## **U.** PORTO

2°CICLO DE ESTUDOS MESTRADO EM PSIQUIATRIA E SAÚDE MENTAL

# Extreme sensory processing patterns and relation with autistic and alexithymic traits among parents of children with Autism Spectrum Disorder and Sensory Processing Disorder – an exploratory study

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#### Abstract

The relationship between sensorial processing, autistic features and alexithymia, has been explored within autism spectrum disorder (ASD), and is not well understood. Recent evidence supports a genetic and phenotypic crossover between sensory processing disorder (SPD) and autism spectrum disorder. Our aims were to compare sensorial profiles and autistic traits between two non-clinical populations with genetic susceptibility to sensory disturbances and to explore associations between alexithymia, sensorial responses and autistic-related domains. Thirty-one parents of children with ASD, 32 parents of children with SPD and 52 parents of typically developed children completed Adolescent/Adult Sensory Profile, Autism Quotient Questionnaire and Toronto Alexithymia Scale. We found higher prevalence of extreme sensorial scorers among parents of children with ASD (17.3% to 41.9%; p=0.014) or with SPD (17.3% to 46.9%, p=0.004) when compared to parents of children with typical development. Autistic features were similar between the parents of children with these neurodevelopmental disorders. Having extreme sensorial patterns expressed was significantly associated with having alexithymia (TAS score>60), p<0.001 (60.0% to 22.4%). We also found several significant correlations between sensory quadrants, alexithymia and autistic features. Our results indicate that broad autism phenotype may be better conceptualized as broad sensorial phenotype, in other words, an atypical neurodevelopment process with variable phenotypic expression. Further studies should aim to replicate these findings among parent-ASD and SPD child dyads and combine neurophysiological measures and genotyping to explore sensorial patterns and alexithymia. Keywords: autism; broad autism spectrum, sensorial disorders, neurodevelopment disorders, alexithymia, emotional regulation, parent-child relations

#### Resumo

A relação entre processamento sensorial, traços autistícos e alexitimia, tem sido explorada na perturbação do especto de autismo e não é totalmente compreendida. Estudos recentes suportam um cruzamento genético e fenotípico entre a perturbação do processamento sensorial e a perturbação do espectro do autismo. Este estudo procurou caracterizar e comparar perfis sensoriais e traços autistícos entre duas populações com suscetibilidade genética para atipicidades sensoriais e explorar associações entre alexitimia, respostas sensoriais e traços autísticos. Trinta e um pais de crianças com perturbação do espectro do autismo (PEA), 32 pais de crianças com perturbação do processamento sensorial (PPS) e 52 pais de crianças neurotípicas preencheram o Perfil Sensorial Adolescente / Adulto, Questionário de Quociente de Espectro do Autismo e Escala de Alexitimia de Toronto. Encontramos maior prevalência de pontuações extremas no perfil sensorial entre pais de crianças com PEA (17,3% a 41,9%; p = 0,014) ou com PPS (17,3% a 46,9%, p = 0,004) comparando com pais de crianças com desenvolvimento típico. As características autistícas eram semelhantes entre os pais de crianças com estas perturbações do neurodesenvolvimento. Exibir padrões sensoriais extremos foi significativamente associado a ter alexitimia (pontuação TAS > 60), (TAS score>60), p <0,001 (60,0% a 22,4%). Também encontramos várias correlações significativas entre quadrantes sensoriais, alexitimia e traços autistícos. Os nossos resultados indicam que o fenótipo alargado do autismo pode ser melhor conceptualizado como uma atipicidade neurodesenvolvimental mais abrangente com com expressão fenotípica variável.

Estudos futuros devem procurar replicar estes resultados em díades de pais-filhos com diagnóstico de PEA ou PPS bem como combinar medidas neurofisiológicas e genotipagem para explorar os padrões sensoriais e alexitimia.

Palavras-chave: autismo; espectro alargado de autismo, perturbações do processamento sensorial, perturbações do neurodesenvolvimento, alexitimia, regulação emocional, relação criança-cuidador

### Index

1.	Introduct	ion	1
	1.1 Senso	ry atypicalities and broad autism spectrum	3
	1.2 Alexith	nymia: a possible third player in the relation between sensory difficulties an	d
	autistic	c traits	5
2.	Purpose	of the study	7
3.	Methods.		9
	3.1. Partic	sipants	9
	3.2. Mater	ials	11
	3.2.1.	Socio-demographic and clinical background	.11
	3.2.2.	Adolescent/Adult Sensory Profile (AASP)	.11
	3.2.3.	Autism Spectrum Quotient (AQ)	.12
	3.2.4.	Toronto Alexythimia Scale	.13
	3.3 Procee	dure	.13
	3.4 Data a	nalysis	.14
4.	Results		.16
	4.1. Chara	acteristics and comparisons of sensorial profiles and of autistic traits	
	betwe	een parents of children with ASD, parents of children with SPD and parents	s of
	childr	en with typical development Sample characterization	16
	4.1.1.	Sensorial Profile	.16
	4.1.2.	Autistic traits	20
	4.2. Assoc	ciations between alexithymia and sensorial responses	.22
	4.2.1.	Correlations between alexithymia and sensorial responses in the total	
	s	ample	.22
	4.2.2.	Alexithymia and the groups of parents	.24

	4.3. Relationship between atypical sensorial patterns and autism-related domains of	-
	social skills and communication	26
5.	Discussion	27
6.	Limitations and Further Studies	33
7.	Conclusions	34
8.	References	.35

#### Tables

Table 1. Sociodemographic characteristics and clinical background of ASD, SPD and TD
parents
Table 2. Extreme sensory patterns and Sensory atypicalities per sensorial quadrant among
ASD, SPD and TD parents18
Table 3. Mean scores in sensorial quadrants and categories <sup>1</sup> among ASD, SPD and TD
parents19
Table 4. Subclinical and clinical autistic traits among ASD, SPD and TD parents
Table 5. Correlation matrix between sensorial quadrant scores, autistic traits and
alexithymia23
Table 6. Alexithymia scores among ASD, SPD and TD parents

#### 1. Introduction

Sensory processing is a neurological process that includes reception, modulation, integration, discrimination, and organization of sensory input in order to elaborate an adaptive emotional and/or motor response (Dunn, 2007). An atypical, clinically relevant sensory processing is manifested by inappropriate responses to sensory stimuli. These responses can be emotional or behavioral disturbances, and interfere with the individual's daily performance. The idea that individuals could struggle in their daily functioning due to sensorial integration dysfunction was first proposed by Ayres & Tickle (1980) based on the observation of children with Autism Spectrum Disorder (ASD) (Ayres, 1980).

Following Ayres, Winnie Dunn (1997) created the Dunn's Four Quadrant Model of Sensory Processing that results from the intersection of two constructs: neurological threshold and self- regulatory behavioral response. The first refers to the threshold for noticing and responding to a sensory stimulus, the higher the threshold the greater the stimulus needed to trigger a response. The second refers to the type of response given to the stimulus and ranges from passive to active strategies. The four main sensory types resulting from the combination of these two constructs are low registration (high neural threshold and passive responding strategies), sensory seeking (high neural threshold and active responding strategies), sensory sensitivity (low neural threshold and passive responding strategies) and sensory avoiding (low neural threshold and active responding strategies) (Dunn & Westman, 1997). This process ultimately builds an individual sensory map of body and environment and is indispensable for the acquisition of knowledge and skills for daily life activities (Dunn, 1997). The difficulties in processing and using sensory information to regulate physiological, motor, affective, and/or attention responses that translate into disruption of behavior and impairment of daily life activities were categorized as sensory processing disorders (SPD) (Zero to Three, 2005). There is evidence on disturbances in sensory inputs from neuroscience

basic and translational research supporting clinical observation. Electrophysiological studies have shown abnormalities in multisensory integration among children with sensory processing disorder, comparing to children with typical development. This includes differences between responses to multisensory stimulation and summed unisensory responses and atypical maturational course of sensory gating (Brett-Green, Miller, Schoen & Nielsen, 2010; Davies, Chang & Gavin, 2009). Imagiological studies have shown reduced white matter microstructural integrity in children with sensory processing disorder involving posterior cerebral tracts and correlating strongly with atypical unimodal and multisensory integration behavior (Owen et al., 2009). Sensorial hyper-reactivity was also associated to a greater expression of D2 in the striatum of animal models who suffered pre-natal stress (Schneider et al., 2008).

