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ORIGINAL PAPER

The Impact of Atypical Sensory Processing on Adaptive Functioning and Maladaptive Behaviors in Autism Spectrum Disorder During Childhood: Results From the ELENA Cohort

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Abstract
Atypical sensory processing is common in autism spectrum disorders (ASD), but their role in adaptive difficulties and problem behaviors is poorly understood. Our aim was to determine the prevalence and type of atypical sensory processing in children with ASD and investigate its impact on their adaptive functioning and maladaptive behaviors. We studied a subsample of 197 children rigorously diagnosed with ASD from the ELENA cohort. Children were divided into atypical and typical sensory processing groups and several independent variables were compared, including adaptive functioning and maladaptive behaviors. Overall, 86.8% of the children had at least one atypical sensory pattern and all sensory modalities were disturbed. Atypical sensory processing explained a significant part of the variance of behavioral problems.

Keywords Autism spectrum disorders · Sensory processing · Adaptive functioning · Maladaptive behaviors · Children

Introduction

Autism spectrum disorders (ASD) are a heterogeneous group of neurodevelopmental conditions characterized by deficits in social interactions and communication, along with unusually repetitive behavior and restrictive interests (American Psychiatric Association 2000). Recent estimates suggest that the prevalence of ASD is as great as one in 68 children (Christensen et al. 2016). Atypical sensory processing (SP) is frequently reported in ASD and is thus an ASD diagnostic criterion (DSM 5; American Psychiatric Association 2000).

Sensory processing allows the selection, organization, and association of various types of sensory information from the environment help to adapt human behavior. Impairment

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of SP can lead to adaptive difficulties (Ayres 1979). The Sensory Profile is the most common tool used to measure SP. This questionnaire, completed by parents, has been validated in a sample of children with ASD (Dunn 1999). The long version of the Sensory Profile presents the results in terms of four sensory profiles for each sensory modality. According to Dunn (1999), sensory responses to sensory stimulation depend on neurological thresholds (high or low) and individual strategies (active or passive). If the threshold of detection is high and the response active, the sensory profile is defined as *sensory seeking* (e.g. children are always to get more sensory input and stay alert) whereas it is defined as *low registration* if the response is passive (e.g. children will miss sensory cues than others notice easily). In contrast, if the threshold of detection is low and the response active, the Sensory Profile is defined as *avoiding sensation* (e.g. children will move away from activities or may choose to work alone), whereas it is defined as *sensory* if the response is passive *sensitivity* (e.g. children will put their hands over their ears.). All sensory modalities (visual, auditory, tactile, olfactory, taste, vestibular, and proprioceptive) can be affected.

Atypical SP is common in ASD (American Psychiatric Association 2000). However, its prevalence depends on the study; a meta-analysis of 14 studies reported a range from 45 to 95% (Ben-Sasson et al. 2009). Such variability could be explained both by the discrepancy of the measures used in the available studies and the limited size of their samples. Some of these studies have shown that sensory symptoms in ASD are stable over time, independent of cognitive level and autistic severity (Ausderau et al. 2014; Perez Repetto et al. 2017), whereas others found that sensory symptoms decrease over time (Baranek et al. 2013). The meta-analysis mentioned earlier shows that SP is associated with chronological age, atypical SP being more prevalent in school-age than preschool-age children (Ben-Sasson et al. 2009). Studies have also found that atypical SP correlates with mental age (Ben-Sasson et al. 2013) or intellectual quotient (Ausderau et al. 2014; Baranek et al. 2013). However, several other studies did not confirm the association between SP and intellectual functioning (O'Donnell et al. 2012) and reported atypical SP by individuals with Asperger syndrome or high functioning autism (Pfeiffer et al. 2005). Moreover, it has been suggested that atypical SP in ASD can appear during childhood (Germani et al. 2014), earlier than social and communication deficits (Sacrey et al. 2015), and can be a major cause of adaptive deficits.

Adaptive functioning is defined as every conceptual, social, and practical skill that has been learned and is performed in everyday life (American Psychiatric Association 2000). Adaptive skills are often measured in practice by the Vineland Adaptive Behavior Scale, which is a standardized instrument based on a semi-directed interview with family members or caregivers, focusing on communication, socialization, and daily life (VABS: Sparrow et al. 2005). Adaptive functioning deficits are not only more prevalent in children with ASD than typical children but also more prevalent than in children with other neurodevelopmental conditions (Mouga et al. 2015). Moreover, adaptive functioning in ASD appears to be more predictive of the outcome in children than intellectual functioning (Farley et al. 2009).

