the community. They filled out the Sensory Profile in the presence of an investigator, who was available to explain the items. Completing the scale took them approximately 10 minutes.

Data Analysis

The data were screened prior to the analysis; there were no missing values or outliers for any of the scales. Because patients with ASD were clustered into care centers, they did not constitute independent observations, which usually poses a problem for statistical analysis. However, a preliminary analysis indicated that the clustering of participants did not influence responses on the sensory profile (i.e., inpatients recruited in the same care center were no more similar than patients recruited in different care centers; all intraclass correlation coefficients [ICCs] < 0.05). For this reason, the clustered structure of the data was ignored and traditional analyses were performed.

The statistical analyses were broken down into three parts. A first series of analyses was conducted to confirm the existence of sensory abnormalities in the ASD sample when compared to the non-clinical norm. We first tested for systematic deviations from the norm in the ASD sample by comparing the average scores of the ASD and control samples on each subscale of the Sensory Profile using a series of unequal variances two-sample t-tests. To determine whether each patient demonstrated sensory abnormalities, individual scores on each subscale were then compared with the normative cut scores of the Sensory Profile (Brown and Dunn 2006; similar results were obtained when computing ad hoc normative cut scores based on the results of the control group in the present study).

The second series of analyses sought to identify distinct sub-profiles of sensory dysfunction in the ASD sample. The data were analyzed using hierarchical cluster analysis (HCA; see e.g. Kaufman and Rousseeuw 1990). The rationale of HCA is to compute the estimated multivariate distances between individuals on measures of interest, and to construct sub-groups or clusters of individuals which lie close to one another. The scores of patients with ASD on each subscale of the Sensory Profile were retrieved and standardized, distances between observations were computed as squared Euclidean distances, and observations were

aggregated using Ward's method (Ward 1963). The stability of the participant classification was ensured by performing a second analysis using the k-means method and confirming that participants were classified in the same clusters by the two methods. The average scores of each cluster on the Sensory Profile were then compared with the control group using Dunnett's test (1955).

The final series of analyses tested the relationship between sensory dysfunction and behavioral dysfunction. To this end, we first examined all bivariate correlations between the Sensory Profile and the two behavioral disorder scales, the BDSI and the EPOCAA. We then investigated whether patients with different sub-profiles of sensory dysfunction suffered from different types of behavioral disorders. Differences between sensory clusters on all subscales of the EPOCAA and BDSI were tested with omnibus analyses of variance (ANOVAs); when an ANOVA was significant or marginally significant, post hoc comparisons were performed using Fisher's Least Significant Difference (LSD) test with a 0.05 significance level.

Results

Preliminary Analysis: Confirming Sensory Dysfunction in Patients with ASD

The results of the comparison between the average scores of the two samples on the Sensory Profile are displayed in Table 1. On average, patients with ASD demonstrated significantly more low registration behaviors and significantly less sensation seeking, sensory sensitivity and sensation avoiding behaviors than control participants. The examination of normative cut scores revealed that 77 % of participants in the ASD sample ($n = 114$) obtained a rating of "much more than most people" or "much less than most people" on at least one subscale of the Sensory Profile; for reference, this was the case for only 14 % of participants in the control group. More specifically, 14 % of participants in the ASD sample ($n = 20$) had extreme scores on the low registration subscale, 6 % of participants ($n = 9$) had extreme scores on the sensory sensitivity subscale, 60 % of participants ($n = 89$) had extreme scores on the

sensation seeking subscale, and 15 % of participants ($n = 22$) had extreme scores on the sensation avoiding subscale. In other words, most patients with ASD were impaired to some extent on at least one dimension of sensory processing.

Objective 1: Identifying Profiles of Sensory Dysfunction in Patients with ASD

The first objective of our study was to determine whether patients with ASD can be classified in distinct sub-profiles of sensory dysfunction. The results of the HCA are displayed in Figure 1. The analysis clearly revealed distinct subgroups of participants with different sensory profiles; the most interpretable cluster solution was obtained by retaining four sub-profiles of participants. A second analysis using the k-means method with the hypothesis of four clusters indicated that 93 % of participants (137 out of 148) were classified in the same cluster as with the HCA, indicating very good stability of the cluster solution.