Despite clinical and biological evidence, SPD is a controversial diagnosis that has been included in Infancy and Childhood Mental Health diagnostic classifications since 1994 (Zero to Three, 2005; ICDL, 2005) but not in the equivalent adult classifications (American Psychiatric Association, 1980, 1987, 1994, 2000, 2013). In fact, whereas there is a clear clinical presentation of sensorial processing disorder in newborns and infants, through late childhood, adolescence and adulthood, sensory disturbances are commonly diluted within associated anxiety (Engel-Yeger & Dunn, 2011) and depressive symptoms (Fitzgerald, 2013; Serafini et al., 2017), internalizing and externalizing behavior problems (Benarous et al., 2020; Dean, Little, Tomcheck & Dunn, 2018; Van Hulle, Lemery-Chalfant & Hill Goldsmith 2018) and even physical health symptoms reported both among clinical and general populations (Hwang, Van Dillen & Haroutounian, 2018; Lehnen et al., 2019).

McMahon et al. (2019) sought to explore the trajectory of SPS symptoms, from childhood to adulthood. In a retrospective study, they assessed sensory symptoms in childhood and the presence of a current psychiatric diagnosis in a heterogeneous adult population. They concluded that sensorial disturbances in childhood were associated

to anxiety disorders trans-diagnostically in adulthood and the association was fully mediated by emotional regulation problems (McMahon, Anand, Morris-Jones & Rosenthal, 2019). The connection between sensorial and emotional processes can explain the ubiquity of sensorial atypicalities in a wide range of psychiatric problems such as affective disorders (Serafini et al., 2016 and 2017; Hjordt & Stenbaek, 2019), addiction disorders (Bashapoor, Hosseini-Kiasari, Daveshvar & Kazemi-Taskooh, 2015; Borges, García del Castillo, Marzo & Castillo-López, 2017; Yalachkov, Kaiser & Naumer, 2010), personality disorders (Niemantsverdiet et al., 2019; Stephan, Sutin, Bosselut & Terracciano, 2017) and neurodevelopmental disorders (Zoenen & Delvenne, 2018). Sensory perception was even recently proposed to be integrated in RDoC (research domain criteria), a framework for investigating mental disorders that seeks to explore basic dimensions of functioning that span the full range of human behavior, from normal to abnormal (National Institute of Mental Health, 2019).

#### 1.1 Sensory atypicalities and broad autism spectrum

Sensory processing difficulties have been shown throughout a variety of measurements and samples of both children and adults with ASD (Baranek et al., 2005; Klintawall et al., 2011; Kirby, Dickie & Baranek, 2015; Tavassoli et al., 2013). This observation led to the inclusion of hyper- or hyporeactivity to sensory input in the ASD diagnosis of the 5th edition of the Diagnostic and Statistical Manual of Mental Disorders (American Psychiatric Association, 2013), as one of the four symptom subcategories. Sensory processing impairment has been proposed as a primary symptom of autism. Hypothetically, if individuals with autism have hyporesponsiveness to sensory stimuli, this will hinder normal development, producing a cascade effect in more complex tasks, particularly in the domains of communication and social skills (Schauder & Bennetto, 2016).

Evidence of a hereditable component to ASD has been accumulating in recent years (Chaste & Leboyer, 2012; Lai, Lombardo & Baron-Cohen, 2014), and unaffected

relatives of individuals with ASD, including parents, have been reported to have a number of autism-related traits and atypicalities in social and communication skills that are labeled as broader autism phenotype (BAP) (Bernier et al., 2011).

Only a few studies have explored sensorial processing characteristics within BAP. A study by Uljarević, Prior & Leekam (2014) reported extreme sensory patterns in 98% of the mothers of children and adolescents with ASD using the Adolescent/Adult Sensory Scale (AASP). In a similar study, De la Marche et al. (2011) assessed sensory processing among adolescent siblings of individuals with ASD, comparing to a control group (adolescents without ASD in the family), and reported similar AASP scores between the two groups except for the sensation seeking quadrant, in which siblings of ASD individuals scored significantly lower.

Glod, Riby, Honey & Rodgers (2017) explored the profiles of sensory processing in child-parent dyads within ASD families in comparison to typically developed dyads. ASD dyads shared more sensory avoidant behaviors and auditory, visual, and vestibular sensory processing atypicalities. In general, the sensory profiles were analogous within parent-child dyads across both groups (Glod, Riby, Honey & Rodgers, 2017).

Considering that the likelihood of BAP traits in relatives is higher among families in which multiple members are diagnosed with ASD (multiple-incidence/multiplex families; MPX), compared to families in which only one relative has ASD (single-incidence/simplex families; SPX), Donaldson, Stauder & Donkers (2017) assessed sensory profiles among parents of children with ASD within MPX and SPX families, comparing to parents of children with typical development. MPX parents scored significantly lower in sensory seeking, and significantly higher in the low registration, sensation avoidance, and sensory sensitivity quadrants of the AASP, when compared to SPX parents and controls, suggesting that atypical sensory processing could be part of the genetic susceptibility for ASD (Donaldson, Stauder & Donkers, 2017).

In fact, pre-clinical and clinical genetic studies have reported a shared genetic liability between sensorial dysfunctions and autism spectrum disorder (DeLorey et al., 2011; Penãgarikano et al., 2011; Tavassoli et al., 2012). In a recent and groundbreaking genetic study, Marco et al. (2018) studied eleven trios of children with sensorial processing disorder and both biological parents using whole exome sequencing, and found 18% de novo, disease-associated, single gene mutations and an enrichment of rare single nucleotide variants shared by children and their parents. These rare single nucleotide variants were located in genes reported to be associated with autism and neurodevelopmental delay. This was the first genetic study involving children with SPD diagnosis and relatives, and supports this as а specific and distinct neurodevelopmental disorder (Marco et al., 2018).

# 1.2 Alexithymia: a possible third player in the relation between sensory difficulties and autistic traits

Alexithymia is a personality construct characterized by the inability to identify, describe, and interpret emotional states that has an estimated prevalence of approximately 10% in the general population (Salminen et al., 1999). It has been recognized as a predisposing factor for psychopathology and as a negative prognostic factor in several physical illnesses (Taylor, Bagby & Parker, 1997). Furthermore, alexithymia has also been associated with a heterogeneous range of sensory modulation related to either hypo- or hyper-sensitivity (Pollatos, Dietel, Gündel & Duschek, 2015; Borhani, Làdavas & Fotopoulu, 2017; Lankes, Schiekofer, Eichhammer & Busch, 2020) and, along with sensory disturbances, is highly prevalent in major affective disorders (Serafini et al., 2016), psychosomatic disorders (Pedrosa Gil et al., 2008, Mazaheri et al., 2012; Moes-Wójtowicz, 2012; Williams et al., 2019), substance abuse disorders (Thorberg, Young, Sullivan & Lyvers, 2009; Bashapoor et al., 2015; Dorard et al., 2017) and, significantly, in autism spectrum disorder (Kinnaird, Stewart & Tchanturia, 2019). In fact, similarly to sensory processing difficulties, alexithymia has been proposed to account for the social and emotional impairments that have always been considered as core domains of ASD (Bird & Cook, 2013). Higher prevalence of alexithymia was also found in relatives of individuals with ASD suggesting that this construct could also be part of the broader autism spectrum (Carlson et al., 2004; Szatmari et al., 2008, Costa et al., 2019).