From our systematic review, the synthesis of the literature shows that atypical patterns of SP are associated with adaptive functioning in children with ASD (Dellapiazza et al. 2018). Children with sensory-seeking or sensory-sensitivity profiles show more deficits in communication skills measured with the VABS (Lane et al. 2010; Tomchek et al. 2015). It was suggested in one study that SP in preschoolers with ASD, particularly sensory seeking (Watson et al. 2011), may influence their receptive and expressive language skills (Tomchek et al. 2015). However, these results cannot be generalized or considered to be specific to ASD, as they could not be replicated when adjusting for mental age (McCormick et al. 2016). In addition, a recent review suggested that atypical SP plays a role in the social skills of ASD children (Thye et al. 2018). In one study, atypical SP was found to be associated with joint attention, which is a specific aspect of socialization skills (Baranek et al. 2013). However, other studies using the Vineland Scale to assess socialization skills were unable to replicate these results (Baker et al. 2008; Lane et al. 2010; McCormick et al. 2016). Concerning other dimensions of adaptive skills, some studies, mainly performed on small samples, have suggested that atypical SP reduces autonomy skills. One study, performed on a sample of 33 preschoolers with ASD, found their adaptive functioning to be lower if they had a sensory-avoiding profile, regardless of their intellectual functioning (Jasmin et al. 2009). Another study, of 22 children with ASD, found a link between the global score of the short Sensory Profile and Vineland personal autonomy skills (Baker et al. 2008). Also, atypical SP have also been found to have a negative impact on familial autonomy skills (Schaaf et al. 2011). A study of a large sample of 144 children with ASD also showed that sensory seeking has a negative effect on autonomy skills (Liss et al. 2006). However, three other studies were unable to replicate these results, but they used the short version of the Sensory Profile, which is less informative than the long form (Lane et al. 2010; McCormick et al. 2016; Tomchek et al. 2015). Another recent study did not find any association between SP and adaptive behaviors when the results were adjusted for the children's verbal mental age (McCormick et al. 2016).

Atypical SP frequently co-occurs with maladaptive behaviors (or problem behaviors) in children with ASD (Baker et al. 2008; Lane et al. 2010; McCormick et al. 2016). Atypical SP has been shown to be a risk factor for self-injurious (Duerden et al. 2012) and repetitive and restricted behaviors in ASD (Lidstone et al. 2014; Renzo et al. 2017). Maladaptive behaviors include irritability, lethargy, hyperactivity, and repetitive behaviors (Aman et al. 1985). Such behaviors have been reported to be more prevalent in children with sensory-seeking difficulties (Baker et al. 2008; Lane et al. 2010). However, this result needs to be interpreted with caution, as the studies did not specifically assess maladaptive behaviors (but with the VABS supplementary section) nor specify the children's IQ, although this can influence behavioral problems (Matson and Shoemaker 2009). Further large-scale studies will be necessary to address the discrepancies of the findings concerning the role of atypical SP on adaptive functioning and maladaptive behaviors in ASD.

Given the ambiguity of the results from past research that has studied SP in ASD, the purpose of the current study is twofold. Our first aim was to assess the prevalence and type of SP in a large sample of children with a rigorous diagnosis of ASD using a standardized and validated tool. The second aim was to investigate the impact of atypical sensory processing on adaptive functioning and maladaptive behaviors in this sample. We anticipate that atypical SP in our sample would be frequently occurring, mainly represented by a sensory-seeking profile, and that atypical SP would be a risk factor for adaptive deficits and maladaptive behaviors.

Methods

Participants

The participants consisted of a subgroup recruited from a large cohort of children diagnosed with ASD, the ELENA cohort (study protocol under submission; http://elena-cohorte.org/), an open, multicenter, longitudinal, prospective study. Overall, 700 children have been recruited for the ELENA cohort after parental consent at the time this paper was written (in October 2018). The inclusion criteria for the ELENA Cohort were: children from 2 to 17 years of age with a diagnosis of ASD, established by a multidisciplinary team using a standardized process, including the Autism Diagnostic Observation Schedule 2 (ADOS 2: Lord et al. 2012), Autism Diagnostic Interview-Revised (ADI-R: Le Couteur et al. 2003), administered by licensed and trained psychologists, a parental interview concerning the child's adaptive functioning using the VABS II, and direct psychological examinations to assess IQ. The only exclusion criterion was the inability of the parents to speak or read French. For the ELENA cohort, the parents were requested to complete web-questionnaires, including a standardized questionnaire (concerning the child's health problems, school, and intervention), the long form of the Sensory Profile, and the Aberrant Behavior Checklist.

