PSYCHIATRY | REVIEW ARTICLE

Sensory processing disorder: Key points of a frequent alteration in neurodevelopmental disorders

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Abstract: Altered neurological sensory integration results in Sensory Processing Disorder (SPD), also known as Sensory Regulation Dysfunction, Sensory Integration Dysfunction or Sensory Dysfunction Disorder. Under this condition, the brain doesn't process sensory inputs correctly, following inappropriate behavioral and motor responses that affect learning, coordination, behavior and language. SPD may lead to stress, anxiety or even depression, and represents a risk of psychopathology. Epidemiological studies carried out in western lifestyle populations have shown a high prevalence of SPD among children (5-15%); however, a large number of health professionals still do not know this condition, giving rise to unattended children and frustrated families. This review aims to provide an updated starting point about some of the most relevant aspects of SPD.

Subjects: Pediatrics & Child Health; Child & Adolescent Psychiatry; Occupational Therapy

ABOUT THE AUTHOR

Our group is established within the Child Developmental Center of Ciudad Real (CDICR), Spain. The CDICR is a health institution specialized in the diagnosis and treatment of neurodevelopmental disorders. Both research and clinical practice are carried out by an interdisciplinary team, that also participates in projects with universities and hospitals, mainly in the areas of psychiatry, pediatrics, psychology and occupational therapy. Our research goals include the characterization of neurobiological, cognitive and behavioral markers to better understand neurodevelopmental disorders. Dr. Adrián Galiana-Simal, earned its PhD at the Physiology Department of Complutense University Medicine School and leads the CDICR Applied Neuroscience department. OT/Psyc María Vela-Romero is the CDICR main director and has a vast experience in Autism and sensory disturbances treatment. Dr. Luis Beato-Fernández leads the Psychiatry Department at the General Hospital of Ciudad Real and is a renowned expert in Neurodevelopmental and Eating Disorders.

PUBLIC INTEREST STATEMENT

The present study is an updated review about sensory processing disorder (SPD). Briefly, SPD is defined as a condition where brain processing of sensory information is not correctly arranged, giving rise to inappropriate behavioral and motor responses. SPD symptomatology is a very frequent feature of neurodevelopmental disorders, and may be present in Autism, ADHD and Learning disabilities. This paper covers and synthesizes all relevant aspects of SPD: a brief introduction to the sensory integration process; a description of SPD subtypes and current classification systems; most relevant prevalence studies; related factors and comorbidity; derived problems; common signs; candidate biomarkers; assessment tools; current treatment strategies; and finally, an integrative conclusion. Our work represents an initial contact with the sensory processing difficulties for those interested in childhood development and its associated disorders.
Keywords: Autism spectrum disorder; developmental disorders; neurodevelopment; sensory integration; sensory processing disorder; sensory regulation dysfunction

1. Introduction
Sensory receptors capture external stimuli from the environment (through tactile, visual, gustatory, olfactory or auditory receptors) or internal stimuli originated in our own body (via tactile, vestibular or proprioceptive receptors). These receptors transform the captured stimulus into sensory information that is sent to the brain to be processed, resulting in a determined motor and behavioral response. The brain processing of sensory information is called Sensory Integration (SI) and allows reacting effectively to stimuli. In the 1970s and early 1980s, Dr. Anna Jean Ayres (OT, PhD) formulated SI theory through extensive research with children with learning disabilities (Ayres, 1977a; Ayres & Mailloux, 1981). In particular, and in the words of Dr. Ayres (1972), SI is “The neurological process that organizes sensation from one’s own body and from the environment and makes it possible to use the body effectively with the environment”.

Some authors divide the SI process in 4 phases (Del Moral Orro et al., 2013): registration (the brain receives sensory information from sense organs); modulation (allows the regulation of stimulus intensity); discrimination (the stimulus is organized and interpreted to distinguish its relevance, characteristics and specific qualities) and response (the brain integrates all the processed stimuli to generate an appropriate response that will lead to a particular behavior and movements). Sensory Processing Disorder (SPD) (also known as Sensory Regulation Dysfunction, Sensory Integration Dysfunction or Sensory Dysfunction Disorder) can be defined as a condition in which one or more of the SI phases are altered, giving rise to unadapted behavioral and/or motor responses (Galiana-Simal et al., 2017). SPD may affect learning, coordination, behavior, language or sensorimotor development, among others, hindering daily life activities and occupational participation (Chien et al., 2016; Corbett et al., 2016; Crozier et al., 2016). Despite this, a large number of health and educational professionals are still unaware of SPD. The present work aims to provide a general and updated starting point about this disorder, reviewing several important aspects such as assessment, prevalence, related factors and comorbidity, related problems, common signs, candidate biomarkers and treatment.