To sum up, although ASD has been established as biologically and psychologically distinct from SPD, recent evidence suggests that they share a genetic pool. Sensorial processing difficulties are part of the diagnostic criteria of ASD and are also present in BAP. Similarly, alexithymia is highly prevalent in autism spectrum disorder and also in broad autism spectrum. Both sensorial disturbances and alexithymia have been separately proposed as ASD endophenotypes, defined as hereditable traits that mediate the interaction between the genes and the observable characteristics. interestingly, emotional regulation, classically impaired in alexithymia, was recently found to mediate the relation between childhood SPD and anxiety disorders in adulthood. Nonetheless, there are no studies simultaneously addressing the relation among sensorial processing, alexithymia and autistic traits, either in individuals with autism and relatives or in other non-clinical and clinical populations.

Despite the clinical and genetic crossover, no previous study has compared the profiles of relatives of individuals with ASD with relatives of individuals with SPD. Importantly, there is no literature on the sensory profile and psychological functioning of families whose children have sensory processing disorder. This research aims to fill in this gap.

#### 2. Purpose of the study

The main goals of this study are (1) to characterize and compare sensorial profiles and autistic traits between parents of children with ASD, parents of children with SPD and parents of typically developed children; (2) to explore associations between alexithymia and sensorial responses; and (3) to investigate the relationship between atypical sensorial patterns and the autistic-related domains of social skills and communication.

Given that sensory sensory atypicalities and autistic features have a significant coexpression supported by a shared genetic susceptibility, we theorize that what is considered to be the broad autism phenotype (BAP) in fact could be non-autism specific. It might be rather a broader neurodevelopmental atypicity with roots in altered sensorial processing and variable phenotypic expression, ranging from the acknowledgment of atypical sensorial and emotional processing, with preserved functionality, to a full-blown psychiatric disorder.

Considering that sensorial disturbances and alexithymia share (1) a high prevalence in many psychiatric disorders (including autism spectrum disorder and broad autism spectrum), (2) hereditability, and (3) problems with emotional regulation, we suggest that this primary altered sensorial processing can interfere with emotional development that later translates into the difficulty in identifying and describing emotional states. This emotional impairment can, in turn, lead to deficits in social skills and communication, that are currently catalogued as subclinical autistic traits.

This conceptualization will be difficult to assess in a clinical population due to the high prevalence of sensorial disturbances and alexithymia in psychiatric disorders and their relation with mental symptoms. However, non-clinical populations with genetic susceptibility to autistic traits (e.g., parents of children with ASD) and to altered sensory processing (e.g., parents of children with SPD) can help to illuminate these relations. The conceptualization will be tested according to the following hypotheses:

H1: Extreme sensory patterns and autistic features are more prevalent both among parents of children with ASD and parents of children with SPD than among parents of children with typical development

If extreme sensory patterns and autistic features are equally prevalent among parents of children with these neurodevelopmental disorders, we obtain support for the idea of BAP's non-specificity because the same characteristics are present in relatives of children with other neurodevelopmental disorders (i.e., SPD) as well. If, in addition, the prevalence of these extreme sensory patterns and autistic features is reduced among parents of children with typical development, then the notion that an atypical phenotype is present among the other two groups of parents is reinforced.

#### H2: Extreme sensory patterns are associated with high alexithymia.

If extreme sensory patterns are more prevalent among parents presenting high alexithymia scores, then we have evidence suggesting that sensorial processing disturbances are linked to difficulties in identifying emotions and describing feelings, reinforcing the role of sensorial processing in the person's emotional development.

### H3: There is a relationship between extreme sensorial patterns and the autismrelated deficits in communication and social skills.

If an association with extreme sensorial patterns is found for the domains of communication and social skills but not for other autism-related domains, then, the idea that some features commonly regarded as autistic traits could arise from primary sensory disturbances is strengthened.

#### 3. Methods

#### 3.1 Participants

The study included 115 parents of children (with ages ranging between one and 18 years-old) diagnosed with autism spectrum disorder (n = 31 parents - ADS group), sensory processing disorder (n = 32 parents - SPD group), and children with typical development (n = 52 parents - TD group).

Participants in the ASD and SPD parents' groups were recruited among parents who were accompanying their children to the Child and Adolescent Psychiatry consultation. Parents were eligible if they had a child with a diagnosis of Autism Spectrum Disorder or Sensorial Processing Disorder established by a specialized team of infancy and early childhood. The control group of parents of children with typical development was recruited within health care professionals and their acquaintances through a snowball procedure. Parents were eligible to the study if they had children aged between 12 months and 18 years old with history of typical development.

Table 1 shows the socio-demographic and clinical characteristics of the groups of parents. The sample's mean age was  $39.27\pm6.98$  and most participants were women (70.4%, n = 81). Chi-square tests showed that men were significantly more prevalent among parents of ASD children than among parents of SPD children ( $\chi^2$  (1, N = 63) = 5.07, p = 0.024). The larger percentage of individuals with superior education in the control group possibly resulted from the snowball sampling method employed. Otherwise, the three groups did not differ significantly regarding age (both the whole sample and split by gender) or past/present physical or mental disorder (all p > 0.05).

(11 - 113)			
	ASD parents <i>n</i> = 31	SPD parents <i>n</i> = 32	TD parents <i>n</i> = 52
Gender – n (%)			
Women	17 (54.8)	26 (81.3)	38 (73.1)
Men	14 (45.2)	6 (18.8)	14 (26.9)
Current age – mean (SD)			
Mother	40.06 (7.07)	36.50 (6.23)	39.11 (6.53)
Father	40.29 (8.67)	40.17 (6.77)	43.00 (6.74)
Age at child's birth – mean (SD	))		
Mother	32.24 (4,52)	31.69 (6.34)	NA
Father	33.57 (7.01)	34.80 (8.38)	NA
Highest Level of Education – n	(%)		
Basic education	10 (34.5)	9 (28.1)	5 (10.0)
Secondary Education	9 (31.0)	14 (43.8)	10 (20.0)
Higher Education	10 (34.5)	9 (28.1)	35 (70.0)
Presence of physical disorder	– n (%)		
	3 (9.7)	5 (15.6)	6 (11.5)
Presence of psychiatric disord	ler – n (%)		
	0	4 (12.5)	1 (1.9)

Table 1 Socio-demographic characteristics and clinical background of ASD, SPD and TD parents (N = 115)

ASD parents – parents of children with Autism Spectrum Disorder, SPD parents – parents of children with Sensorial Processing Disorder, TD parents – parents of children with typical development, SD – Standard deviation.

Current age has three missing values (two fathers and one mother in the TD parents group); Education has four missing values: two in the ASD parents group and two in the TD parents group.

#### 3.2 Materials

#### **3.2.1** Socio-demographic and clinical background

Participants answered questions on general socio-demographic variables, such as gender, age group and education level. Considering that both sensory disturbances and alexithymia are prevalent among various psychiatric disorders (Bashapoor et al., 2015; Benarous et al., 2020; Dean et al., 2018; Dorard et al., 2017; Engel-Yeger & Dunn, 2011; Fitzgerald, 2013; Hwang et al., 2018; Kinnaird et al., 2019; Lehnen et al., 2019; Mazaheri et al., 2012; Moes-Wójtowicz, 2012; Pedrosa Gil et al., 2008; Serafini et al., 2016, 2017; Thorberg et al., 2009; Van Hulle et al., 2018; Williams et al., 2019) and have been associated to physical health (Hwang et al., 2018; Lehnen et al., 2019; Porcelli & Taylor, 2018), we asked participants about present or past mental and physical disorders (Table 1).