For the current study, we used a subset of the ELENA data for 197 children from 3 to 11 years of age, in accordance with the age boundaries of the Sensory Profile used to assess the children's SP (Dunn 1999). We excluded children who were blind and/or had auditory deficits, and we also excluded the two children diagnosed with comorbid Down syndrome or Phelan-Mc Dermid syndrome as far as they commonly exhibit atypical sensory reactions and low adaptive level (Bruni et al. 2010; Mieses et al. 2016).

Measures

Sensory processing was assessed using the long-form Sensory Profile (Dunn 1999). This questionnaire of 125 items asks parents about sensory responses in the daily life of their children aged from 3 to 11 years. The results are given according to four quadrants that summarize behavioral responses to sensory stimulation in four profiles: sensory seeking, low registration, sensory avoiding, and sensory sensitivity. The results can also be presented according to each sensory modality (auditory, visual, vestibular, oral, touch, and multisensory). Their frequency is recorded on a five-point Likert scale and the scores are interpreted as follows: « typical performance » (less than 1 standard deviation (SD)), « probable difference » (between 1 and 2 SD), and « definite difference » (more than 2 SD). Lower Sensory Profile scores imply greater SP difficulties. Internal consistency of the Sensory Profile ranged from 0.70 to 0.90 and internal validity correlations from 0.25 to 0.76 (Dunn 1999).

Adaptive functioning was assessed using the Vineland Adaptive Behavior Scales second edition (VABS II: Sparrow et al. 2005). This standardized caregiver interview of 297 items, measures adaptive behaviors from childhood to adulthood in the subdomains of communication, daily living skills, and socialization. In our study, we used the standard scores of the three subdomains. The reliability of the VABS II for each domain was excellent ($\alpha = 0.80$) and the intra-class coefficient of the test/re-test 0.89.

Maladaptive behaviors were assessed using the Aberrant Behavior Checklist (ABC: Aman et al. 1985), a 58-item scale concerning maladaptive or problem behaviors, with each item scored from 0 (= no problem) to 3 (= severe problem). The scale includes five factors: (I) irritability, agitation, crying; (II) lethargy, social withdrawal; (III) stereotypic behavior; (IV) hyperactivity, noncompliance; and (V) inappropriate speech. The ABC showed good internal consistency among subscales ($\alpha = 0.91$), an excellent test–retest reliability of 0.98, and an acceptable interrater reliability of 0.63.

ASD severity was established using the Autism Diagnostic Observation Schedule second version (ADOS-2: Lord et al. 2012), a semi-structured behavioral observation protocol to assess ASD symptomatology. This scale is composed of 25 to 30 items across symptom domains: social interaction, communication, repetitive and stereotyped behaviors, and play. The internal consistency was good (α = 0.50–0.92), the test–retest reliability acceptable (0.64–0.88), and the interrater reliability excellent (0.79–0.98). For this study, we used the Calibrate Severity Score (CSS), ranging from 1 to 10.

The *"Best-Estimate" intellectual functioning level* was established from several psychometric scales, depending on the age and developmental level of each participant (Howlin et al. 2014). The intellectual quotient (IQ) or, when not available, developmental quotient (DQ) (developmental age score/chronological age * 100), were used for analysis. IQ was estimated from the WISC-IV, WISC-V, WPPSI-R (n = 68); KABC (n = 3); or BECS (n = 8). The DQ was estimated from the PEP-R (n = 64) and Brunet-Lezine scales (n = 15).

Data Analyses

Descriptive statistics were calculated to characterize the sample and study variables. The scores from the Sensory Profile quadrants and sections were used to define two groups of children based on their SP functioning: a typical group (scores under 1 SD from the norm) and an atypical group (scores over 1 to 2 SD from the norm). First, the prevalence of typical and atypical sensory scores was calculated. Then, intergroup (atypical/typical) comparisons based on gender, chronological age, best-estimate IQ, and autism severity, Vineland, and ABC scores, were conducted using Student's T test for parametric variables and the Mann–Whitney test for non-parametric variables. Finally, multivariate analysis of covariance (ANCOVA) was performed on dependent variables (Vineland II and ABC scores) significantly associated with atypical SP (quadrants) in the univariate analysis. Age and IQ were entered as covariates. Results were considered to be statistically significant for $p < 0.05$. SAS 9.3 was used to perform the statistical analyses.

Results

Participants

Overall, there were 197 participants aged from 3 to 10 years and 11 months (mean age 5.7 ± 2.2 years). Their clinical characteristics are shown in Table 1. Approximately 64% were aged below 6 years. Approximately 52% had no intellectual disability (best-estimate IQ over 70), 19.6% a mild intellectual disability, 12.7% a moderate intellectual disability, and 15.8% a severe intellectual disability.