Descriptive statistics for each cluster are displayed in Table 2. On average, the four clusters were significantly different on each subscale of the Sensory Profile (all F s > 40.00 , all p s $< .001$), but they were comparable in terms of age ($F = 0.12$, $p = 0.951$) and gender ($\chi^2 = 1.85$, $p = .603$). An analysis using an ordered logistic model revealed that patients in the four clusters differed in terms of verbal level ($\chi^2 = 7.97$, $p = .047$): patients of cluster C had lower verbal levels and patients of cluster D had higher verbal levels on average.

The average scores on the Sensory Profile of participants in the four clusters were then compared with the control group. The results are summarized in Figure 2. Overall, the four clusters demonstrated different patterns of sensory dysfunction in reference to control participants.

- Patients in Cluster A were characterized by their over-sensitivity: they were the only patients to score significantly higher than the control group on sensory sensitivity and on sensation avoiding, and they displayed extremely low sensation seeking.

- Patients in Cluster B had a profile of under-sensitivity: they were not different from the control group on sensory sensitivity and sensation avoiding, but they had significantly greater low registration and demonstrated the most sensation seeking behaviors of all patients with ASD, suggesting that they actively sought to compensate their under-sensitivity.
- By contrast, patients in Cluster C were globally under-responsive, in terms of both behavior and sensory processing: they demonstrated extreme low registration, but they also displayed extremely little sensation seeking and scored significantly below controls on sensation avoiding. The significantly lower verbal level of patients in this cluster suggests that they had more severe autistic symptoms altogether.
- Lastly, patients in Cluster D had a relatively balanced sensory profile with limited sensory dysfunction: they were neither over-sensitive nor under-sensitive, demonstrated little sensation avoiding, and had relatively high sensation seeking compared to other clusters. These patients also had significantly higher verbal levels than the others.

Table 1

Two-sample t-tests for differences between ASD and control participants

Subscale	ASD sample		Control group		t-statistic	p	η^2
	M	SD	M	SD			
Low registration	34.59	7.89	29.74	6.67	5.72	<.001	.10
Sensory sensitivity	30.61	7.33	32.55	7.44	2.25	.025	.02
Sensation seeking	33.78	7.64	48.67	6.83	17.68	<.001	.52
Sensation avoiding	30.84	9.58	33.57	7.92	2.66	.008	.02

Table 2

Descriptive statistics per cluster

Cluster	N	Gender	Age	Verbal level	Low registration	Sensory sensitivity	Sensation seeking	Sensation avoiding
Cluster A	44	32% male	32.82 (9.40)	0.80 (1.13)	31.32 (5.83)	36.61 (6.29)	30.05 (5.75)	42.68 (5.20)
Cluster B	35	37% male	32.74 (8.19)	0.71 (1.10)	38.23 (5.10)	32.43 (5.22)	40.20 (5.33)	29.49 (5.50)
Cluster C	24	25% male	33.96 (10.19)	0.63 (1.06)	44.92 (7.08)	29.46 (5.18)	25.88 (5.04)	26.62 (5.58)
Cluster D	45	24% male	32.80 (8.19)	1.31 (1.31)	29.47 (4.69)	23.96 (4.58)	36.64 (6.19)	22.58 (4.42)

Note. Means with standard deviations in parentheses. Verbal level is scored on a scale from 0 to 3 (0 = no spoken language, 1 = echolalia, 2 = simple words, 3 = simple sentences)

Table 3

Correlations between the Sensory Profile and the BDSI

Subscale	GHA	DIA	DI	HD	IA	EEL	SPB	ED	SBD
Low registration	.40	.41	.38	.04	.00	-.07	.16	-.18	.13
Sensory sensitivity	.17	.15	.14	.08	.06	-.05	.22	.04	.14
Sensation seeking	-.35	-.39	-.45	.14	.24	.29	-.09	.28	.22
Sensation avoiding	.14	.07	.13	-.14	-.08	-.16	.21	-.16	-.11

Note. Significant correlations are in bold. GHA global hypoactivity and apathy, DIA difficulty for initiating activities, DI disinterest and indifference, HD hyperactivity and distractibility, IA irritability and aggressiveness, EEL euphoria and emotional lability, SPB stereotyped and perseverative behavior, ED environmental dependency, SBD social behavior disorders