#### **3.2.2** Adolescent/Adult Sensory Profile (AASP)

The Adolescent/Adult Sensory Profile (AASP) is a 60-item self-report questionnaire based on Dunn's four quadrants model, and examines the subject's sensory modulation and processing. Each of the four quadrants or subscales comprises 15 items that are distributed by six sensory modalities (Taste/Smell, Auditory, Visual, Tactile, Movement, and Activity). Using a five-point Likert scale, participants indicate how often they respond to a sensory event in the manner described in each item. For scoring, the 60 items are sorted into the four subscales – low registration, sensation seeking, sensory sensitivity and sensation avoiding – reflecting different sensory processing patterns. Scores for each sensorial quadrant range from 15 to 75, higher scores indicating higher frequency of the respective sensory response. Normative means within five classification groups of scores have been described for the general population aged 18–64 years (Brown & Dunn, 2002). A score below the 2nd percentile is considered "Much Less than Most People". A score between the 2nd and the 16th

percentiles corresponds to "Less than Most People". Scores between the 16th and the 84th percentiles correspond to "Similar to Most People". Scores between the 84th and the 98th percentiles represent "More than Most People", and a score above the 98th percentile corresponds to "Much More than Most People" (Brown & Dunn, 2002). The questionnaire has shown an acceptable internal consistency, with *alpha* values for each quadrant varying between .639 and .775 (Brown & Dunn, 2002). The validity construct was guaranteed by a panel of experts, who assured that the instrument reflected the theoretical framework outlined by Dunn (1997) (Brown & Dunn, 2002). The AASP was translated into Portuguese and culturally adapted, showing values of internal consistency similar to the original study ( $\alpha$ =.631 to .754) (Borges et al., 2017).

#### **3.2.3** Autism Spectrum Quotient (AQ)

The Autism Spectrum Quotient (AQ) was developed by Baron-Cohen, Wheelwright, Skinner, Martin & Clubley in 2001. It is a self-assessment tool of intelligence autistic traits in adult individuals with normal intelligence quotients. The scores place the individual on a continuum between presence and absence of autism. It consists of 50 questions in total and assesses five different areas, each with 10 items: social skill (AQ Social), communication (AQ Communication), imagination (AQ Imagination), attention to detail (AQ Attention to Detail) and attention switching (AQ Attention Switching). The AQ has an adequate internal consistency in each of the five domains ( $\frac{1}{10}$ Communication = .65;  $\frac{1}{10}$ Social, = .77;  $\frac{1}{10}$ magination = .65;  $\frac{1}{10}$ Attention to Details = .63;  $\frac{1}{10}$ Attention Switching = .67) and excellent test-retest reliability (Baron-Cohen et al., 2001). Currently, the psychometric properties of the AQ for the Portuguese population have not been studied, but the English version was translated into Portuguese (Castro & Lima, 2009) and has already been used in studies with the Portuguese population, showing an adequate global Cronbach's *alpha* value of 0.60 (Veigas, 2017).

In the first study with the AQ in the United Kingdom, a 32-point cut-off bridge was established for screening individuals with clinically significant levels of autistic traits

(Baron-Cohen et al., 2001). Broad Autism Spectrum (BAP) studies usually use the threshold of 1.5 standard deviations above the mean of the control group to identify affected cases (Losh, Childress, Lam & Piven, 2008; Piven & Palmer, 1999).

#### **3.2.4** Toronto Alexithymia Scale (TAS)

The 20-item Toronto Alexithymia Scale (TAS-20) is a self-assessment tool developed by Bagby, Parker and Taylor (1994) that provides an accurate and valid assessment of the alexithymia construct. The TAS-20 has been translated into Portuguese and its factorial structure has been cross-validated through confirmatory factor analysis showing a stable and replicable factorial structure that is congruent with the alexithymia construct: (F1) Difficulty to identify feelings; (F2) Difficulty in describing feelings to others; and (F3) Externally-oriented thinking style. Cronbach's *alpha* coefficient reported for the complete Portuguese instrument is 0.75, which translates into an adequate internal consistency (Veríssimo, 2001). The empirically derived cut-off score of 61 is used for identifying individuals with "high" or "low" alexithymia (Bagby et al., 1994).

#### 3.3 Procedure

We conducted a cross-sectional study in the Child and Adolescent Psychiatry Outpatient clinic in a tertiary university hospital center. Eligible parents were invited to participate at the end of their children's medical appointments, and those who accepted to participate were taken to a separate room where the investigator gave information on the study. Parents in the group of children with typical development history received the information and the questionnaires via e-mail. After giving their informed consent, participants completed the study's battery of questionnaires. The study was approved by the Institutional Review Board of the University Hospital Center of Porto (ref. 2019-334(271-DEFI/290-CE).

#### 3.4 Data analysis

Participants' scores on each sensorial guadrant of the AASP were converted into percentiles according to the manual's references for adolescents and adults (Brown & Dunn, 2002), for comparison of our results with past studies (Donaldson, 2016; Uljarević et al., 2014). Extreme sensorial patterns were defined by scores falling below the 2nd percentile (AASP classification: "Much Less than Most People") or above the 98th percentile (AASP classification: "Much More than Most People"). We additionally explored, for each sensory quadrant, any increased (above the 82nd percentile) or decreased (below the 16th percentile) behavior response. Mean scores for each quadrant were calculated as additional information, though they fail to accurately represent the extremes of the distribution (i.e., the focus of this study). Subclinical autistic traits (BAP spectrum) was defined by threshold calculation according to previous studies (Losh & Piven, 2008; Piven & Palmer, 1999) (1.5 SD above the AQ mean score of the control group, i.e., AQ scores > 23), and we additionally included the percentage of individuals who actually showed clinically relevant autistic traits (i.e., AQ > 31). Following the recommended value (Bagby et al., 1994), individuals scoring higher than 61 on the TAS-20 were considered high in alexithymia.

For descriptive analyses, categorical variables were presented as frequencies and percentages, and continuous variables as means and standard deviations, or medians and interquartile ranges for variables with skewed distribution.

To inspect hypothesis 1, we characterized the sensorial profile and autistic traits of parents of children with ASD, parents of children with SPD and parents of typically developed children, for comparison. Chi-square or Fisher's exact test was used to assess group differences regarding (1) the proportion of extreme scorers (<2th and >98<sup>th</sup> percentiles) on sensorial processing, both in any quadrant and by quadrant, and (2) increased (above the 82nd percentile) or decreased (below the 16th percentile) behavior responses per quadrant. The same statistical tests were used for group

comparisons regarding subclinical autistic traits (BAP spectrum). Analysis of variance (ANOVA) were used to compare sensory quadrant and autism quotient scores (and respective subdomains) among the three groups of parents, followed by the Bonferroni post-hoc test when findings were significant in the ANOVA.

To test hypotheses 2 and 3, we correlated sensorial quadrant scores with alexithymia and with autism quotient scores (including the respective domains) using Pearson correlations or the non-parametric equivalent (Spearman correlation). The relation between sensorial responses and alexithymia were further inspected through analysis of variance (ANOVA), followed by the Bonferroni post-hoc test when findings were significant in the ANOVA. Fisher's exact test or the Chi-square test were used to compare the groups of parents regarding the presence of high alexithymia. The relationship between atypical sensorial patterns and the autistic-related domains of social skills and communication was further investigated through tests of the associations between different extreme sensory patterns and BAP spectrum using Chisquare or Fisher's exact test.

Finally, to examine how gender and present/past presence of mental disorders might influence sensory quadrant, autism quotient and alexithymia scores independently from group effects, we conduced independent samples *t*-tests to compare sensory quadrant scores between genders and also between those with and without a current mental or physical disorder. For these tests, all participants were pooled together and separated based only on the demographic variable in question.

All reported p values are two tailed, with a significance level of 0.05. Analyses were performed with the use of SPSS version 26.

#### 4. Results

### 4.1 Characteristics and comparisons of sensorial profiles and of autistic traits between parents of children with ASD, parents of children with SPD and parents of children with typical development

#### **4.1.1** Sensorial Profile

Extreme sensory patterns in any sensorial quadrant (sensorial quadrant scores below the 2nd percentile or above the 98th percentile) were found in 32.2% (n = 37) of the study's population. Table 2 displays the distribution of AASP percentiles per sensorial quadrant.