A total of 145 mothers (mean age 36.7 ± 5.5 years) and 143 fathers (mean age 40.2 ± 7.0 years) participated in the study. Overall, 38.4% of the mothers and 48.7% of the fathers had a high school-level education and 61.5% of the mothers and 51.3% of fathers had a higher than a high school-level education.

Prevalence of Atypical Sensory Processing

In the whole sample, mean Sensory Profile scores were in the atypical range for every quadrant and section, except for visual processing (Table 2). The frequency of typical and atypical scores for the quadrants and sections of the Sensory Profile are described in Table 2. Most of the children (86.8%, N = 171) had Sensory Profile scores in the atypical range for at least one quadrant. Approximately one quarter (24.4%, N = 48) had SP scores in the atypical range for all four quadrants. Only 13.2% of the children had SP scores in the typical range for all quadrants (N = 26). The prevalence of the children with SP scores in the atypical range was particularly high when considering by quadrant (78.1% sensation avoiding, 68.2% sensation seeking, 64.5% sensation sensitivity, and 53.3% low registration) and remained high when considering by section (58.3% multisensory processing, 56.9% touch processing, 53.0% auditory processing, 45.3% oral processing, and 32.6% vestibular processing).

Intergroup comparisons of Sensory Profile quadrants showed no significant differences based on age, gender, cognitive level, or ADOS scores (all $p > 0.05$), except for the sensory-avoiding group, for which children in the atypical group were older than those in the typical group (6.0 ± 2.3 vs 4.9 ± 1.7 years, $p < 0.05$). Intergroup comparisons of SP sections showed no significant differences based on gender (all $p < 0.05$). However, there were significant differences depending on age and cognitive level. For auditory processing, children in the atypical group were older than those in the typical group (6.2 ± 2.4 vs 5.1 ± 1.9 years, $p < 0.05$). For visual processing, children in the atypical group were also older than those in the typical group (6.5 ± 2.4 vs 5.4 ± 2.1 years; $p < 0.05$). However, for oral processing, children in the atypical group were younger than those in the typical group (5.3 ± 2.2 vs 6.1 ± 2.3, $p < 0.05$). For visual processing, the cognitive level was higher in the atypical group than in the typical group (81.7 ± 25.9 vs 67.8 ± 25.7, $p < 0.05$), whereas for oral processing, children in the atypical group had a lower cognitive level than those in the typical group (63.3 ± 26.0 vs 81.3 ± 24.8, $p < 0.05$).

Table 1 Clinical characteristics of the children

Measure	*N*	%	
Gender			
Male	164	83.2	
Female	33	16.8	
Diagnosis			
F84.0 childhood autism	152	77.2	
F84.1 atypical autism	6	3.0	
F84.5 Asperger syndrome	32	16.2	
F84.8—other F84.9 unspecified	7	3.6	
Prematurity			
Yes, before 37 weeks	21	17.8	
No	97	82.2	
	N	Mean	SD
Best estimate IQ	158	71.6	26.8
Severity score ADOS	175	6.9	2.0
Vineland 2	192		
Communication		69.8	16.5
Daily living skills		73.0	12.6
Socialization		68.1	11.4
Aberrant Behavior Checklist	154		
Irritability		35.3	18.8
Lethargy		28.2	17.6
Stereotypy		34.2	22.1
Hyperactivity		47.9	22.3

Table 2 Description and comparison of typical and atypical Sensory Profile scores

	Total sample			Typical group		Atypical group		p value*
	N	Mean (SD)	Range of typical performance	N	Mean (SD)	N	Mean (SD)	
Quadrant								
Low registration	180	**60.2 (9.7)**	72–64	84	**68.1 (2.9)**	96	**53.2 (8.2)**	**< 0.0001**
Sensation seeking	167	**92.8 (17.2)**	123–103	53	**111.45 (5.8)**	114	**84.1 (13.4)**	**< 0.0001**
Sensory sensitivity	158	**75.8 (11.7)**	94–81	56	**88.1 (4.4)**	102	**69.0 (8.5)**	**< 0.0001**
Sensation avoiding	155	**100.0 (15.1)**	133–113	34	**118.9 (5.3)**	121	**94.6 (12.4)**	**< 0.0001**
Section								
Auditory processing	166	**26.7 (6.0)**	40–30	78	**31.5 (3.1)**	88	**21.9 (3.9)**	**< 0.0001**
Visual processing	170	34.2 (6.3)	45–32	122	**37.3 (3.4)**	48	**26.1 (4.6)**	**< 0.0001**
Vestibular processing	181	**44.2 (5.8)**	55–48	122	**47.37 (3.3)**	59	**37.6 (3.8)**	**< 0.0001**
Touch processing	179	**68.6 (10.7)**	90–73	79	**78.5 (4.8)**	100	**60.8 (6.8)**	**< 0.0001**
Multisensory processing	192	**25.0 (4.4)**	35–27	82	**29.3 (2.0)**	110	**22.4 (3.3)**	**< 0.0001**
Oral sensory processing	179	**45.0 (10.3)**	60–46	98	**53.0 (4.5)**	81	**35.3 (6.3)**	**< 0.0001**

