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Phenomenology of repetitive and restrictive behaviors and sensory phenomena in neurodevelopmental disorders: an exploratory study

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Abstract

Background Repetitive and restrictive behaviors (RRB) include simple motor stereotypes, tics and complex ritualized and rigid behaviors that are core symptoms in neurodevelopmental disorders such as obsessive–compulsive disorder (OCD), Tourette syndrome (TS) or autism spectrum disorder (ASD). Sensory phenomena (SP) are uncomfortable feelings, including bodily sensations, sense of inner tension, “just-right” perceptions, feelings of incompleteness, or “urge-only” phenomena, which have been described to precede, trigger, or accompany RRB. In such clinical contexts RRB and SP may be considered common variables that affect multiple aspects of daily functioning and are treatment targets.

Objective This study aims to further understand RRB and SP phenomenology in children and adolescents diagnosed with OCD, TS or ASD and identify whether specific RRB or SP can distinguish these groups.

Methods We assessed RRB and SP in participants aged between 6 and 17 with a main diagnosis of OCD ($n=23$), TS ($n=19$), or ASD ($n=21$) with the Repetitive Behavior Scale-Revised (RBS-R) and The University of Sao Paulo-Sensory Phenomena Scale (USP-SPS).

Results The RBS-R mean was 17.3 ± 14.9 with no group differences for total RBS-R symptom severity, except for the routine subscale (OCD > ASD, $p=0.03$). Ninety percent of participants showed at least one type of SP on the USP-SPS with a mean total severity of 5.3 ± 3.8 , with no statistical differences between groups. The most frequent SP subtype was physical sensations (68.4%).

Conclusion RRB and SP are transdiagnostic features in neurodevelopmental disorders and the RBS-R and the USP-SPS might be useful in their assessment and treatment plan.

Keywords Neurodevelopmental disorders, Repetitive and restrictive behaviors, Sensory phenomena, Obsessive–compulsive disorder, Tic disorders, Autism spectrum disorder

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Introduction

Restrictive and repetitive behaviors (RRB) are a wide range of responses characterized by their repetition, rigidity, or inflexibility, and by a frequent lack of function or objective [37, 91]. RRB include simple motor stereotypes, tics and more complex ritualized and rigid behaviors such as: compulsions, rituals and routines, insistence on doing any activity in the same way, self-injurious behavior, attachment to specific objects and limited and circumscribed interests that can vary in frequency, intensity and duration [97]. RRB are common in typical child development [21, 23]. Preference to keep ambiances unchanged, object alignment in straight lines, rigid routines and a keen perception of minute detail often occur in children. Some repetitive routines are positively associated with anxiety [31, 55]. These typical RRB may play an adaptive role in the maturation of the motor nervous system and emotional regulation [21] helping the child to achieve a sense of control [107]. Nevertheless, even when ritualistic behavior is normative and adaptive at young age, children at the top of the distribution could be more prone to develop OCD [107], reflecting continuity [9, 106]. De Caluwé et al. proposed an integrative conceptual model based on continuity of such phenomena [16], supported by behavioral-genetic [9] and neuropsychological evidence [70]. RRB are nuclear symptoms in neuropsychiatric and/or neurodevelopmental disorders, such as pediatric-onset obsessive-compulsive disorder (OCD), Tourette syndrome (TS) and autism spectrum disorder (ASD) [63], in which RRB are main diagnostic features [8]. In ASD, the social communication and the repetitive/restrictive behaviors domains are considered two separate symptom areas, probably with different development trajectories [54]. Basal ganglia are central to the development of RRB [33]. Corticostriatal circuits, which function to control goal-directed motor, cognitive and motivational behavior, are involved in OCD [77, 100], TS [93] and ASD [41, 88]. Disruption of those may result in inadequate repetition of a behavioral set (as, inability to switch to other behavior) or facilitation of inappropriate behavioral sets [22]. Therefore, current classification of these disorders might complicate research of RRB and SP [56, 57].

RRB are often preceded by a feeling of incompleteness or other uncomfortable sensations, such as premonitory impulses or the need to perform actions until it feels "just right". These sensory phenomena (SP) are unpleasant feelings, including bodily sensations, sense of inner tension, "just-right" perceptions, feelings of incompleteness, or "urge-only" phenomena, which have been described to precede, trigger, or accompany RRB in OCD and TS [26, 82]. In TS, premonitory urges (PU) are uncomfortable physical sensations preceding tics that can be relieved for

a time by the expression of tics (Rajagopal & Cavanna; [76]). Converging evidence supports the presence of sensory gating dysfunction in such phenomenon [76]. The Premonitory Urge for Tics Scale (PUTS) is a reliable and valid instrument for quantifying PU in adults and in youth (Openneer et al.; Rajagopal & Cavanna; [76, 101]).

Some findings suggest a relationship between sensory sensitivity and excessive ritualism [6, 38, 83], even after controlling for anxiety [15, 89].

Contrary to traditional categorical nosological limits, recent findings indicate that most neurodevelopmental disorders are overlapping syndromes [42] that include a set of dimensional traits that extend to the general population [24]. In some patients, the number of symptoms outreaches the criteria for more than one disorder, although the implications of such comorbidity are still unclear [96]. Additionally, the association between such symptoms may change over time [78].