Table 4

Correlations between the Sensory Profile and the EPOCAA

Subscale	IS	SI	EC	EDA	SA	OA	EA	SBSS	RCF	OU	RSS	ISB	LA
Low registration	.22	.36	.24	.20	.27	-.07	.27	.27	-.04	.18	-.11	.17	.39
Sensory sensitivity	.28	.33	.22	.14	.10	.00	.19	.24	.43	.07	.26	.05	-.01
Sensation seeking	-.40	-.42	-.30	.06	.09	.23	.14	.00	.13	.23	.24	.24	.03
Sensation avoiding	.49	.33	.31	.01	.03	-.09	.00	.25	.34	.02	.27	-.06	-.11

Note. Significant correlations are in bold. IS isolation seeking, SI social interactions, EC eye contact, EDA emotional disorders and anxiety, SA self-aggression, OA other-aggression, EA expression of affectivity, SMAR stereotypical behavior and self-stimulation, RCF reactivity to change and frustration, OU object use, RSS reactivity to sensory stimuli, ISB inappropriate social behavior, LA limited autonomy

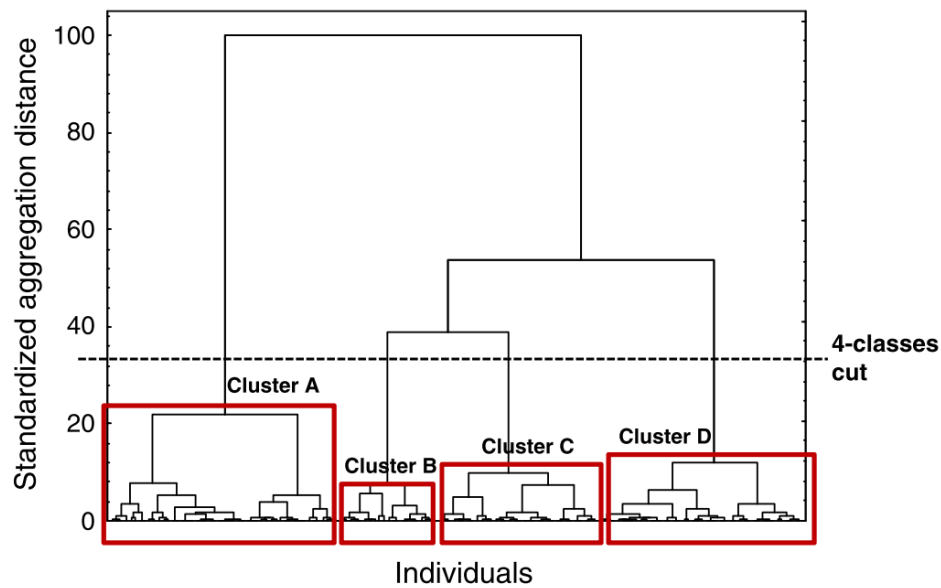


Figure 1. Dendrogram for a hierarchical cluster analysis on patients with ASD

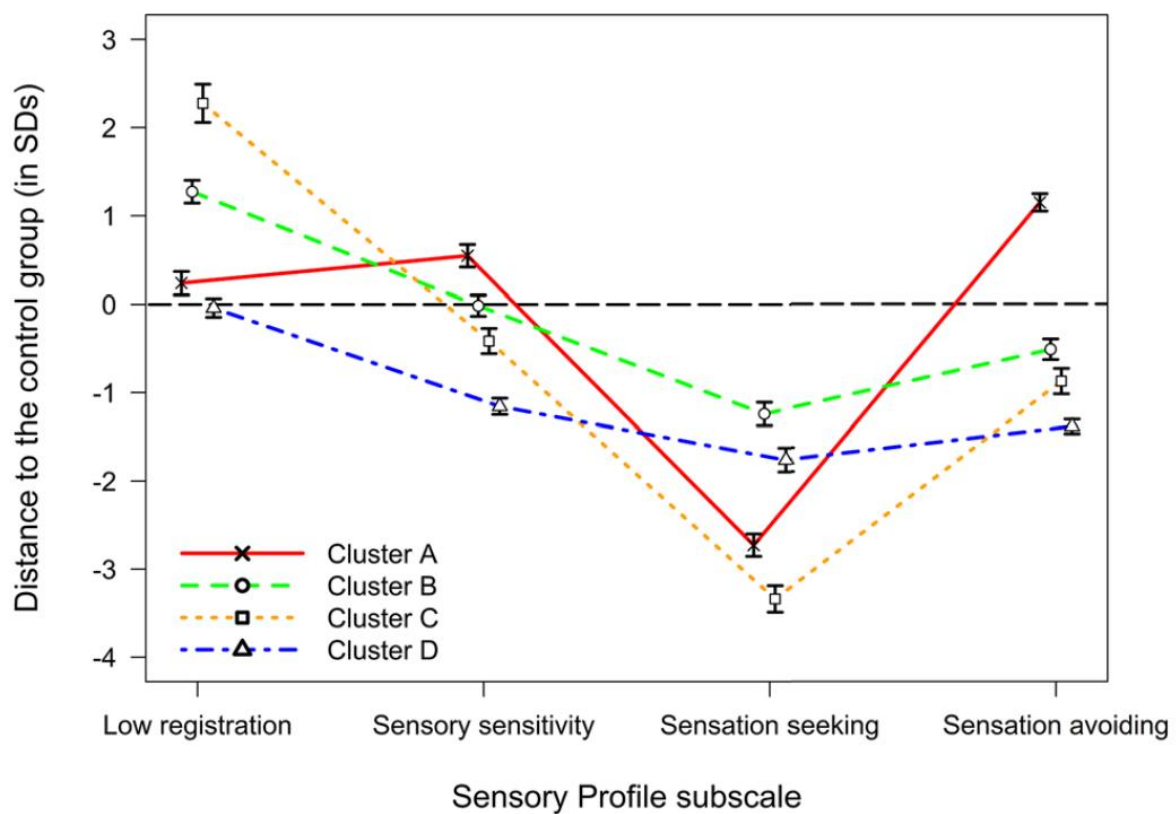


Figure 2. Standardized average scores on the Sensory Profile for the four clusters in reference to the control group. The horizontal dashed line represents the mean of the control group. The vertical axis represents the standardized distance between the mean of a cluster and the mean of the control group (ranging from 4 standard deviations below the control group to 3 standard deviations above the control group). Error bars represent standard errors of the mean.

Objective 2: Testing the Relationship Between Sensory and Behavioral Dysfunction

The second objective of our study was to determine how sensory processing abnormalities relate to behavioral dysfunction in patients with ASD. The correlations between subscales of the Sensory Profile and the EPOCAA and BDSI are summarized in Tables 3 and 4. The results confirmed that different sensory abnormalities were predictive of different patterns of behavioral dysfunction. For example, a high level of sensory sensitivity was predictive of isolation seeking, dysfunctions of social interactions, sensory-motor reactivity, reactivity to change, and reactivity