Significant results appear in bold

*p value of comparisons between typical and atypical group

Table 3 Comparison of the VABS scores between typical and atypical SP groups, based on quadrants

	Low registration		Sensory seeking		Sensory sensitivity		Sensory avoiding	
	Typical N = 83	Atypical N = 92	Typical N = 51	Atypical N = 111	Typical N = 55	Atypical N = 99	Typical N = 34	Atypical N = 117
	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)
Vineland								
Communication	70.3 (17.6)	70.0 (15.7)	**74.0 (19.1)**	**68.0 (15.2)***	69.7 (18.2)	69.0 (14.9)	**63.9 (18.2)**	**73.3 (15.6)****
Daily living skills	75.4 (13.1)	72.0 (11.5)	**76.4 (12.8)**	**71.9 (12.1)***	74.1 (12.0)	71.2 (12.3)	74.2 (13.4)	73.4 (12.5)
Socialization	68.0 (12.6)	69.0 (9.4)	**71.5 (11.5)**	**67.1 (11.5)***	68.8 (11.8)	67.7 (10.4)	67.8 (12.3)	69.2 (10.9)

Significant results appear in bold

*Significant at $p < .05$ level, **significant at $p < 0.01$ level

Group Comparisons Based on Adaptive Behavior (VABS)

We compared the mean scores for each VABS domain between the atypical and typical SP groups. The results are summarized in Table 3.

Based on SP quadrants (see Table 3), children with atypical sensory avoiding had significantly higher VABS communication scores than children with typical sensory avoiding, whereas children with atypical sensory seeking had lower adaptive functioning than those with typical reactions, regardless of the Vineland domain. Based on SP sections, children in the atypical group for tactile processing had lower socialization scores than those in the typical group (66.7 ± 10.8 vs 70.6 ± 10.9, $p < 0.05$). For oral processing, children in the atypical group had lower scores in each VABS domain: communication (66.3 ± 16.7 vs 73.6 ± 15.6, $p < 0.01$), autonomy (69.4 ± 13.5 vs 76.0 ± 11.0, $p < 0.01$), and socialization (65.1 ± 11.9 vs 70.9 ± 10.2, $p < 0.01$). There were no intergroup differences for VABS scores for auditory, visual, vestibular, or multisensory processing.

Multivariate Analysis of Covariance on Adaptive Behaviors (VABS)

The results of the multivariate ANCOVA model explained 62% of the variance of the communication scores, 34% of the socialization scores, and 34% of the autonomy scores. There was no effect of sensory quadrant on the Vineland scores (all $p > 0.05$), but there was a positive effect of the best-estimate IQ on the Vineland domains. For each one-unit increase of the best-estimate IQ score there was an increase in the communication score ($\beta = 0.5$, $p < 0.0001$), as well as that for socialization ($\beta = 0.3$, $p < 0.01$) and autonomy ($\beta = 0.3$, $p < 0.0001$). Scores for socialization ($\beta = -1.1$, $p < 0.01$) and autonomy ($\beta = -0.98$, $p < 0.05$) decreased with age.

Intergroup Comparisons for Maladaptive Behaviors (ABC)

We performed intergroup comparisons for ABC scores based on SP quadrants (Table 4) and SP sections (Table 5).