Overlapping symptoms and comorbidity frequently occur in the three disorders that we aim to study. Pediatric-onset OCD is often accompanied by motor tics [14] and TS is frequently accompanied by obsessive-compulsive symptoms or diagnosed together with OCD [40]. It is common for children diagnosed with TS to be diagnosed with OCD when they reach adolescence (Sanderson et al.). Comorbidity between OCD and TS is so common in a specific group of patients that these two disorders represent a specific subtype of disease, named obsessive-compulsive tic disorder [17]. In ASD, in which RRB are central symptoms, OCD or TS are often diagnosed [45], Kalyva et al.; [69]. Nonetheless, differences between RRB exist, such as ego-dystonic compulsions in OCD and TS that differ from those performed by ASD patients, with more ego-syntonic characteristics, low perception of personal responsibility and low neutralization efforts (Scahill, 2016). Stereotypes, typical in ASD, are extremely rigid, repetitive sequences of behavior, and are most prominent under aversive conditions, including stress, social isolation and sensory deprivation [79]. Thus, stereotypes may not be governed in the same way by reinforcement learning, compared to other RRB [33]. Finally, PUTS items 3,4,5 and 6 (sensation of being 'wound up' because of inner tension, the feeling that something is not 'just right' or complete and the feeling that there is energy in the body that needs to get out in order to get relief) [66], or the revised non-just-right experiences scale (NJRE-QR) [10] showed that different types of these phenomena are more closely related to compulsive symptoms than tics.

Additional to this clinical context, findings from the last decade emphasize genetic overlap between ASD, TS and OCD [12, 35, 46, 102], including the identification of copy number variants that confer risk for these disorders

[59, 65, 84]. Family, twin, and molecular studies suggest some genetic overlap between ASD, OCD and TS [67]. Genome-wide association studies (GWAs) show that these three disorders are highly hereditary and genetically related [25, 99, 103, 104]. Immune and autoimmune pathways also appear to be involved in the development of a set of neuropsychiatric disorders such as pediatric-onset OCD, TS and ASD [2, 4, 32, 64, 94]. Several lines of research suggest that dysfunction of innate immunity can occur in these neuropsychiatric conditions [27, 81, 95].

On this basis, RRB and SP may be considered diverse manifestations along a single continuum versus qualitatively distinct behavioral expressions with different etiological factors [16]. From the intervention perspective, different etiologies may need different treatment approach (e.g. antidopaminergic versus serotonergic pharmacotherapy). Either way, RRB adversely affect multiple aspects of daily functioning in OCD, TS and ASD, and are therefore important targets for clinical intervention [52]. This study aims to further understand RRB and SP phenomenology in children and adolescents diagnosed with OCD, TS or ASD and identify whether specific RRB or SP can distinguish these groups.

Methods

Patients and methods

We gathered a sample of patients aged 6–17 years from the outpatient child and adolescent psychiatry department of Hospital Clinic (Barcelona, Spain), with a main diagnosis of OCD ($n=23$), TS ($n=19$), or level 1 ASD ($n=21$) established according to the criteria of the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5). “Main diagnosis” refers to the diagnosis for which the patient and their family were consulting the department, and which caused the greatest interference in their functioning (although comorbidities could also be diagnosed during follow-up). All these patients were treated in tertiary-level units of OCD, TS and ASD of the child and adolescent psychiatry department of Hospital Clinic. They were invited to participate in the study between mid 2020 and mid 2023.

Procedure

The study was approved by the Hospital Clinic ethics committee, and the parents or guardians of all patients (and those patients from the age of twelve) gave written informed consent prior to assessment. Children under 12 years of age gave written assent. All patient data were anonymized in compliance with Spanish data protection legislation.

A list of eligible patients from the OCD, TS and ASD specialized units was made, and they were invited to participate. All participants were evaluated when they

consented to participate in the study by experienced specialists in child and adolescent clinical psychology or psychiatry from Hospital Clinic (not necessarily when they were at their worst clinical period). In all groups, we excluded individuals with an intelligence quotient (IQ) below 80, psychotic disorder or disabling neurological disease. All participants had no difficulty using language. Comorbidity data for OCD, TS, and ASD groups were provided by their regular physicians (psychiatrists or clinical psychologists) via clinical records, as screening for all mental disorders is performed during the first consultations. The test battery applied has an approximate duration of 3 h, and includes the Repetitive Behavior Scale-Revised (RBS-R) [8], The University of Sao Paulo-Sensory Phenomena Scale (USP-SPS) [82], the Children's Yale-Brown Obsessive Compulsive Scale (CY-BOCS) [86] for patients with OCD, the Yale Global Tic Severity Scale (YGTSS) [58] for patients with TS, the ADOS (Autism Diagnostic Observation Schedule) [60] for patients with ASD, and the Clinical Global Impression Severity Scale (CGI) [36]. The CY-BOCS, YGTSS and the USP-SPS instruments were administered to children and adolescents, but parents were present in the assessments and their input was taken into consideration. Although RBS-R is a self-report, participants completed it at the assessment appointment so they or their parents could ask any doubts.