2. Method
A bibliographic review was carried out in December 2017 to find relevant information on several aspects of the disorder: assessment, prevalence, comorbidity, related factors, derived problems, habitual signs and treatment. In order to find peer-reviewed works with scientific relevance in the SPD field, a wide search was performed in some of the most relevant scientific and academic databases: PubMed, Scopus and Google Scholar, comprising more than 60 studies from the 1970s to the present. Descriptors used were: “prevalence”, “comorbidity”, “factors”, “signs”, “symptoms”, “valuation”, “biomarker”, “diagnostic”, “assessment” and “treatment” along with “sensory processing disorder”, “sensory regulation dysfunction”, “sensory integration dysfunction” and “sensory dysfunction disorder” descriptors.

3. Sensory Processing Disorder (SPD)
SPD is recognized today as a diagnostic entity in the manual Diagnostic Classification of Mental Health and Developmental Disorders of Infancy and Early Childhood-Revised generally known as Zero to Three (Egger & Emde, 2011) and in the Interdisciplinary council on developmental and early disorders (Greenspan & Wieder, 2008). In addition, sensory difficulties are a frequent feature in Autism Spectrum Disorder (ASD) and are included in DSM-5 (DSM5 American Psychiatric Association A, 2013) as a manifestation within the diagnostic criteria “restricted, repetitive patterns of behavior, interests or activities” (Schaaf & Lane, 2015). Individuals with SPD have impaired responses to, processing of, and organization of sensory information that affects participation in functional daily life routines and activities (Miller et al., 2009). SPD is especially difficult to understand and diagnose due to the fact that one or more of the sensory systems could be jointly...
affected (visual, auditory, tactile, smell, taste, vestibular, proprioception and interoception). This scenario gives rise to a wide variety of symptoms, requiring different treatment strategies depending on the case. Because of its heterogeneity, SPD includes subtypes that are classified according to different models. Two of the most widely used are Miller’s and Schaaf’s classifications.

3.1. Miller’s classification
According to Dr. Lucy Jane Miller’s (OT, PhD) model, SPD can be subclassified into 3 types that can co-exist combined: sensory modulation disorder (SMD), sensory-based motor disorder (SBMD) or sensory discrimination disorder (SDD) (Miller, Anzalone et al., 2007; Miller et al., 2009) (Figure 1). SMD refers to difficulty regulating responses to sensory stimulation and include 3 subtypes: sensory over-responsive (SOR), sensory under-responsive (SUR) and sensory craving (SC). SBMD denotes difficulty with balance, motor coordination, and the performance of skilled, non-habitual and habitual motor tasks. Within SBMD two subtypes are proposed: dyspraxia and postural disorder. Regarding SDD, it refers to difficulty interpreting the specific characteristics of sensory stimuli and can be present in any of the sensory systems (Miller et al., 2009).