Based on SP quadrants (see Table 4), children categorized as atypical had significantly more maladaptive behaviors, regardless of the ABC domain considered, except for

Table 4 Intergroup comparisons of ABC scores based on SP quadrants

	Low registration		Sensory seeking		Sensory sensitivity		Sensory avoiding	
	Typical N = 58	Atypical N = 81	Typical N = 43	Atypical N = 85	Typical N = 43	Atypical N = 76	Typical N = 26	Atypical N = 92
	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)
ABC								
Irritability	**31.3 (17.3)**	**39.0 (19.0)***	**28.5 (17.8)**	**39.7 (19.3)****	**29.1 (17.5)**	**39.2 (19.5)***	**27.2 (20.1)**	**37.2 (18.5)***
Lethargy	**22.3 (16.8)**	**32.8 (16.7)*****	**21.7 (15.7)**	**32.4 (17.8)****	**21.5 (17.6)**	**31.9 (17.9)****	**19.8 (16.1)**	**30.6 (16.8)****
Stereotypy	**26.2 (19.6)**	**39.2 (21.3)*****	**27.1 (22.4)**	**37.8 (21.4)****	**26.6 (23.2)**	**39.2 (21.7)****	**22.3 (17.1)**	**36.2 (21.0)****
Hyperactivity	44.7 (21.1)	51.4 (22.6)	**31.4 (19.3)**	**56.6 (18.7)*****	**39.9 (21.2)**	**53.2 (22.4)****	41.5 (22.1)	49.1 (22.1)

Significant results appear in bold

*Significant at $p < 0.05$ level, **significant at $p < 0.01$ level, ***significant at $p < 0.001$ level

Table 5 Intergroup comparisons of ABC scores based on Sensory Profile sections

	Auditory		Visual		Vestibular		Touch		Multisensory		Oral	
	Typical N = 57	Atypical N = 68	Typical N = 40	Atypical N = 90	Typical N = 96	Atypical N = 43	Typical N = 75	Atypical N = 62	Typical N = 61	Atypical N = 88	Typical N = 78	Atypical N = 61
	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)
ABC												
Irritability	32.4 (19.3)	39.5 (18.5)*	31.0 (18.4)	45.1 (18.5)***	34.6 (19.8)	38.6 (18.2)	28.1 (17.5)	40.1 (17.8)***	27.9 (17.0)	39.9 (18.2)***	31.6 (19.2)	39.3 (17.7)*
Lethargy	22.4 (15.8)	33.3 (18.7)**	25.6 (15.3)	32.2 (20.9)	27.1 (18.3)	32.0 (16.0)	21.1 (15.5)	34.3 (17.0)***	22.3 (16.8)	32.5 (17.9)***	26.0 (17.5)	31.9 (17.6)*
Stereotypy	27.9 (20.4)	40.5 (22.5)**	31.0 (20.6)	39.6 (23.8)	31.1 (22.0)	40.6 (22.2)**	24.5 (20.2)	42.1 (21.0)***	25.6 (19.0)	39.1 (21.8)***	30.2 (21.2)	40.3 (22.6)*
Hyperactivity	43.1 (22.4)	54.5 (21.0)**	46.0 (22.9)	51.4 (20.3)	44.5 (22.3)	56.3 (21.3)**	37.1 (19.5)	57.5 (20.3)***	37.6 (20.6)	54.1 (20.7)***	42.1 (23.6)	53.7 (19.4)**

Significant results appear in bold

*Significant at p < 0.05 level, **significant at p < 0.01 level, ***significant at p < 0.001 level

hyperactivity, which did not differ between typical and atypical groups for low registration and sensory avoiding.

Based on Sensory Profile sections (see Table 5), children with atypical reactions for the auditory, touch, multisensory or oral dimensions had significantly more maladaptive behaviors than those in the typical group. Also, children with atypical visual reactions had significantly higher irritability scores and those with atypical vestibular reactions had more stereotypies and hyperactivity.

Multivariate Analysis of Covariance on Maladaptive Behavior (ABC)

We performed an ANCOVA analysis to study the variance of atypical scores, based on quadrants, to explain the ABC scores (Table 6). ANCOVA explained 16% of the variance of the irritability score, 24% of that for lethargy, 39% of that for stereotypical behaviors, and 35% for that of hyperactivity. In addition, the irritability, lethargy, stereotypy, and hyperactivity scores decreased when the best-estimate IQ increased (*all* $p < 0.05$) and the stereotypy scores increased when the children's age increased ($\beta = 2.9$, $p < 0.01$). There was an 18.8-point increase in the irritability score for each once-point increase in the sensation-avoiding score. The lethargy score was higher ($\beta = 10.0$, $p < 0.05$) when the sensory-sensitivity scores were in the atypical range. The stereotypy scores were higher when the scores were atypical for low registration ($\beta = 12.3$, $p < 0.01$) and sensory sensitivity ($\beta = 13.2$, $p < 0.01$). Finally, the hyperactivity scores were higher when the scores for sensation seeking ($\beta = 16.0$, $p < 0.01$) and sensory sensitivity ($\beta = 11.3$, $p < 0.05$) were in the atypical range.