Assessment instruments

Children's Yale-Brown Obsessive–Compulsive Scale (CY-BOCS) [86, 98]

The CY-BOCS is the gold standard instrument for assessing the severity of OCD in children. It is a semi-structured clinical scale comprising 10 items (5 on obsessions and 5 on compulsion/rituals) that evaluate time, interference, distress, resistance, and control of symptoms. The maximum score for each item is 4 points (maximum total score of 40). Severity of OCD is classified as sub-clinical (≤ 10 points), mild (11–16), moderate (17–24), severe (25–32), or extreme (33–40). The validation study in a Spanish population found a Cronbach alpha of 0.87. Administration time ranges from 45 to 60 min.

Yale Global Tic Severity Scale (YGTSS) [29, 58]

The YGTSS is the gold standard instrument for assessing the severity of TS. This semi-structured clinical scale begins with a semi-systematic inventory of tic symptoms, which the clinician marks as being present or absent over the past week and at the worst ever moment of the disease. Current motor and verbal tics are evaluated separately according to number, frequency, intensity, complexity, and interference, on a 6-point ordinal scale (0: absent; 1–5: level of severity),

yielding 3 scores: total motor tic score, total verbal tic score, and total tic score (sum of the previous 2 scores, with a maximum of 50). Finally, the scale provides a global severity score (0–100), which is the sum of the total tic score (0–50) and the overall impairment rating (0–50). Severity is classified as subclinical (≤ 10 points), mild (11–20), marked (21–40), or severe (> 40). The validation study in a Spanish population found a Cronbach alpha of 0.99 for both dimensions (motor tics and phonic tics). Administration time ranges from 45 to 60 min.

Autism Diagnostic Observation Schedule (ADOS) [60, 61]

ADOS is a semi-structured observational assessment designed to evaluate aspects of communication, social interaction, play, and stereotyped behaviors and restricted interests. Depending on an individual's language level and age, certified staff in ADOS administration (e.g., clinicians, psychologists, research staff) administered to participants one of several modules (modes of implementation) of ADOS. Participants in this study received ADOS-2, module 3 for those more verbally fluent and older children, as module 4 for adolescents with fluent speech. Based on the scoring of the ADOS-2, participants receive a classification of autism, ASD, or non-spectrum, as well as a standardized severity score and a RRBs score. All ADOS-2 examiners were certified research-reliable in both administration and scoring. The assessment takes around 1 h to complete.

Repetitive Behaviors Scale – Revised (RBS-R) [8, 62]

The RBS-R is a 43-item self-report questionnaire that is used to measure the breadth of repetitive behavior in children, adolescents, and adults with autism spectrum disorder. The RBS-R provides a quantitative, continuous measure of the full spectrum of repetitive behaviors. The RBS-R consists of six subscales including: Stereotyped Behavior, Self-injurious Behavior, Compulsive Behavior, Routine Behavior, Sameness Behavior, and Restricted Behavior, that have no overlap of item content. This permits differential identification and scoring of discrete varieties of repetitive behaviors. The assessment of repetitive behaviors is performed based on the observations and interactions during the past month of a family member, caregiver or professional who knows the person well. Behaviors are rated on a 4-point scale: 0-Behavior does not occur, 1-Behavior occurs and is a mild problem, 2-Behavior occurs and is a moderate problem, 3-Behavior occurs and is a severe problem. Time to complete takes from 10 to 15 min.

University of São Paulo's Sensory Phenomena Scale (USP-SPS) [82]

This is a semi-structured scale developed to investigate the presence and severity of different types of SP occurring before or during the performance of repetitive behaviors. The USP-SPS is divided in two parts, a checklist, and a severity scale. The USP-SPS checklist is composed of items assessing past and current examples of different types of SP and encompasses all previous descriptions of SP in the literature. If symptoms were endorsed, patients were also asked to provide their age of onset. Subtypes of SP included:

1. Physical sensations (tactile and/or muscle-joint): uncomfortable sensations localized in a specific region of the body (skin, muscles, or joints) which precede or occur along with repetitive behaviors. Subjects also frequently report that there is a momentary sense of relief once the repetitive behaviors have been performed.
2. “Just right” perceptions triggered by tactile, visual, or auditory sensations including the need for things to: feel “just right”—a need to touch objects or people until it feels “just right”; look “just right”—a need for objects to look a certain way (e.g., perfectly symmetrical); and/or to sound “just right”—a need for a person's voice or an audio recording to sound “just right” or have the “just right” pitch.
3. Feeling of incompleteness/need to feel “just right” internally: an inner feeling and/or perception of discomfort that makes the patient do things until feeling relieved. To have an inner sense of not being “just right” and a necessity to repeat a behavior until getting the “just right” feeling.
4. Energy release: a generalized inner tension or energy that builds up and needs to be released by doing a movement or action.
5. Urge only: no sensations or feelings, just an urge to do the repetitive behavior.

The USP-SPS severity scale measures the severity of the SP by three ordinal scales with six anchor points that focus on the frequency of the SP (0–5), the amount of distress they cause (0–5), and the degree to which they interfere with the patient's functioning (0–5). The total score is obtained by combining the scores for frequency, distress, and interference, ranging from 0–15. Scores are obtained both for current severity and the time when the SP were at their worst severity. Administration time ranges from 10 to 15 min.