3.2. Schaaf’s classification
Dr. Roseann C. Schaaf (OT, PhD) model can be consulted in her “Clinician’s guide for Implementing Ayres Sensory Integration” (Schaaf & Mailloux, 2015). This classification refers common patterns of SPD derived from studies comparing typically developing children and children with different developmental disorders, including, but not limited to ASD; it also includes the Ayres Sensory Integration Assessment Interpretation Tool© (ASI–IT) to interpret patterns (Schaaf & Mailloux, 2015). Schaaf’s classification is based on Dr. Ayres Sensory Integration Test (Ayres, 1977b) and the Sensory Integration and Praxis Test (SIPT) (Mailloux, 1990). Common patterns of SPD are classified as follows: Poor sensory perception overview; Somatodyspraxia (SD); Vestibular and bilateral integration deficits (VBID); Visuodyspraxia (VP); and Sensory reactivity (Figure 2). The pattern of poor sensory perception overview is characterized by difficulty in the ability to identify, discriminate, and interpret sensory information in more than one sensory system. SD is a pattern associated with poor sensory perception (especially tactile) combined with signs of poor motor planning involving the imitation, planning, and sequencing of actions. VBID refers poor vestibular processing along with difficulties in related motor functions such as muscle tone, postural and ocular-motor control, balance, midline integration and bilateral coordination. VP means poor visual perception with poor visual-motor skills and planning. Finally, sensory reactivity refers excessive or excessive reactions to typical levels of sensation that interfere with participation in daily activities; may manifest as the fight, flight, or freeze reaction and may produce anxiety, high activity level or inattentiveness. Sensory reactivity can manifest as either sensory hyperreactivity or hyporreactivity. These SPD patterns can be detected by some of the most used sensory processing scales and

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**Figure 1. Classification of Sensory Processing Disorder and subtypes proposed by Dr. L.J. Miller (Miller, Anzalone et al., 2007).**
evaluation tools (see “assessment” section). Following assessment of SPD patterns, ASI–IT must be applied in order to interpret them (Schaaf & Mailloux, 2015). The ASI–IT classifies detected problems in 3 main categories: problems in sensory perception (subclassified in vestibular processing, proprioception, tactile perception and visual perception); problems in motor-related function (subclassified in postural or ocular mechanisms, bilateral integration, postural mechanisms, body-centered praxis and visuopraxis); and problems in sensory reactivity (subclassified in hyperreactivity and hyporeactivity). When problems in vestibular processing, proprioception, postural or ocular mechanisms, bilateral integration and postural mechanisms are present in the same case, ASI–IT classifies it as problems in vestibular bilateral integration (or VBID). In the same way, problems in somatopraxis (or SD) appear when problems in proprioception, tactile perception, postural mechanisms and body-centered praxis occur together. Finally, problems in visuopraxis (or VD) are present when visual perception and visuopraxis problems are detected jointly.

Then, to better understand this complex disorder and provide a starting overview, we briefly present some interesting studies about prevalence, related factors and comorbidity, related problems, common signs, candidate biomarkers, assessment and treatment of SPD.

4. Assessment
At the moment, the lack of validated biomarkers makes SPD assessment difficult. What can be observed concerning SPD is behavior and performance problems, in contrast, for example, to acquired brain damage, which can be observed by neuroimaging techniques.

The most commonly used tools for assessing children sensory processing are the Sensory Integration and Praxis Test (SIPT), the Sensory Processing Measure (SPM) and the Sensory Profile (SP) (Jorquera-Cabrera et al., 2017). Among them, SP (and its new version, SP2) is one of the most used SPD assessment tools due to its simplicity and reliability (Chien et al., 2016; Dunn & Westman, 1997). This questionnaire examines the sensory performance of children in combination with other assessments, observations and reports and must be completed by adults that have been present and involved during children development (generally their parents). The short version of SP has been the most widely used tool in the cited epidemiological studies (Ahn et al., 2004; Engel-Yeger, 2010; Gouze et al., 2009; Roman-Oyola & Reynolds, 2013) due to its versatility (it has several versions depending on the pursued objectives).

Finally, it should be mentioned the current efforts in the development of a novel and free assessment tool to evaluate the nuclear aspects of SPD: the Evaluation in Ayres Sensory Integration (EASI) test (Mailloux et al., 2017). More information about this project is available at https://www.cl-asi.org/easi.