Discussion

We assessed the prevalence and types of SP and their links with adaptive skills and maladaptive behaviors in a large sample of children with ASD (ELENA-Cohort). Regarding atypical SP, our study reveals that is extremely common in children with ASD, especially sensation avoiding and sensation seeking. Gender, age, IQ, and autistic severity did not influence these processes. Atypical SP was strongly associated with maladaptive behaviors and explained a large proportion of them. In addition, the sensation-seeking profile was associated with lower Vineland adaptive scores.

Relative to typical children, we detected that children with ASD demonstrated atypical responses to sensory stimulation, including hypo-and hyper-reactivity, in accordance with the literature (Ashburner et al. 2008; Ben-Sasson et al. 2009; Tomchek et al. 2015). Most of the children (86.8%) showed atypical SP in at least one quadrant of the Sensory Profile, whereas this prevalence ranges from 45 to 95% in

Table 6 ANCOVA analysis (adjusted for age and best-estimate IQ) of ABC scores by atypical profile for SP sensory quadrants (selection forward with the best AIC)

	ABC			
	Irritability Beta (SD)	Lethargy Beta (SD)	Stereotypy Beta (SD)	Hyperactivity Beta (SD)
Intercept	34.9 (7.7)***	18.25 (7.2)**	25.3 (7.7)**	48.1 (7.2)***
Age	–	–	2.9 (1.1)**	–
Best estimate IQ	−0.2 (0.09)*	−0.17 (0.08)*	−0.3 (0.1) **	−0.27 (0.08)**
Low registration (dich)	–	–	12.3 (4.9)**	–
Sensation seeking (dich)	–	–	–	16.0 (4.7)**
Sensory sensitivity (dich)	–	10.0 (4.7)*	13.2 (5.1)**	11.3 (4.8)*
Sensation avoiding (dich)	18.8 (5.8)**	–	–	–
R^2 total	0.16	0.24	0.39	0.35

Dich: atypical vs. typical; Dashes indicate that variable not entry in the model

*Significant at $p<0.05$ level, **significant at $p<0.01$ level, ***significant at $p<0.0.0001$

the literature (Ben-Sasson et al. 2009). The most prevalent sensory profile found is sensory avoiding, whereas it was sensory seeking in the review mentioned earlier.

Regarding sensory modalities, we observed a high rate of atypical SP for oral, tactile, auditory, and vestibular modalities. The highest rate of atypical processing was identified for multisensory processing in 58.3% of the sample, suggesting that deficits for each modality disturbed the integration of multimodal information (Baum et al. 2015). Surprisingly, children with ASD showed similar visual processing to that of typical children, as reported in another study (Little et al. 2018). However, atypical visual processing in ASD is frequently reported by clinicians. One reason for this discrepancy may be that atypical visual behaviors queried in the Sensory Profile are difficult to ascertain by parents.

In terms of maladaptive behaviors, we observed a strong association with atypical SP, in accordance with the literature (Baker et al. 2008; Lane et al. 2010; Nieto et al. 2017). In our sample, atypical SP was particularly related to behavioral problems, whereas intellectual level is thought to be the significant predictor (Hartley et al. 2008). More precisely, irritability was related to avoiding-sensation behaviors and hyperactivity by sensory-seeking and sensory-sensitivity behaviors. We showed that atypical SP explained a part of maladaptive behaviors (from 16% until 39% of explained variance) when age and intellectual level are controlled. We can hypothesize that other variables, which were not taking into account in our analysis, might be related to maladaptive behaviour, such as ASD symptom severity or comorbid gastrointestinal and sleep disorders (Rattaz et al. 2018). These findings are particularly important, as the quality of life of the families (McStay et al. 2014) and maternal stress are both influenced by behavioral problems of the children (Nieto et al. 2017).

In our sample, atypical SP was not associated with age, gender, cognitive level, or autistic severity, however there is also no consensus in the literature (for a review see: Hazen et al. 2014). Atypical SP can be found in many neurodevelopmental disorders, such as ADHD (Little et al. 2018) or intellectual disabilities (Ausderau et al. 2014). Our results underline that atypical SP are particularly generalized in ASD, regardless of the child's chronological age (Ben-Sasson et al. 2009), justifying their use as a core diagnostic criterion (American Psychiatric Association 2000). Consistent with previous studies, we observed that children with higher hyper-responsiveness had significantly greater repetitive and restricted behaviors (Boyd et al. 2010; Gal et al. 2010). This relationship has also been found in children with high-functioning autism (Chen et al. 2009). These findings can be interpreted in light of the hypothesis that stereotypies regulate atypical SP (Joosten and Bundy 2010).