Clinical Global Impression Severity Subscale (CGI) [36]

The CGI comprises two companion one-item measures evaluating the following: (a) the CGI Severity Subscale that measures severity of psychopathology from 1 to 7 and (b) the CGI Improvement Subscale that measures change from the initiation of treatment on a similar seven-point scale. For the current study only the CGI Severity Subscale was used. Subsequent to a clinical

evaluation, the CGI form can be completed in less than a minute by an experienced rater. In practice, the CGI captures clinical impressions that transcend mere symptom checklists. It is readily understandable and can be used with relative ease by the non-researcher clinician. The CGI is rated on the following seven-point scale: 1 = normal, not at all ill; 2 = borderline mentally ill; 3 = mildly ill; 4 = moderately ill; 5 = markedly ill; 6 = severely ill; 7 = among the most extremely ill patients. This rating is based upon observed and reported symptoms, behavior, and function in the past seven days.

Statistical analysis

Continuous variables were presented as the mean value \pm standard deviation and to compare means between groups we used the Student's *t* test or Wilcoxon rank sum test as appropriate. ANOVA was used for comparisons with more than two groups. Categorical variables were expressed as total number with percentages

and compared between groups using Pearson's Chi-squared test or Fisher's exact test as appropriate. Logistic regression was used for statistical comparisons for binary outcomes. Statistical comparisons were done with emmeans library (v. 1.8.2). All analyses were addressed considering a two-tailed type 1 error of 5%. All analyses and figures were done using R (v. 4.3.2).

Results

Descriptive statistics

Table 1 displays descriptive statistics from the sample. Mean age in the total sample was 14.0 and sixty-three percent of the total sample were male (same gender), with no statistically significant differences between the 3 groups. The TS group showed greater proportion of comorbidities (68%), followed by the ASD group (67%) and the OCD group (48%), although the difference did not reach statistical significance. Most of the sample (65%) received combined treatment (CBT plus

Table 1 Descriptive statistics of the participants

	Total sample (<i>n</i> = 63)	OCD (<i>n</i> = 23) (37%)	TS (<i>n</i> = 19) (30%)	ASD (<i>n</i> = 21) (33%)	<i>P</i> -value
Age (years)	14.0 (2.7)	14.4 (2.4)	13.9 (2.8)	13.8 (2.9)	0.83
Sex (male)	40 (63%)	12 (52%)	13 (68%)	15 (71%)	0.36
Presence of any comorbidity	38 (60%)	11 (48%)	13 (68%)	14 (67%)	0.11
Type of treatment					0.05
CBT	16 (25%)	10 (43%)	2 (11%)	4 (19%)	
MEDICATION	6 (9.5%)	1 (4.3%)	4 (21%)	1 (4.8%)	
CBT + MEDICATION	41 (65%)	12 (52%)	13 (68%)	16 (76%)	
Number of medications					0.24
None	16 (25%)	10 (43%)	2 (11%)	4 (19%)	
Monotherapy	21 (33%)	6 (26%)	8 (42%)	7 (33%)	
Two	18 (29%)	6 (26%)	6 (32%)	6 (29%)	
Three	8 (13%)	1 (4.3%)	3 (16%)	4 (19%)	
Type of medication					0.00
Antipsychotic	31 (49%)	5 (22%)	14 (74%)	12 (57%)	
SSRIs	29 (46%)	13 (57%)	6 (32%)	10 (48%)	
Methylphenidate	5 (8%)	-	4 (21%)	1 (5%)	
Adrenergic agonist	5 (8%)	-	3 (16%)	2 (10%)	
Benzodiazepines	4 (6%)	2 (9%)	2 (11%)	-	
Atomoxetine	3 (5%)	1 (4%)	-	2 (10%)	
Mood stabilizer	2 (3%)	-	-	2 (10%)	
Tricyclic antidepressant	2 (3%)	-	-	2 (10%)	
CYBOCS (<i>n</i> = 32; OCD = 23, TS = 5; ASD = 4)	20.4 (7.5)	19.3 (7.8)	20.6 (6.4)	27.0 (3.8)	0.10
YGTSS (<i>n</i> = 19)	-	-	25.7 (11.2)	-	
ADOS (<i>n</i> = 15)	-	-	-	10.9 (3.0)	
CGI Severity Subscale	5.0 (1.2)	4.7 (1.4)	5.6 (0.9)	4.6 (0.9)	< 0.01

ADOS autism diagnostic observation schedule, ASD Autism spectrum disorder, CBT cognitive-behavioral therapy, CGI Clinical Global Impression, CYBOCS Children's Yale-Brown Obsessive-Compulsive Scale, OCD obsessive-compulsive disorder, SSRIs Selective serotonin reuptake inhibitors, TS Tourette syndrome, YGTSS Yale Global Tic Severity Scale

medication) whereas 25% of the total sample did not take any psychopharmacological treatment. Atypical antipsychotic was the most prescribed medication (49%), followed by selective serotonin reuptake inhibitors (SSRIs) (46%) with significant differences between groups ($p=0.002$). The CY-BOCS mean showed moderate severity except within the ASD group (marked severity) and the YGTSS mean showed marked severity. The CGI Severity Subscale in the TS group showed

significant differences with the other two groups ($p<0.01$).

Table 2 presents comorbidities identified in the three patient groups as percentages, which statistically differed (0.02). The most frequent comorbidities were anxiety disorders in the three groups, attention-deficit/hyperactivity disorder (ADHD) in the TS and ASD groups, and OCD in the TS group.