to sensory stimuli; conversely, low registration was primarily associated with global hypoactivity and apathy, difficulty for initiating activities, disinterest and indifference, and self-aggression. Sensation avoiding was predictive of isolation seeking, deficits in social interactions and eye contacts, and excess reactivity to change and to sensory stimuli; whereas a high level of sensation seeking came with less isolation and less deficits in social interactions and eye contacts, but with more other-aggression, inappropriate social behaviors, irritability and emotional lability.

The final series of analyses examined whether patients in the different sensory clusters displayed different behavioral disorders. Results are synthesized in Tables 5 and 6 (see Online Information 1 for detailed descriptive statistics per cluster and per behavioral disorder subscale). The difference between clusters was statistically significant at the 0.05 level for all but four behavioral disorder subscales.

- Patients in Cluster A (over-sensitive) had a fairly typical level of impairment on most behavioral disorder subscales. These patients were mostly remarkable for a high level of isolation seeking, and for displaying less emotional lability than others. They were also relatively more autonomous than other clusters.
- Patients in Cluster B (under-sensitive) had a larger range of behavioral disorders than the over-sensitive patients of Cluster A. In particular, they displayed more emotional

disorders than other clusters, with high scores in irritability and aggressiveness, emotional disorders and anxiety, and expression of affectivity. They had more difficulties with relationships to others, with high scores in social behavioral disorders and in other-aggression; their behavior was more influenced by environmental stimuli ("environmental dependency"), and they displayed more dysfunctional use of everyday objects (such as exploratory or destructive behaviors). These patients were also remarkable for demonstrating a significantly higher level of self-aggression than other clusters.