Figure 3 shows the main features of available tools to assess sensory processing.
5. Prevalence
Most epidemiological studies carried out in western-lifestyle populations indicate a surprisingly high prevalence of SPD among children. For example, a study of 703 children aged 3 to 6 years in North American public schools concluded that 13.7% met the diagnostic criteria of SPD (Ahn et al., 2004). In a sample of 796 boys and girls aged 3 to 10 years recruited from across all regions of the USA revealed an 11.6% of SPD prevalence (Gouze et al., 2009). In that study, it was observed that: boys were more likely to have SPD than girls (14.6% vs 8.6% respectively); 63% of participants with SPD also had a psychiatric disorder; 37% of participants with SPD did not present other disorders (meaning 5% of total), indicating that SPD exists independently of psychiatric disorders; and, the high occurrence of psychiatric disorder in children with sensory regulation problems suggests that poor sensory regulation is a significant risk factor for psychopathology (Gouze et al., 2009). Analyzing this study, we can estimate that the prevalence of “pure” SPD (when children show sensory processing alterations in the absence of any other disorder, also known as idiopathic SPD (Miller, Anzalone et al., 2007)), could be around 5%. Similarly, another study showed that 15% of 395 boys and girls from Israeli schools aged 3 to 10 years met criteria of SPD (Engel-Yeger, 2010). Another study, with a sample of 141 preschoolers in three different regions of Puerto Rico, revealed that 19.9% of them showed SPD (Roman-Oyola & Reynolds, 2013). Summarizing, most SPD prevalence studies show a worrying cipher of 15%, however, SPD remains almost unknown for families, teachers and even health professionals.

6. Related factors and comorbidity
Nowadays, the etiology of SPD is unknown. However, it has been described that SPD development is related to some specific factors such as prenatal or birth complications, low birth weight (less than 2.2 kg) and premature birth (46% of children born with less than 32 weeks of gestation present SPD symptoms at 4 years) (Craizer et al., 2016), parental stress (Gourley et al., 2013), alcohol and drugs consumption during pregnancy (Hansen & Jirikowic, 2013), as well as certain genetic factors, high exposure to chemical agents during childhood and poor sensory stimulation (Ben-Sasson et al., 2009; Keuler et al., 2011; May-Benson et al., 2009; Schneider et al., 2008).

However, it should be noted that SPD often appears along with other disorders, such as ASD (comorbidity between 80–90% of cases) (Al-Heizan et al., 2015; Baker et al., 2008; Baranek et al., 2006; Leekam et al., 2007). In addition, SPD symptomatology could be present in about 60% of cases of attention-deficit hyperactivity disorder (ADHD) (Ahn et al., 2004; Mangeot et al., 2001), in
Down syndrome (49%) (Bruni et al., 2010), the opsoclonus myoclonus syndrome (63%) (Green et al., 2016), dysfunctional evacuation syndrome (53%) (Pollock et al., 2014), urinary incontinence (44%) (Cupelli et al., 2014) and some diseases such as atopic dermatitis (Engel-Yeger et al., 2007), asthma (25.7%) (Engel-Yeger et al., 2014) and epilepsy (49%) (Van Campen et al., 2015). It has been estimated that a person with SPD has a 4 times increased risk to develop emotional problems (such as anxiety) and 3 times higher risk to develop external behavioral problems (such as aggressive conducts) (Ben-Sasson et al., 2009).

7. Derived problems
Individuals affected by SPD have problems in learning and academic performance, fine and gross motor coordination, muscle tone, behavior, language, carrying out activities of daily living, disproportionate rejection or pursuit of certain sensory stimuli, distractibility, impulsivity, hyperactivity or hypoactivity, sensorimotor retardation, irritability, malaise, dizziness, anxiety and gastrointestinal problems (Baum et al., 2015; Gourley et al., 2013; Hazen et al., 2014; Zimmer & Desch, 2012). Problems derived from SPD have been shown to be especially harmful to school-aged children, generating chronic stress, low self-esteem and depression, which seriously affect their development during childhood and their personal and professional life as an adult (Bar-Shalita et al., 2008; Chien et al., 2016; Gearhart & Bodie, 2012; Kinnealey et al., 2011). It has also been described that SPD has a negative impact on the social participation of affected ones (Baker et al., 2008).

8. Common signs
Some of the most common signs that Dr. Ayres detected in her observations, which remain important for current professionals dealing with a case of SPD are summarized below, and are based on (Ayres, 2008).

9. Hyperactivity or distractivity
It is one of the most frequent reasons for consultation. The children do not sit still and spend all their time moving. Difficulties exist to connect with activities that involve an effort of concentration or attention in a single objective to realize a task because children are not able or cannot disconnect of all the other stimuli that are present at the same time.