Phenomenology of repetitive and restrictive behaviours as evaluated with the Repetitive Behaviour Scale-Revised (RBS-R)

Ninety-eight per cent of the total sample showed presence of RRB in the RBS-R. The RBS-R mean was 17.3 ± 14.9 with no group differences for total RBS-R symptom severity. The group scoring highest in the total score (sum of all six subscale scores) was the ASD group (20.6 ± 14.4), followed by the TS group (16.6 ± 14.8) and the OCD group (15 ± 15.6), although no significant differences between groups were observed. Within the whole sample the sameness behavior showed the greatest score, followed by the compulsive, the routine, the self-injurious, the stereotyped and the restricted behavior. As depicted in Fig. 1, the ASD group scored highest in the self-injurious behavior (4.7 ± 3.1 in the ASD group; 3.3 ± 2.0 in the TS group; 2.5 ± 1.7 in the OCD group), routine (6.3 ± 4.1 in the ASD group; 4.0 ± 4.1 in the TS group; 3.0 ± 2.3 in the OCD group), sameness (8.9 ± 5.6 in

Table 2 Comorbidities in the three groups in the study sample

	OCD (n=23)	TS (n=19)	ASD (n=21)	p-value
				0.02
OCD	-	7 (37%)	4 (19%)	
ASD	1 (4%)	-	-	
ANXIETY DISORDER	7 (30%)	5 (26%)	5 (24%)	
TS or CTD	3 (13%)	-	-	
ADHD	4 (17%)	6 (32%)	6 (29%)	
DEPRESSIVE DISORDER	-	1 (5%)	3 (14%)	
ODD	-	2 (11%)	-	
LEARNING DISORDER	1 (4%)	3 (16%)	1 (5%)	

ADHD Attention-Deficit/Hyperactivity Disorder, ASD Autism spectrum disorder, CTD Chronic tic disorders, OCD obsessive-compulsive disorder, ODD Oppositional defiant disorder, TS Tourette syndrome

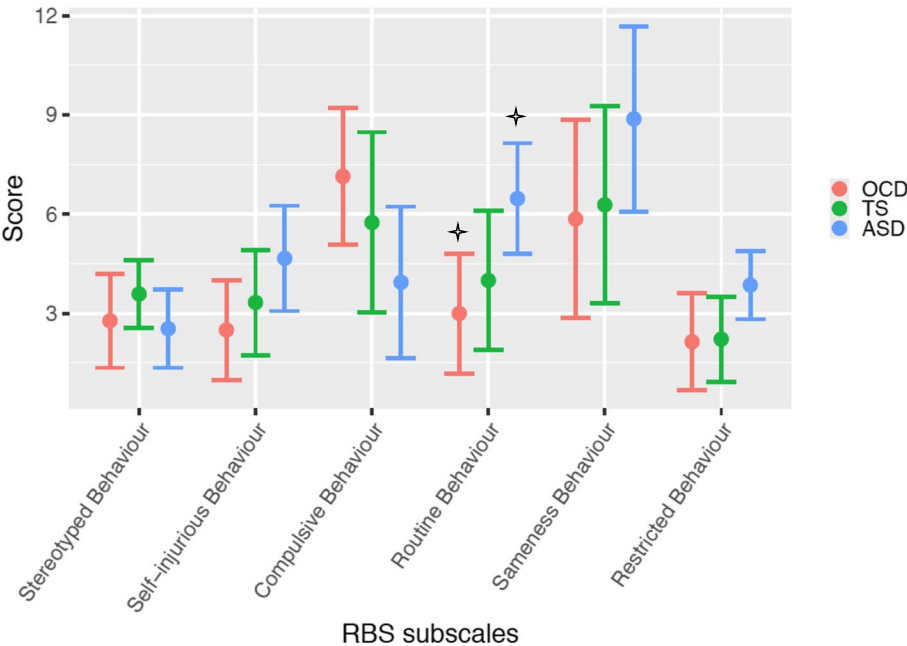


Fig. 1 Restrictive and repetitive behaviors as evaluated with the Repetitive Behavior Scale-Revised (RBS-R). Abbreviations: ASD: Autism spectrum disorder; OCD: obsessive-compulsive disorder; RBS-R: Repetitive Behavior Scale-revised; TS: Tourette syndrome: Significant difference in the routine behavior subscale between ASD and OCD groups ($p=0.03$)

the ASD group; 6.3 ± 4.2 in the TS group; 5.9 ± 6.5 in the OCD group) and restricted behavior subscales (3.5 ± 2.0 in the ASD group; 2.2 ± 1.7 in the TS group; 2.1 ± 1.8 in the OCD group). The OCD group scored highest in the compulsive behavior subscale (7.1 ± 5.4 in the OCD group; 5.8 ± 5.2 in the TS group and 3.9 ± 3.1 in the ASD group). The TS group scored highest in the stereotyped behavior subscale (3.6 ± 2.0 in the TS group; 2.8 ± 2.3 in the OCD group and 2.5 ± 2.0 in the ASD group). Among the RBS-R six subscales, significant differences were observed in the routine behaviour subscale between the ASD and the OCD group ($p = 0.03$).