Our study reveals that children exhibiting sensory seeking had lower adaptive scores for all Vineland domains and more maladaptive behaviors. These results are consistent with previous findings highlighting that children with ASD seeking sensory stimulation had lower socialization skills and were less receptive to their social environment (Tomchek et al. 2015). This association between SP and socialization skills appears to occur very early in childhood, as children with ASD display more atypical sensory seeking with their siblings at 18 months. Moreover, the level of atypical sensory seeking in early childhood is predictive of their later socialization skills (Damiano-Goodwin et al. 2018). Minimizing the impact of the sensory-seeking strategy on adaptive development requires early intervention and adaptation of the environment.

Regarding the significant link between atypical SP and hyperactivity, it could explain the elevated prevalence of hyperactivity in individuals with ASD (Murray 2010). Indeed, SP difficulties are related and predict ADHD symptoms in ASD (Ashburner et al. 2008; Sanz-Cervera et al. 2015). Moreover, ASD teenagers frequently report that problems in SP reduce their concentration at school (Howe and Stagg 2016), affecting academic performance. Such a reduction in concentration also contributes to the inattention of children with ASD (Ashburner et al. 2008). The effectiveness of strategies to reduce sensory stimulation in the context of learning has to consequently be investigated. Future studies need to focus on the association between atypical SP and ADHD symptomatology in children with ASD.

Concerning the associations between atypical SP and adaptive behaviors, we found a limited number of significant results. Other recent studies have underlined that early atypical SP in childhood may result later in lower adaptive behaviors through a "cascade effect" (Baranek et al. 2018; Williams et al. 2018). There may also be a "developmental" explanation, as data on SP and adaptive skills in our study were collected during the same period. It may be necessary to collect longitudinal data to detect a latent "cascade effect" over time of atypical SP on adaptive skills.

As shown in previous studies, we observed that children with atypical scores for oral SP had lower adaptive functioning in all Vineland domains and more maladaptive behaviors (Lane et al. 2010; Tomchek et al. 2015). A recent study showed that atypical taste processing is potentially related to atypical connectivity in the primary gustative cortex and is a risk factor for feeding problems in ASD (Avery et al. 2018). Atypical oral processing was related not only with poorer adaptive functioning (communication, socialization, and daily living skills) but also more behavioral problems, suggesting the need of targeted intervention.

Atypical SP is generally reported to have a negative impact on communication skills, as shown by parental questionnaires (Baker et al. 2008; Liss et al. 2006; Tomchek et al. 2015) and direct measures (Demopoulos et al. 2015). However, children with an avoiding-sensory profile presented unexpectedly better communication skills than ASD children with typical SP. Although these findings need to be replicated, they suggest that the avoiding-sensory profile in children with ASD could be a marker of their ability to focus their attention on speech components and thus to improve their communication skills.

The strengths of our study include a large sample (Lane et al. 2010; O'Donnell et al. 2012) of children rigorously diagnosed with ASD and the standardized assessment of their SP using the long form of the Sensory Profile (Lane et al. 2010; McCormick et al. 2016; O'Donnell et al. 2012). There were also several limitations, including the absence of direct examinations of SP and no use of a control group.

In conclusion, atypical SP are very common in children with ASD in our sample and related to some important aspects of their adaptive functioning. Indeed, atypical SP explained a significant part of maladaptive behaviors, suggesting that they have to be considered in routine practice to prevent and manage subsequent maladaptive behaviors. Moreover, children with a sensory-seeking profile require specific attention, as they have lower adaptive skills and a greater risk for maladaptive behaviors. Further studies are needed to better understand the course of atypical SP and better target and personalize interventions. In addition, these studies have to begin early during childhood and to be prospective to enhance our knowledge of the role of SP on adaptive trajectories.

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Author Contributions FD conceived of the study, contributed in collection, analysis, and interpretation of the data and drafted the manuscript; AB is the PI of ELENA cohort, she also participated in the design of the current study, drafted the work and revised it critically for main intellectual content; CV participated in the design of the study and contributed to revised it critically. CM and M-CP analyzed and interpreted data; RS and NB revised critically the final manuscript. M-JO, LR, MC, CC, SV and TM contributed to collect data in ELENA cohort used in this sample. All authors read and approved the final version to be published.

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Compliance with Ethical Standards

Conflict of interest The authors have no conflicts of interest to declare.

Ethical Approval The study and informed consent procedure have been approved by the Ethics Committee on the Research of Human Subjects at Marseille Mediterranean (CNIL. number DR-2015-393).

Informed Consent Signed informed consent is obtained from all participating families included in the ELENA cohort.

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