Comparing with parents of TD children, there was a significant association between having any extreme sensory quadrant score and being the parent of a child with ASD (17.3% to 41.9%;  $\chi^2(1, N = 83) = 6.05$ ; p = 0.014) or with SPD (17.3% to 46.9%;  $\chi^2(1, N = 84) = 8.49$ ; p = 0.004). In particular, extreme scorers in the sensorial sensitivity quadrant were significantly more concentrated in both the ASD and SPD parents' groups than in the group of TD parents (respectively, 22.6% to 5.8%;  $\chi^2(1, N = 83) = 5.18$ ; p = 0.035 and 25.0% to 5.8%; p = 0.018).

Three differences were found between parents in the ASD and in the SPD groups, but only in one did the two groups actually differed from each other. This difference referred to a higher concentration of sensorial avoidance scores below the 16<sup>th</sup> percentile among parents of ASD children than among parents of SPD children (35.5% to 9.4%),  $\chi^2(1, N = 63) = 6.21$ ; p = 0.013). In the remaining two differences, both ASD and SPD groups of parents registered greater proportions of the respective sensorial response than the TD group of parents, but the difference from the latter group was significant only for one of the former groups (and not for the other). Thus, the group of ASD (but not of SPD) parents was significantly associated to a higher prevalence of decreased sensorial seeking scores (below the 16<sup>th</sup> percentile), when compared to the group of TD parents (16.1% to 5.8%,  $\chi^2(1, N = 83) = 9.13$ ; p = 0.003). In the group of SPD (but not of ASD) parents, we also found a significantly higher prevalence of low registration extreme scores in comparison with TD parents (21.9% to 3.8%, p = 0.024).

No statistically significant differences were found between the three groups of parents regarding their mean scores in each sensory quadrant, or in each sensory modality, using analysis of variance (Table 3). However, mothers presented significantly higher scores on the vestibular (t(113) = 2.58; p = 0.011) and visual processing categories (t(83) = 3.33; p = 0.001), and on the avoidance quadrant (t(113) = 2.16; p = 0.033) than fathers did. Being a woman also was associated to heightened low registration response ( $\chi^2(1; N = 115) = 4.45$ ; p = 0.035). No statistically difference in sensory quadrants or categories were found among parents who reported physical or mental disorders.

		ASD parents	SPD parents	TD parents		<i>p</i> -value <sup>2</sup>	
		<i>n</i> = 31	n = 32	n = 52	ASD vs. SPD parents	ASD <i>vs</i> .TD parents	SPD vs. TD parents
Extreme ser	sory pattern	s in any qua	adrant – n (%	6)			
<2nd >98th		13 (41.9)	15 (46.9)	9 (17.3)	.693	.014	.004
Sensory aty	picalities per	quadrant –	n (%)				
Low	<2nd >98th	2 (6.5)	7 (21.9)	2 (3.8)	.148	.627	.024
Registration	<2nd	2 (6.5)	4 (12.5)	1 (1.9)	.672	.553	.067
	<16th	8 (25.8)	11 (34.4)	12 (23.1)	.459	.779	.259
	>82nd	4 (12.9)	8 (25.0)	8 (15.4)	.222	1.000	.276
	>98th	0	3 (9.4)	1 (1.8)	.238	1.000	.152
Sensory	<2nd >98th	5 (16.1)	3 (9.4)	3 (5.8)	.474	.143	.670
Seeking	<2nd	5 (16.1)	3 (9.4)	3 (5.8)	.474	.003	.670
	<16th	15 (48.4)	11 (34.4)	9 (17.3)	.259	.332	.075
	>82nd	1 (3.2)	3 (9.4)	2 (3.8)	.613	1.000	.364
	>98th	0	0	0	NA	NA	NA
Sensory	<2nd >98th	7 (22.6)	8 (25.0)	3 (5.8)	.822	.035	.018
Sensitivity	<2nd	2 (6.5)	1 (3.1)	0	.613	.137	.381
	<16th	6 (19.4)	5 (15.6)	7 (13.5)	.697	.539	.761
	>82nd	8 (25.8)	12 (37.5)	14 (26.9)	.319	.911	.339
	>98th	5 (16.1)	8 (25.0)	3 (5.8)	.384	.277	.018
Sensory	<2nd >98th	3 (9.7)	8 (25)	4 (7.7)	.109	1.000	.050
Avoidance	<2nd	2 (6.5)	3 (9.4)	2 (3.8)	1.000	.592	.364
	<16th	11 (35.5)	3 (9.4)	10 (19.2)	.013	.099	.353
	>82nd	5 (16.1)	11 (34.4)	9 (17.3)	.096	.890	.075
	>98th	1 (3.2)	5 (16.5)	2 (3.6)	.196	1.000	.100

Table 2 Extreme sensory patterns and Sensory atypicalities per sensorial quadrant<sup>1</sup> among ASD, SPD and TD parents (N = 115)

ASD parents – parents of children with Autism Spectrum Disorder, SPD parents – parents of children with Sensorial Processing Disorder, TD parents – parents of children with typical development.

<sup>1</sup>Assessed with the AASP (Adolescent/Adult Sensory Profile). Scores on each sensorial quadrant

range from 15 to 75 (with higher scores indicating higher frequency of the respective sensory response). Scores between the 16th and the 84th percentiles correspond to "Similar to Most People". Extreme sensory patterns are defined as scores below the 2nd or above the 98th percentile.

<sup>2</sup> Chi-square or Fisher exact test (as appropriate), with p-value < 05 indicating statistically significant differences.

Table 3

Mean scores in sensorial quadrants and categories<sup>1</sup> among ASD, SPD and TD parents (N = 115)

	ASD parents <i>n</i> = 31	SPD parents <i>n</i> = 32	TD parents <i>n</i> = 52
Sensory Quadrants – mean (SD)			
Low Registration	27.97 (6.78)	29.47 (10.02)	28.79 (6.15)
Sensory Seeking	43.06 (7.10)	44.78 (8.26)	45.83 (6.06)
Sensory Sensitivity	34.45 (11.03)	38.03 (11.81)	36.06 (8.23)
Sensory Avoidance	33.55 (10.15)	36.00 (11.26)	33.81 (7.99)
Sensory categories – mean (SD)			
Taste/Smell processing	19.48 (4.37)	19.75 (3.22)	18.94 (3.44)
Movement processing	18.19 (5.01)	19.69 (5.54)	18.31 (3.55)
Visual processing	23.39 (5.48)	25.91 (6.00)	24.81 (4.98)
Touch processing	29.94 (8.20)	32.31 (7.55)	31.58 (6.01)
Activity Level	24.31 (4.81)	25.06 (6.08)	25.79 (4.15)
Auditory processing	23.94 (7.65)	25.41 (7.61)	24.83 (4.27)

ASD parents – parents of children with Autism Spectrum Disorder, SPD parents – parents of children with Sensorial Processing Disorder, TD parents – parents of children with typical development, SD – Standard Deviation.

<sup>1</sup>Assessed with the AASP (Adolescent/Adult Sensory Profile). Scores on each sensorial quadrant range from 15 to 75 (with higher scores indicating higher frequency of the respective sensory response).

ANOVA Analysis of Variance showed non-significant group differences (p-value > .05).

#### 4.1.2 Autistic traits

The study's threshold for broad autism spectrum was 24 points (mean AQ score for parents of typically developed children + 1.5 SD). Within the study population (N = 115), only 11.7% (n = 13) met the criterion for BAP (AQ score>23) and only one father of a child with ASD showed clinically relevant autistic traits (AQ score>31). The differences in the proportions of parents with subclinical autistic traits of BAP spectrum for the three groups was non-significant (Table 4).