Phenomenology of sensory phenomena as evaluated with the USP-SPS University of São Paulo's Sensory Phenomena Scale

Ninety-one per cent of the sample reported at least 1 type of SP lifetime preceding and/or accompanying repetitive behaviors (87% of the OCD group, 100% of the TS group and 86% of the ASD group). The five types of SP did not show significant differences between the three diagnostic groups. The most frequent subtype of SP in the whole sample was physical sensations (69.8%). Physical sensations, “just right” perception and feeling of incompleteness scored highest within the OCD group, whereas energy release and “urge-only phenomena” did in the TS group. These results are summarized in Table 3. Among patients with SP, the mean total severity of SP was 5.3 (3.8), with no significant differences between groups.

Figure 2 depicts the overlap between types of sensorial phenomena according to the USP-SPS. Within the whole group ($N = 63$) five cases (9% of the sample) reported the presence of all SP and seven cases (12% of the sample) reported all SP except urge SP. Physical sensation was the most overlapped with other SP in contrast with feeling of incompleteness that showed less overlap with other SP.

Correlations between the RBS-R and the USP-SPS scales and demographic/clinical variables

The CYBOCS showed positive correlations with the RBS-R total score ($r = 0.44$; $p = 0.01$) and the USP-SPS severity score ($r = -0.43$; $p = 0.02$). Though, the ADOS was negatively correlated with the RBS-R total score ($r = -0.52$; $p = 0.04$), the sameness behavior RBS-R subscale ($r = -0.79$; $p = 0.004$) and the USP-SPS severity score ($r = -0.67$; $p = 0.006$). When social affect and RRB domains were analyzed separately, the social affect domain was negatively correlated with the sameness behavior RBS-R subscale ($r = -0.76$; $p = 0.007$) and the USP-SPS severity ($r = -0.56$; $p = 0.029$). The RRB domain did not correlate significantly with any of the outcomes. No significant correlation was found between the outcomes and the YGTSS. The RBS-R total score and the USP-SPS severity scale showed a moderate positive correlation (0.35 ; $p = 0.004$).

Significant correlations were not found between the outcomes and sex or age.

Table 3 Sensory Phenomena evaluated with the USP-SPS University of São Paulo's Sensory Phenomena Scale

	Total ($n = 63$) mean (SD)	OCD ($n = 23$) mean (SD)	TS ($n = 19$) mean (SD)	ASD ($n = 21$) mean (SD)	<i>p</i> -value
USP-SPS severity	5.3 (3.8)	5.7 (4.1)	6 (3.8)	4.1 (3.2)	OCD-TS: 0.97 OCD-ASD: 0.34 TS- ASD: 0.27
	Total <i>n</i> (%)	OCD <i>n</i> (%)	TS <i>n</i> (%)	ASD <i>n</i> (%)	<i>p</i>-value
Physical sensations	44 (69.8)	17 (73.9)	14 (73.7)	13 (61.9)	OCD-TS: 0.99 OCD-ASD: 0.67 TS- ASD: 0.71
“Just right” perception	37 (58.7)	16 (69.6)	9 (47.4)	12 (57.1)	OCD-TS: 0.32 OCD-ASD: 0.67 TS- ASD: 0.81
Feeling of incompleteness	25 (39.7)	11 (47.8)	7 (36.8)	7 (33.3)	OCD-TS: 0.76 OCD-ASD: 0.59 TS- ASD: 0.97
Energy release	26 (41.3)	10 (43.5)	10 (52.6)	6 (28.6)	OCD-TS: 0.83 OCD-ASD: 0.56 TS- ASD: 0.28
“Urge-only” phenomena	17 (27)	7 (30.4)	7 (36.8)	3 (14.3)	OCD-TS: 0.90 OCD-ASD: 0.42 TS- ASD: 0.25

ASD Autism spectrum disorder, OCD obsessive-compulsive disorder, RBS-R Repetitive Behavior Scale-revised, SD standard deviation, TS Tourette syndrome, USP-SPS University of São Paulo's Sensory Phenomena Scale