- Patients in Cluster C (passive) were characterized by their unresponsive behavior, with high isolation, hypoactivity and apathy, disinterest and indifference, but also deficits in social interactions and eye contacts. They displayed less reactivity to change and to sensory stimuli than other clusters, as well as less emotional lability. Unsurprisingly, these patients demonstrated less other-aggression than others.
- Patients in Cluster D (balanced profile) had lower scores on self-stimulation and reactivity to sensory stimuli, confirming their overall milder sensory dysfunction. These patients also had the least behavioral disorders of all: they were either equally impaired or significantly less impaired than other clusters on virtually all subscales. The only exception was that these patients demonstrated a high level of other-aggression, presumably related to the fact that they were more active than other patients.

Discussion

Our results lead to three main conclusions concerning sensory processing abnormalities in low-functioning adults with ASD and their relationship with behavioral disorders. First, the data confirm that sensory abnormalities are highly prevalent in low-functioning adults with ASD, in line with prior studies (Degenne-Richard 2014; Leekam et al. 2007). The current study extends prior work in showing that sensory abnormalities can be reliably observed in a large

sample of adults with a wide range of ages, even when assessed with a widely-used scale such as Dunn's Sensory Profile. Second, our results provide evidence for clearly distinct sub-profiles of sensory dysfunction in low-functioning adults with ASD (see also Degenne-Richard 2014). Participants in our study could be split into four clusters: over-sensitive, under-sensitive, passive and balanced. Patients in the different clusters were similar in terms of age and gender, but they had different verbal levels and behavioral impairments, thus confirming the validity of the classification. Our results converge with prior studies that distinguished sub-profiles of sensory dysfunction in children with ASD based on under- and over-sensitivity and on the overall level of impairment (see Ausderau et al. 2014; Baker et al. 2008; Ben-Sasson et al. 2008; Liss et al. 2006). Third, sensory dysfunction appears related to behavioral dysfunction: responses on the various subscales of the Sensory Profile were predictive of impairments on the BDSI and EPOCAA, and critically, the four sub-profiles of sensory abnormalities were associated with different types of behavioral disorders. These results support the conclusions of prior studies showing an interplay between sensory dysfunction and behavioral disorders (Baker et al. 2008; Baranek et al. 1997b; Hilton et al. 2007; Kern et al. 2007b), and confirm that this relationship is also found in low-functioning adults. In sum, our results indicate that sensory abnormalities do play an important role in low-functioning adults with ASD.

These conclusions have significant implications for clinical care. Low-functioning adults with ASD are especially vulnerable to sensory dysfunction, because their low autonomy affords few strategies to cope with inappropriate stimuli. Both the prevalence of sensory abnormalities in our sample and their link with behavioral disorders vouch for the clinical relevance of systematically assessing sensory processing in this population. In particular, our results suggest that it may be beneficial to identify a patient's profile of sensory dysfunction, because understanding their specific pattern of sensory abnormalities may help to prevent and ultimately care for their behavioral disorders. This idea seems especially interesting in light of

the possibility that sensory symptoms may actually be one cause of behavioral disorders in ASD patients (e.g. Baker et al. 2008; Baranek et al. 1997b). For example, Kern et al. (2008) asserted: "It is possible that behavioral difficulties (i.e., willful acts of noncompliance) in autism may not be behavioral at [their] core, but a manifestation of difficulties in sensory modulation, e.g., discomfort from light, sounds, somatosensory information, or confusion from proprioceptive or vestibular information" (pp. 8–9).

Sensory symptoms may also impede adaptive behaviors, as well as social skills (see Pfeiffer et al. 2005). For example, some authors have suggested that sensation avoiding leads to resisting social interactions whereas sensation seeking leads to initiating unconventional or inappropriate interactions (see Kern et al. 2007a), an idea that is backed by the present results. As a consequence, sensory-based intervention strategies may help counteract the emergence of maladaptive behaviors and could be an effective way to care for low-functioning adults with ASD (Lane et al. 2010). A number of sensory-based strategies for care have been developed over the years and could be used for this purpose (for a review, see Gal et al. 2007). These include adapting the patients' environment to promote stimulations to which they are likely to respond positively, teaching them basic self-employed strategies designed to cope with unpleasant stimuli, and directly confronting them with stimulations tailored to their needs (Gal et al. 2007). Of course, the present results indicate that sensory-based care strategies should take into account the specific profile of sensory dysfunction of each patient: a patient from the "passive" cluster might benefit from increased stimulation, but a patient from the "over-sensitive" cluster certainly would not.