An analysis of variance (ANOVA) yielded significant differences among parents' groups on autism quotient total scores and on the specific domains of attention to details and communication, respectively, F(2, 112) = 4.41; p = 0.014; F(2, 112) = 3.65; p = 0.029; and F(2, 112) = 5.12; p = 0.007. Differences between parents in the ASD and in the SPD groups were statistically non-significant. However, even though parents in both these groups scored higher than parents in the TD group, Bonferroni post hoc tests showed significant differences only between parents in the SPD group and in the TD group for autism quotient total score (p = 0.011) and for the domains of attention to details (p = 0.025) and communication (p = 0.005) (Table 5). There was no statistically significant difference in autism quotient total scores regarding parents' gender and physical or mental disorders.

# Table 4 Subclinical and clinical autistic traits<sup>1</sup> among ASD, SPD and TD parents (N = 115)

	ASD	SPD	TD	<i>p</i> -value <sup>2</sup>						
	parents parents n = 31 $n = 32$		parents <i>n</i> = 52	ASD vs. SPD parents	ASD <i>vs</i> .TD parents	SPD <i>vs</i> . TD parents				
Clinically relevant autistic traits - n (%)										
AQ scores >32	1 (3.2)	0	0							
Subclinical autistic	traits (BAP) –	n (%)								
AQ scores >23	3 (9.7)	5 (15.6)	5 (9.6)	.708	1.000	.495				
Autism Spectrum C	Quotient (AQ) –	mean (SD)								
Total	16.81 (6.53)	19.56 (6.12)	15.50 (5.82)	.227	1.000	.011				
Att.Details	4.74 (2.46)	5.91 (2.44)	4.48 (2.34)	.171	1.000	.028				
Att. Switching	4.26 (1.61)	4.63 (1.74)	4.15 (1.94)	1.000	1.000	.742				
Communication	2.48 (1.91)	3.22 (2.06)	1.94 (1.49)	311	.547	.005				
Social Skills	2.48 (1.95)	2.87 (2.11)	2.00 (1.52)	1.000	.720	.170				
Imagination	2.84 (1.73)	3.03 (1.99)	2.92 (1.88)	1.000	1.000	1.000				

ASD parents – parents of children with Autism Spectrum Disorder, SPD parents – parents of children with Sensorial Processing Disorder, TD parents – parents of children with typical development, SD – Standard Deviation, BAP – Broad Autism Phenotype, Att. Details – Attention to Details, Att. Switching – Attention Switching

<sup>1</sup> Assessed with the AQ (Autism Spectrum Quotient), which ranges from 0 to 50 and places the individual on a continuum between presence (higher scores) and absence (lower scores) of autism. <sup>2</sup> Chi-square/Fisher exact test for differences in Broad Autism Spectrum (BAP). ANOVA Analysis of Variance for mean differences in AQ scores, followed by Bonferroni post-hoc tests, with *p*-value < 05 indicating statistically significant differences.

#### 4.2 Associations between alexithymia and sensorial responses

4.2.1 Correlations between alexithymia and sensorial responses in the total sample

Having extreme sensorial patterns, expressed as scoring below the 2nd percentile and above the 98th percentile in at least one sensorial quadrant, was significantly associated with having alexithymia (TAS score>60),  $\chi^2(1, N = 111) = 14.40$ ; *p* < 0.001 (60.0% to 22.4%).

Only sensory seeking registered non-significant correlations with alexithymia and its subscales. Low registration, sensorial avoidance and sensorial sensitivity scores were all significantly correlated with alexithymia (r = 0.517; p < 0.001; r = 0.495; p < 0.001; r = 0.469; p < 0.001, respectively) and with its subscales, namely with difficulty describing feelings (r = 0.544; p < 0.001; r = 0.543; p < 0.001; r = 0.447; p < 0.001, respectively) and with difficulty identifying feelings (r = 0.417; p < 0.001; r = 0.437; p < 0.001, respectively) (Table 5).

Table 5
Correlation matrix between sensorial quadrant scores (AASP), autistic traits (AQ) and alexithymia (TAS-20)

	1	2	3	4	5	6	7	8	9	10	11	12	13	14
0. Sensorial quadrant														
1. L.Registration	1													
2. S. Seeking	.202	1												
3. S. Sensitivity	,676 <sup>**</sup>	,034	1											
4. S. Avoidance	,670**	-,040	,813 <sup>**</sup>	* 1										
5. AQ total	,352**	-,152	,395*	* ,392**	1									
6. Att. to Details	,133	,170	,156	,126	,529**	1								
7. Att. Switching	,367**	-,175	,487*	* ,480 <sup>**</sup>	,667**	,123	1							
8. Communication	,336**	-,101	,301*	* ,212*	,694**	,129	,360**	1						
9. Social Skills	,283**	-,246*	,352 <sup>**</sup>	* ,414**	,733**	,151	,449**	,493**	1					
10. Imagination	,050	-,228*	-,015	,080,	,630**	,072	,327**	,353**	,375**	1				
11. TAS total	,517**	-,094	,495 <sup>**</sup>	* ,469**	,397**	,168	,373**	,299**	,322**	,147	1			
12. TAS DIF	,544**	-,036	,543 <sup>**</sup>	* ,447**	,334**	,144	,333**	,285**	,252 <sup>*</sup>	,087	,842**	1		
13. TAS DDF	,417**	-,097	,437*	* ,427 <sup>**</sup>	,427**	,217*	,369**	,293**	,324**	,193 <sup>*</sup>	,847**	,677**	1	
14. TAS EOT	,243*'	* -,118	,201*	,263**	,250**	,092	,237*	,177	,219 <sup>*</sup>	,106	,705**	,280**	,447*'	່ 1

\**p* < 0.05; \*\**p* < 0.01.

AASP – Adolescent/Adult Sensory Profile; L.Registration – Low registration, S. Seeking – Sensorial Seeking; S.Sensitivity – Sensorial Sensitivity, AQ – Autism Quotient and respective domains Att. to details – Attentio to Details; Att. Switching – Attention Switching; TAS – Toronto Alexithymia Score; DIF – Difficulty identifying feelings; DDF – Difficulty describing feelings; EOT – Externally-oriented thinking.

#### **4.2.2** Alexithymia and the groups of parents

The trait alexithymia (TAS-20 score > 60) was present in 26.1% (n = 30) of the study population, and the difference between parents in the ASD and in the SPD groups was non-significant. Comparing to parents with TD children, this trait was more expressed in both parents of ASD children and parents of SPD children, although the difference reached statistical significance only for the latter group of parents (43.8% to 13.5%;  $\chi^2(1, N = 83) = 9.69; p = 0.002$ ) (Table 6).

Consistently, analysis of variance of mean alexithymia scores showed that differences between the groups of ADS and SPD parents were non-significant regarding alexithymia total score and subscales. Both ASD parents and SPD parents' groups registered higher alexithymia (total and subscale scores) than parents in the TD group. Bonferroni post hoc tests showed significant differences regarding TAS-20 total and externally-oriented thinking scores (respectively, p = 0.04 and p < 0.001; p = 0.002 and p < 0.001). The ANOVA had shown significant differences in TAS-20 total score and the externally-oriented thinking subscale among the parents' groups, respectively, F(2, 110) = 8.45; p < 0.001 and F(2, 110) = 40.65; p < 0.001 (Table 6).

Parents who reported current or past psychiatry disorders registered higher scores in alexithymia's total scale (t(110) = 2.751; p = 0.007) and also in the difficulty identifying feelings (t(110) = 2.236; p = 0.027) and externally-oriented thinking (t(110) = 2.382; p = 0.019) subscales. We found no statically difference on the TAS-20 scores between genders or in association with having a physical disorder.