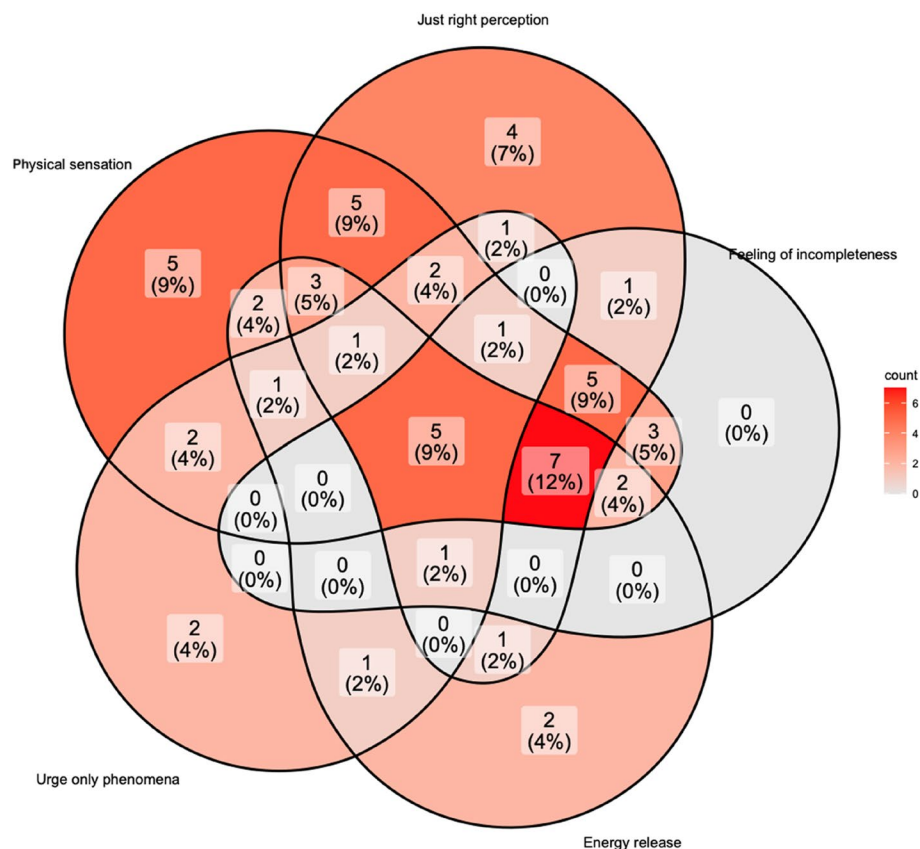


Fig. 2 Venn diagram of SP as evaluated with the USP-SPS

Discussion

Although RRB are well studied in ASD [3, 19, 28, 48, 49, 62] and literature on sensory phenomena in TS and OCD is rapidly expanding [7, 10, 72], our study of pediatric populations addressing RRB and SP in these three groups of patients together advances the knowledge in the field.

The primary objective of this study was to further understand RRB and SP phenomenology in a clinical sample of children and adolescents with a lifetime diagnosis of OCD, TS or ASD using two different instruments, the RBS-R and the USP-SPS. Most results are consistent with the main characteristics of these three disorders definition and with what it is clinically observed, although only one difference between groups was statistically significant, probably due to the small sample size. The compulsive behavior subscale, which items include main OCD symptom dimensions (washing/cleaning, completeness, checking, counting, hoarding, repeating, touching/tapping) scored highest in the OCD group. In contrast, the self-injurious, routine, sameness and restricted subtypes showed higher rates in the ASD group, being the routine and sameness subtypes those with higher scores. That might be explained because only ASD patients with

level 1 of severity according to DSM-5 were included in this study, which is consistent with the study by Jasim and Perry which found that high need of support ASD groups showed higher rates of RBS-R subtypes except for Routine/Sameness [48]. Another explanation may be that motor behaviors improve with age whereas rigidities, insistence on routine and difficulty in adapting to changes are more stable over time. An unexpected finding was that stereotyped behavior was higher in the TS group than in the ASD group. Again, one possible reason for these results is that stereotypes are more prevalent in ASD patients with high need of support [19] or that the stereotype subscale items (defined as *apparently purposeless movements or actions that are repeated in a similar manner*) could be confounded by tics. Also, along verbal developmental stages, obstacles resulting from ASD are more frequently due to social communication problems rather than to RRB, as might be the case in this study. RRB and SP were shown to be moderately correlated with OCD symptom severity, whereas unexpectedly they were strongly negatively correlated with the ADOS. According to the high rates of RRB observed with the RBS-R scale within the ASD group, one hypothesis could be that RRB

may exist in patients' daily life even though they could not be captured with the ADOS-2 administration, which is a semi-structured instrument. Also, social communication deficits could end up in more isolation, turning into avoidance of the stress derived from social environments and, therefore, leading to reduction of RRB. Response bias due to a self-report instrument might be plausible as well. The positive correlation between the RBS-R total score and the USP-SPS severity scale could be explained because sensory dysfunction is distressing and upsetting, therefore children could seek, consequently, ways to calm down, create order and predictability through excessive ritualistic behaviors [15]. However, ASD cases exhibited higher scores in self-injurious, sameness, and restricted behaviors, consistent with established literature.

We did not find significant correlations between the outcomes and age and sex. Across studies, RRB assessed with the RBS-R showed mixed findings regarding correlation with sex [3, 48]. Regarding age, RRB seem to appear in patterns that change over development (more frequent and severe in younger compared to older individuals [19]), and the type of RRB differ between childhood and adolescence. Some studies, though, found no age correlation with RRB [28, 49].

The second objective was to identify whether specific RRB or SP can distinguish these groups. Our results show that, overall, ASD cases showed higher rates in the RBS-R whereas OCD and TS patients did in the USP-SPS. However, Prato et al. recently found that SP assessed with the USP-SPS were significantly more represented in the ASD-group than in the TS-group, except for sound just-right perceptions and energy release [73]. In our sample physical sensations, "just right" perception and feeling of incompleteness scored highest within the OCD group, whereas energy release and "urge-only phenomena" did in the TS group. Several studies reported the association of SP in patients with OCD and TS [26, 71, 72]. Particularly, feeling of incompleteness and non-just-right experiences (NJRE) have been vastly studied in patients with OCD [90] and TS or other tic disorders [10, 73], suggesting they are more related to compulsions than tics. A factorial-network analysis of a large multinational cohort found feeling of incompleteness to be the most central dimension which could represent a core phenotype in OCD [13]. One study showed that SP severity was positively related to hyperactivation of the insula but dissociable from other symptoms in OCD [11]. Nevertheless, according to our results, RRB and SP seem to be correlated with each other and with OCD severity. RRB and SP may be considered transdiagnostic constructs in neurodevelopmental disorders with overlapping and predisposing neural substrates [71], as previous research suggests [56, 57]. Empirical findings relate

SP to sensorimotor deficits [43, 44] and to too sensible mechanism of error detection [80], hypothesizing that early severe motor deviances could facilitate the emergence of OCD symptoms along developmental years [71].