How would this all play out in practice? Our results suggest at least one possible example. Under-sensitive patients displayed a range of specific behavioral disorders, prominently including self-aggression, one of the most disabling symptoms of ASD from a clinical standpoint. Indeed, patients belonging to the "under-sensitive" cluster demonstrated

significantly higher self-aggression than all other patients in the sample (see Tordjman et al. 1999, for a similar result). The idea that sensory dysfunction may be at the root of behavioral dysfunction suggests that self-aggression behaviors may actually be caused by sensory under-sensitivity, possibly because under-sensitive patients use self-aggression as a form of self-stimulation. If that is the case, self-aggression behaviors might be discouraged by offering increased sensory inputs to these patients.

Several methodological concerns can be discussed in light of the present findings. The existence of different profiles of sensory dysfunction suggests that researchers should avoid comparing average sensory scores across samples: averaging the scores of participants of ASD will necessarily confound opposite patterns of deficits. For example, the average sensory sensitivity of patients with ASD was very close to the average of control participants in the present study (with an effect size of $\eta^2 = .02$ for the difference), but this average is clearly misleading: one cluster of ASD patients was firmly over-sensitive whereas another was under-sensitive (see Fig. 2). Another important point is that both research and clinical practice in the field of ASD would benefit from the use of dedicated measurement tools for sensory abnormalities. Although Dunn's Sensory Profile has been used in a very large share of studies on ASD (Gal et al. 2007), it was created for non-clinical participants and demonstrates relatively poor psychometric qualities in this population (Gonthier et al. in preparation). A few scales have been specifically designed to assess sensory processing in ASD patients (such as the Sensory Profile Checklist Revised; Bogdashina 2005) and may be much more suited to the specificities of autism. Direct observation may also be a very good way to assess sensory processing. This approach can be fruitfully used in research works, especially when complemented with experimental studies (see Iarocci and McDonald 2006), and the development of standardized observation scales (e.g. Tavassoli et al. 2016) may prove very helpful for identifying sensory problems and selecting suitable interventions in clinical settings.

Importantly, using better methods to assess sensory abnormalities would also allow for more fine-grained analyses of the results. For example, the clustering of participants in the present study seemed to be driven partly by their overall level of activity and the severity of their autistic symptoms, rather than by specificities in sensory processing: patients from cluster C ("passive") had a lower level of activity and more severe symptoms overall, whereas patients in cluster D ("balanced") were more active and suffered from less severe autistic and behavioral symptoms. These differences may or may not reflect underlying differences in sensory processing. Future studies should also strive to study intra-individual variability in sensory abnormalities: for example, anecdotal evidence indicates that some patients can be under-responsive to certain stimuli and over-responsive to others. This point suggests that the clusters of sensory impairment found in the present study may not reflect homogeneous profiles. As a consequence, strategies for care will necessarily have to take into account the idiosyncrasies of each patient.

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Author Contributions

CG reviewed the literature, analyzed and interpreted the data and wrote the manuscript; LL participated in the design and coordination of the study and performed the data collection; MB participated in the design, supervised the study and helped to draft the manuscript.

Conflicts of interest

The authors declare that they have no conflict of interest.

Ethical Approval

The study was approved by the research ethics committee of the University of Savoy, in compliance with French regulations (approval date: January 29 2012). All procedures were in accordance with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed Consent

All relevant information about the purpose and the conditions of the study was communicated to the heads of the care centers, to the participating health and social workers, and to the families of the participants. Informed consent was provided by the legal guardians of all participants with autism prior to their inclusion in the study. The research was purely non-interventional: for adults with ASD, the questionnaires used the other-report method, and the patients themselves were never approached during the course of the study. The data were fully anonymized, and no information allowing the identification of participants was collected at any point.

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