Table 6 Alexithymia scores<sup>1</sup> among ASD, SPD and TD parents (N = 115)

	ASD	SPD	TD	<i>p</i> -value <sup>2</sup>				
	parents <i>n</i> = 31	parents <i>n</i> = 32	• •		ASD vs.TD parents	SPD <i>vs</i> . TD parents		
High Alexithy	vmia – n (%)							
TSA-20 > 60	9 (29.0)	14 (43.8)	7 (13.5)	.225	.082	.002		
Toronto Alex	ithymia Scale–	mean (SD)						
Total	56.06 (12.60)	56.73 (15.86)	45.81 (13.01)	1.000	.004	.002		
DIF	14.65 (7.40)	17.07 (7.87)	14.63 (6.92)	.596	1.000	.049		
(DDF)	13.58 (4.03)	14.17 (4.65)	12.13 (4.52)	1.000	.459	.143		
Externally- Oriented Thinking (EOT)	25.13 (3.82)	23.00 (5.85)	16.42 (4.25)	.223	.<.0001	<.0001		

ASD parents – parents of children with Autism Spectrum Disorder, SPD parents – parents of children with Sensorial Processing Disorder, TD parents – parents of children with typical development, SD – Standard Deviation.

<sup>1</sup> Assessed with the TAS-20 (the 20-item Toronto Alexithymia Scale), which ranges from 20 to 100, identifying individuals with "low" or "high" alexithymia (corresponding, respectively, to higher or lower scores).

<sup>2</sup> Chi-square test for differences among parents scoring high in alexithymia. ANOVA Analysis of Variance for mean differences in alexithymia scores, followed by Bonferroni post-hoc tests, with p-value < 05 indicating statistically significant differences.

# 4.3 Relationship between atypical sensorial patterns and autism-related domains of social skills and communication

Sensorial quadrants of low registration, sensorial avoidance and sensorial sensitivity scores were all significantly and positively correlated with autism quotient total score (r = 0.352; p < 0.001; r = 0.395; p < 0.001; r = 0.392; p < 0.001, respectively) especially in the areas of attention switching (r = 0.367; p < 0.001; r = 0.487; p < 0.001; r = 0.480; p < 0.001, respectively), communication (r = 0.336; p < 0.001; r = 0.301; p = 0.001; r = 0.414; p < 0.001, respectively) (Table 5).

We also found a significant positive correlation between autism quotient and alexithymia (r = 0.397, p < 0.001), also expressed in two of its components, namely difficulty to describe feelings (r = 0.427; p < 0.001) and difficulty to identify emotions ( $r = 0.334 \ p < 0.001$ ). These components were also correlated with specific autism domains, specifically attention switching (r = 0.369; p < 0.001 and r = 0.333; p < 0.001, respectively), communication (r = 0.293; p = 0.002 and r = 0.285; p = 0.002) and social skills (r = 0.324; p < 0.001 and r = 0.252; p = 0.007, respectively) (Table 5).

Having increased sensorial avoidance responses (scores above the 98<sup>th</sup> percentile) and extreme sensorial responses (scores under the 2th and above the 98<sup>th</sup> percentile) were significantly associated to the presence of BAP spectrum (AQ score>23), 30.8% to 3.9% (p = 0.006) and 38.5% to 9.8% (p = 0.013).

#### 5. Discussion

By comparing sensorial atypicalities and autism features among relatives of children with autism spectrum disorder and sensory processing disorder, this exploratory study sough to expand the knowledge about sensorial disturbances and their relation with autistic features and alexithymia and inspect whether broad autism phenotype reflects a primary neurodevelopmental atypicity related to sensory processing.

In support of hypothesis 1, we found that extreme sensorial patterns (i.e., scores below the 2<sup>nd</sup> and above the 98<sup>th</sup> percentiles) were more prevalent both among parents of children with SPD and parents of children with ASD, when compared to parents of children with typical development. The observed prevalence of extreme sensory patterns in the ASD parents' group (48.4%) and in the SPD parents' group (41.9%) was similar to Uljarević et al.'s (2014) study reporting a 44% prevalence among mothers of children with ASD. Globally, sensorial patterns in ASD and SPD were similar in their aticipicity.

When compared with parents of TD children, both ASD and SPD showed a higher concentration of extreme scores in sensorial sensitivity (significant for both groups) and low registration (significant for SPD parents group) and a decreased sensory seeking score (significant for ASD parents group).

The only significant difference between ASD and SPD parents was that decreased sensory avoidance score was more prevalent in the first group.

This finding has to be interpreted with caution because we found a gender effect on increased sensory avoidance responses and these two groups have significant differences regarding the proportion of men.

Glod et al (2017) also found atypical sensory responses among parents of children with ASD, reflected in deviant mean scores, on low registration, sensorial sensitivity and sensorial avoidance. In our study, the higher prevalence of sensory atypicalities was not accompanied by differences in quadrant and categories scores between the

groups. We suggest that the concentration of scores in both extreme outer percentiles (<2th and >98th) in the groups of parents of children with neurodevelopmental disorders might have contributed to a non-skewed distribution.

Although non-significant differences were found among groups regarding the prevalence of broad autism spectrum, 15.6% of SPD parents registered scores within the BAP spectrum, and their mean AQ scores were significantly higher when compared to the TD parents' group. Other studies have suggested a 14-23% prevalence of phenotypic features of autism in first-degree relatives of individuals with ASD (Georgides et al., 2013; Sasson, 2013). We did not observe these results in the group of ASD parents, in which less than a tenth of ASD parents were included within the BAP spectrum. We suggest that the reduced proportion of BAP spectrum in ASD parent's group is related to the smaller prevalence (and associated incidence) of ASD (3.22%) (Saito et al., 2020) when compared to SPD (5-15%) (Galiana-Simal et al.,2020). A smaller incidence suggests a lower genetic penetration, so this sample may be undersized in number of parents of children with ASD which albeits the detection of significant differences regarding autistic features. In this sense, we argue that results obtained in parents of children with SPD support alone the presence of an atypical phenotype related to sensorial disturbances and the idea of BAP-nonspecificity supporting hypothesis 1.

Our results confirmed hypothesis 2. Low registration, sensorial avoidance and sensorial sensitivity responses were moderately correlated with alexithymia supporting the idea that atypical sensorial processing may interfere with emotion knowledge and regulation.

These findings add to the existing literature regarding the deficits in interoception and body ownership in this personality construct. It has been theorized that a disruption in multisensory integration between exteroceptive and interoceptive channels might

explain the difficulties to detect and interpret bodily sensations in alexithymic individuals (Herbert, Herbert, & Pollatos, 2011; Shah, Hall, Catmur, & Bird, 2016). Atypical sensory processing also contributes to alterations in the bodily self as the interaction with the world through our sensorial apparatus is itself responsible for establishing the internal representation of our body limits. Also, when an individual has increased sensorial sensitivity, a stimulus neutral to the majority of people, may be charged with a particular positive and negative affect and integrated in emotional memory. We propose that this process in early stages on emotional development could promote the indiscrimination of affective states and, alternatively, an adaptive reliance on external cues for action (external-oriented thinking).

Another view concerns alexithymia as a psychoanalytic concept. The relation with the primary caregiver has been proposed as the basis for establishing the child's symbolizing function and capacity to understand his or her own emotional states. A child with atypical sensory processing poses increased difficulties for caregivers' reading and interpretation of affective states and fulfillment of needs. The frustration of the caregiver and the repetition of unpleasant experiences in the child may interfere with integration of mind-body experiences and the establishing of the psychosomatic unit. It may also interfere with attachment that shapes the relation with significant others with a possible deficit in communication and social skills.

Although the cross-sectional design of our study prevents the establishment of causal relations, this study's population was chosen due to a genetic susceptibility strongly favoring the role of sensorial disturbances as the primary alteration.

Considering that the sensorial quadrants that were correlated with alexithymia were the same that showed extreme patterns in parents of children with ASD and parents of children with SPS, we further compared the prevalence of alexithymia among the study's groups. The personality trait alexithymia was highly prevalent both in parents of children with ASD and parents of children with SPD. The single previous study that assessed alexithymia in parents of children with ASD found a much lower prevalence

(10.3% in Szatmari et al., 2008). Interestingly, the difference was not translated in the subscales related to affect and emotion regulation but only in externally-oriented thinking (EOT). EOT has been associated with lower internal consistency (Komaki et al, 2003; Taylor, Bagby, & Parker, 2003) and higher liability to cultural factors (Dere et al., 2012, 2013). The cultural factor is not suitable to explain this result because the study's population shares the same cultural background (Portuguese people living in the North of Portugal). Furthermore, the education level was similar between groups (except in the control group, that has a significantly higher proportion of individuals with higher education).