Nonetheless, the characteristics of this sample need to be considered to further understand the results. First, all these patients were treated in tertiary-level units of OCD, TS and ASD and had moderate to severe symptoms at referral. Unfortunately, not all were at the same point of their clinical evolution when recruited and assessed. Additionally, 12.5% of the OCD group met criteria for TS or chronic tic disorder, 35% of the TS group was diagnosed with OCD and 19% of the ASD group was diagnosed with OCD, as observed in Table 2. These complex clinical phenotypes were consistent with the complex pharmacological treatments described in Table 1 and the frequency of RRB and SP observed across the sample. This clinical sample showed high rates of RRB in the RBS-R even when 46% of participants were on SSRIs. Despite SSRIs are the first-line evidence-based drug for OCD [92], 21% of the OCD group also was on atypical antipsychotics, which has been evidenced as a useful augmentation strategy in severe cases [1]. The TS group, with comorbid OCD in 35% of the sample, received atypical antipsychotics in 75% and SSRIs in 30% of the cases. That is in line with the literature that has shown that obsessive-compulsive behaviors in TS cases respond best to a combination of an SSRI and a dopamine antagonist [47]. In the ASD group, 48% were on SSRIs (19% of the sample also met criteria for OCD) and 57% on atypical antipsychotics. Although SSRIs are frequently used in clinical practice, there is limited evidence supporting the efficacy of SSRIs for RRBs in children with ASD (Herscu et al.; [51, 105]) and modest effect of atypical antipsychotics in repetitive behavior in ASD [105]. Finally, almost one-third of the sample suffered from an anxiety disorder. Anxiety might play an important role in all three disorders since such behaviors may relieve stress and anxiety [5, 21, 34] and that diagnosis justifies pharmacological treatment with SSRIs.

The main limitation of this study is the small sample size, which reduces its statistical power. We acknowledge that a greater sample size could have shed light on potential differences among groups. Also, the cross-sectional design is of limited validity. Another limitation is that we did not obtain neuropsychological variables (e.g., IQ, executive dysfunction) which could have contributed elucidating the relationship between neurobiological circuitry and repetitive behavior, since research has suggested that the severity of RRB is inversely correlated to IQ in ASD [30] and child's maturation [20]. Finally, the cross-sectional design of this study and the period when it was performed (post-pandemic) captured RRB and SP

at different time-points of these patients' clinical evolution, also within diagnostic categories. Even though all patients came from tertiary units the results show that the group with TS was worst, clinically wise, compared to the other two groups.

Our results support the hypothesis of considering RRB and SP as symptomatic dimensions in neurodevelopmental disorders, as it has been suggested with social cognition [74] or with sensory over-responsivity [43, 71]. RRB and SP share a common neural basis in the operation of cortico-basal ganglia circuits and sensorimotor systems [33, 71]. This theory would be in line with the hypothesis proposed by De Caluwé [16] and recent developments on the classification of psychopathology, such as the Hierarchical Taxonomy of Psychopathology (HiTOP) [53]. Then, RRB and SP could be acknowledged on a continuum that would allow comparison between individuals affected with neurodevelopmental disorders rather than between categories. Additionally, an evaluation of RRB and SP impairment level could be of value considering their widespread presence across disorders.

In the light of all the above, future research should track the developmental trajectory of RRB and SP to better understand their stability, progression, and response to interventions, since corticostriatal circuits have a protracted developmental trajectory [18] and neurobiological changes associated with RRB and SP may change over development and treatment. It would be valuable the use of genetic and immunological data in addition to these clinical assessments of RRB and SP in patients with neurodevelopmental disorders, to further comprehend the genetics of this phenomena and its relationship with immune and autoimmune pathways. Such research could contribute elucidating whether these phenomena across disorders fit in the continuity model. The most relevant clinical implication of our results is, statistics-wise, that the RBS-R and the USP-SPS instruments could not distinguish specific RRB and SP between OCD, TS and AS, except for routine behavior in the RBS-R (OCD > ASD, $p=0.03$). However, ASD patients were better represented by the RBS-R scale and OCD and TS patients by the USPS-SPS scale. Based on our results, we may conclude that RRB and SP might be worth exploring in neurodevelopmental disorders with a dimensional approach that focuses on the neurobiological basis of such behaviors. These instruments may help to further comprehend symptoms which can help structuring interventions.

Authors' contributions

Author Contributions: Ms.A. Vigil-Pérez had full access to all the data in the study and take responsibility for the integrity of the data and the accuracy of the data analyses. Study concept and design: L. Lázaro, R. Calvo Escalona, A. Morer, A. Vigil-Pérez. Acquisition, analysis, or interpretation of data: All authors. Drafting of the manuscript: Vigil-Pérez, L. Lázaro, R. Calvo Escalona, A. Morer

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Data availability

The data that support the findings of this study are available on request from the corresponding author.

Declarations

Ethics approval and consent to participate

The study was approved by the Hospital Clinic ethics committee, and the parents or guardians of all patients (and those patients from the age of twelve) gave written informed consent prior to assessment. Children under 12 years of age gave written assent. All patient data were anonymized in compliance with Spanish data protection legislation.

Competing interests

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