10. Muscle tone and coordination problems
These are highly related to alterations in motor planning or praxis. Coordination problems can be seen in gross or fine motor activities. Some people may have poor balance, while others may have great difficulty in learning how to perform new tasks that require motor coordination.

11. Alterations in language, motor skills or academic performance
These symptoms may be evident in preschool as signs of a deficit sensory integration. At school age, there may be problems in some academic areas despite normal intelligence.

12. Poor organizational behaviour
Children can be impulsive and easily distracted and show a lack of planning when tackling tasks. Some children have difficulty adjusting to a new situation. Others may react with frustration, aggression, flight or rejection when they realize that they fail. They even need to modulate their basal state, and they need to perform actions that from the adult perspective appears to be only behavioral problems or bad behavior.

13. Poor self-esteem
Sometimes, children who experience the above problems do not feel well enough. Intelligent children with these types of problems may know that some tasks are more difficult for them than for other children, but may not know why. These children may seem lazy, bored, or unmotivated. Some children with these characteristics may find ways to avoid those tasks that are hard or embarrassing. When this occurs, children are often considered problematic or stubborn. When
a problem is difficult or incomprehensible, parents and children may both feel guilty and family stress, poor self-concept, and generally the feeling of hopelessness prevails.

14. Candidate biomarkers
SPD symptomatology is commonly present without a neurological injury or acquired brain damage, and therefore does not “appear” in diagnostic imaging tests, which greatly hampers its assessment. In addition, there are currently no validated SPD biomarkers for clinical use; regarding this, only some few studies have been published to date, so this field of knowledge remains challenging. One of the aforementioned studies, published in 2010, showed that children with severe SPD had a lower basal activity of the parasympathetic nervous system compared to children with normotypic development, suggesting that such activity could be a candidate for severe SPD biomarker (Schaaf et al., 2010). Another study, published in 2011, demonstrated by electroencephalography (EEG) that children with SPD showed unique patterns of brain processing that were different from those of children with normotypic development (Gavin et al., 2011). In this study, researchers found that brain activity measured by EEG was able to correctly distinguish children with SPD from non-SPD children with 77% accuracy. They also showed a significant relation between neurophysiological measures and functional performance in sensory and motor tasks between groups. Later, in a work published in 2015, it was observed that children with SPD showed altered white matter microstructure organization (without injury) in posterior cerebral tracts that connect the sensory inputs to the cognitive processing, which was quantified by diffusion tensor imaging (DTI), a magnetic resonance imaging technique (Chang et al., 2015). Thus, according to the author’s opinion, DTI could be a useful tool for the evaluation of children with SPD; however, more research would be required to be validated as such. In 2016, a group of researchers studied the plasma levels of 8-isoprostane and cysteinyl leukotrienes (CysLTs) in a sample of ASD patients with sensory deficits and healthy controls, to measure the predictive value of the above molecules as oxidative stress-related parameters (Qasem et al., 2016). The study concluded that 8-isoprostane and CysLTs could be used as biomarkers for the early recognition of autistic patients with sensory deficits phenotypes; unfortunately, there is no information about 8-isoprostane and CysLTs plasmatic levels from pure or idiopathic SPD samples to date, so we cannot assert whether these molecules are useful biomarkers of this phenotype. Finally, a recent study dated 2017, compared magnetoencephalographic (MEG) imaging-derived indices of auditory and somatosensory cortical processing in 3 groups of children: ASD, SPD without ASD and normotypic development (Demopoulos et al., 2017). This study showed that the SPD group would have an intermediate phenotype between ASD and normotypic development with regard to somatosensory processing that may be detected by MEG-based techniques.

These studies highlight the efforts that scientist and clinicians have been done for the last years to find tools of clinical utility that allow better diagnosis, prognosis and both monitoring SPD evolution and effectiveness of treatments. Despite the previously reported results, the few published studies to date shown several limitations such as low number of participants, low phenotype homogeneity or expensive imaging tools that make difficult for these approaches to become clinically useful biomarkers. However, they represent a very important point of support for the larger and deeper studies searching for SPD biomarkers and physiological basis that are to come in the near future.