Although many studies reported high internal consistency and the validity of a threefactorial structure of the TAS-20, Reise et al. (2013) argued that alexithymia is best measured as a total scale score (Reise, Bonifay, & Haviland, 2013). Since total alexithymia score stands on its own for the assessment of alexithymia construct, the fact that only EOT scores (and not difficulty identifying and describing feeling) were significantly higher in parents of children with neurodevelopmental disorders is not enough to exclude the existence of a disturbed affect regulation.

The five participants who, in our study, reported present/past psychiatric disorders, had significantly higher alexithymia scores. This adds to the collected evidence of the relation between alexithymia and psychiatry disorders (Honkalampi et al., 2018). The trait alexithymia has been reported to be present in 13.8-33% of individuals with anxiety disorders (Honkalampi, De Berardis, Vellante & Viinamäki, 2018), and it seems to affect the search for treatment (Rufer, Moergeli, Moritz, Drabe, & Weidt, 2014). This evidence strengthens the need to control for this confounding factor in similar future studies.

Regarding hypothesis 3, we found a moderate positive correlation between both sensorial avoidance, sensorial sensitivity and social skills, communication and attention switching. In the autism quotient questionnaire, items referring to social skills are linked to feeling unease in social situations and active behavior toward avoiding them. Items

related to communication refer to difficulties in reading people's emotions and intentions. In fact, social situations are frequently charged with sensorial stimuli, and it is possible that an individual may avoid socialization because he or she is oversensitive to stimuli and/or tends to give sensorial avoidance responses. On the other hand, an individual with a high threshold to sensory experiences (increased low registration) may not notice or detect changes in sensory situations, missing emotional clues necessary for interacting and empathizing with others.

This assumption gathers further support in the correlation of deficits in social skills, communication and attention switching were correlated with the conceptual framework of alexithymia, specifically, difficulty describing and identifying feelings.

We had not expected the correlation with attention switching. However, integrating this finding with Dunn's sensory quadrants model, it is reasonable that a low neurological threshold (sensorial avoidance and sensorial sensitivity) may interfere with the ability to maintain and diverge attention. This is commonly expressed in another neurodevelopmental disorder, i.e., attention deficit and hyperactive disorder (ADHD) (Bijlenga, Tjon-Ka-Jie, Schuijers, & Kooij, 2017).

Our results are in line with Liss, Mailloux, & Erchull's (2008) study, which also unveiled an association between sensorial processing sensitivity, autism symptoms (attention to details, poor social skills and communication) and alexithymia in a non-clinical population.

A direct practical application of the study's results refers to the possible impact of these characteristics in the caregiver-child interaction. Regarding sensorial processing, parents with a high sensorial sensitivity and tendency to avoidance of sensorial stimuli could produce a modelling effect in their children by signaling threat or disgust toward stimulus that are irrelevant to most population. If this child is already genetically predisposed to atypical sensory processing, he or she is amenable to a potentiation and sustainment of atypical behavioral responses. Even more importantly, alexithymia,

which was highly prevalent among both parents of children with SPD and parents of children with ASD, has been shown to exacerbate the challenges in the parent-child relationship, especially if a child has a neurodevelopmental disorder (Crowell, Keluskar, & Gorecki, 2019). A previous study identified alexithymia as a possible contributor to the reduced interaction between children with ASD and their parents (Costa et al., 2019). Accumulated evidence indicates a relationship between alexithymia and dysfunctional patterns of affective involvement in the family (Cuzzocrea, Barberis, Costa, & Larcan, 2015; Pellerone, Tomasello, & Migliorisi, 2017). Considering the increased prevalence of extreme sensorial patterns and alexithymia in a population of parents whose children have increased difficulties in equivalent and other domains, they may benefit from individual or dyad approaches to interventions like multisensory integration therapy and training in identifying and communicating feelings (e.g., mentalization-based therapy, mindfulness).

We found a similar atypical sensory processing and increased autistic traits in parents of children with two neurodevelopmental disorders (ASD and SPD). This phenotype was atypical in the sense that it was not present among individuals with typical development (i.e., with no genetic relationship with autism or with sensory processing disorders). Thus, reducing the clinical features labeled as subclinical autistic traits to the BAP spectrum might be misleading because they may have different biological foundations. In our study parents have, in common, a genetic susceptibility to sensory disturbances that was phenotypically expressed favoring a primary sensory disturbance underpinning this neurodevelopmental atypicity.

The high prevalence of atypical sensorial patterns and alexithymia in non-clinical populations expresses the dimensionality of mental functioning and the necessity of integrating them in the full range of human behavior. These characteristics may not translate into functional impairment or other mental health symptoms. However, they

may have a direct impact in caregiver-child interaction that is already extra challenging in children with neurodevelopmental disorders.

Finally, the supported relationship between atypical sensory processing and alexithymia indicates the importance of the sensorial apparatus to higher-order functions as cognition and emotion. Although the categorical system remains necessary in clinical contexts, research approaches that incorporate the dimensionality and heterogeneity of mental phenomena are more useful to access psychiatric disorders' etiology. Globally this study places sensory processing as an essential dimension in the study of mental functioning

## 6. Limitations and Further Studies

We identify a few limitations in this study. Firstly, the selection of the parents in the control (TD) group was conducted in the absence of a developmental test to confirm the normal development of their children, although mental health professionals conducted the snowball sampling procedure to potential participants that they were aware had children without neurodevelopmental or other developmental atypicities. In any case, the strict selection criteria applied to the other two study groups mitigates this aspect. Also, parents of ASD and SPD children usually resort to child and adolescence psychiatry consultations for behavior alterations that take place between the infants' first and second years of life. These changes in behavior are usually noticeable in child-caregiver interactions. The diagnosis of these children is based on a multi-assessment method by a specialized team. Considering the type of symptoms and age of presentation of these neurodevelopmental disorders, parents' report of a child's normal development probably qualifies as suitable comparison.

Another limitation was the small sample, especially regarding the sensorial quadrants. Lastly, this is a cross-sectional study, and both sensorial patterns (McMahon et al., 2019) and alexithymia (Matilla et al., 2010) tend to alter throughout life, which may

influence the establishment of longitudinal relationships. Also, adult population is not ideal to study characteristics that have an early development as they have already established their personality and submitted to various modelling effects and have established their personality and been exposed to diverse environmental factors.

Future replication of this study with an additional comparison of these characteristics between parent- ASD or SPD child dyads is important to cast further light on our results. The combination of neurophysiological measures to assess sensory disturbances could help to distinguish biologically sensory and emotional processing. Both anxiety and depression symptoms shape sensorial experience, and alexithymic individuals may have difficulties in detecting and describing these symptoms, so we suggest the assessment of these features in further investigation on a similar population. Finally, we propose genotyping individuals with altered sensory processing as primary dimensional trait could enlighten the genetic cross-over between psychiatric disorders like bipolar disorder, schizophrenia, autism, ADHD, addiction disorders as well as the co-morbidity between them.

## 7. Conclusions

Our results are suggestive of an increased prevalence of extreme sensorial patterns and alexithymia in different clinically-susceptible populations, challenging the concept of broad autism spectrum and supporting a broader neurodevelopmental atyipicity built on sensorial processing. Also, the observed increased sensorial behavioral responses in low registration, sensory avoidance and sensory sensitivity associated to autistic features and high alexithymia reinforce the role of sensorial processing in the modelling of cognition and emotion systems. The high prevalence of alexithymia should raise concern on the impact on the caregiver-child interaction and encourage further studies and timely interventions. Future investigations should pursue the neurophysiological and genetic markers of this association.

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