15. Treatment
According to Dr. Ayres, Occupational Therapy provides the appropriate knowledge to treat SPD (Ayres, 2008). The role of the occupational therapist (OT) is to generate a relationship with the affected children to develop an environment of trust and fun intended for improve the arousal regulation, increase participation and establish a solid foundation of learning adapted to their nervous system. The purpose is to reduce sensory difficulties in daily life, increase children self-esteem and improve family dynamics and quality of life. In this context, one of the most used OT treatments for SPD is Ayres sensory integration therapy (ASI) or sensory integration therapy (SIT) (Case-Smith & O’Brien, 2009). ASI intervention model is based on three concepts: neural plasticity makes brain change possible, active participation is required for neural changes, and enriched environments are needed to guide neural changes (Ayres, 1972; Miller et al., 2009). In this type of
intervention model, developed specifically for children with SPD, the OT must introduce challenges in which children can move forward and leave behind their difficulties of sensorial modulation and participation, achieving adapted behaviors. The challenges posed by the OT are performed by active exercises that generate an intrinsic motivation that serves as an engine to drive the effort while entertaining (it is very important to keep in mind that children motivation is achieved through play) (Hunt et al., 2017). Thus, through the search for motivated effort, children improve their sensory integration capacities by transferring these learnings to home, playgrounds, school and community (Case-Smith & O’Brien, 2009). The treatment sessions are designed to provide controlled sensory experiences in order to obtain adapted behavior or response, as well as to help the nervous system to efficiently modulate, organize and integrate the information provided (Baranek, 2002). In short, the goal is to generate a “fair challenge” in which children strive but achieve their goal in a fun and at the same time learn to respond to daily activities in a more adapted way (Schaaf & Nightlinger, 2007; Smith et al., 2005).

We should not overlook the specific needs of many children with SPD because of their communication difficulties. Ayres (1972) stated that “communication and language are one of the final products of sensory integration”. For this reason, it is often necessary to perform a combined treatment of SIT and speech therapy. The importance of interdisciplinary collaboration and consultation between the OT and the speech therapist was pointed out for more than 15 years ago (Yack, 1989). Likewise, it is very advisable to train the speech therapists in SIT concepts in order to know and apply them to the design of more complete and effective treatment programs (Iskowitz, 1997). The efficacy of SIT as a treatment for SPD has been widely demonstrated in specific cases and in studies with a relatively low number of participants (Tudela-Torras et al., 2017), nevertheless, its scientific evidence still remains under study (Miller, Coll et al., 2007; Zimmer & Desch, 2012). Figure 4 summarizes the fundamentals of SIT as the main developed therapy for SPD.

Finally, owing to its unknown etiology and the few basic-translational studies with animal models focused on biological or molecular targets and mechanisms (Gogolla et al., 2014; Sinclair et al., 2017), currently, there is no clinically accepted pharmacological treatment for SPD at date.

16. Conclusion

Despite the high prevalence of SPD in children population (around 5–15%), as published by epidemiological studies whose measurement tool was the SP, its comorbidity in about 80–90% of ASD cases and its demonstrated detrimental effects on children development and subsequent adult life, there is a disturbing lack of knowledge about this disorder among many health and educational professionals as well as in families that have to deal with it. This fact makes difficult for the society to become aware of what SPD is, giving rise to inaccurate diagnosis, treatments and interventions of symptomatic children. Additionally, the lack of accepted pharmacotherapy for SPD makes SIT the only available treatment for it. SIT has shown a moderate
beneficial effect for decades; however, the underlying neural mechanisms remain unclear. In our opinion, this is fundamentally due to three aspects: (1) the designing of large studies with participants with comparable sensory profiles is very difficult because of the variable singularities presented by children with SPD; (2) the lack of standardized and internationally accepted methodology for research on SIT (an issue which is currently being address (Schaaf et al., 2015)); and (3) the lack of clinically available biomarkers to improve diagnosis, prognosis and follow-up of treatment and evolution of the disorder. All these characteristics make SPD a disorder that, although its fundamentals remain unclear, is attracting a growing interest among the international scientific community specialized in developmental disorders. This fact will undoubtedly have a positive impact on the number of studies, improving knowledge, treatments and quality of life of those affected children and their families.

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