Running head: ADHD AND SLD VINELAND-II PROFILE

Application of the Repetitive Behavior Scale-Revised – Italian Version – In Preschoolers With Autism Spectrum Disorder

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Abstract

Restricted repetitive and stereotyped patterns of behavior, interests, and activities (RRB) are mandatory features for a diagnosis of Autism Spectrum Disorder (ASD) according to the Diagnostic and Statistical Manual of mental disorders-fifth edition (DSM-5). Despite the strong diagnostic role of RRB, their expressiveness and their relationship with other clinical/demographic features in ASD is not fully elucidated. The Italian version of the Repetitive Behavior Scale-Revised (RBS-R) was applied to a relatively large sample of preschool-aged children with ASD who underwent a comprehensive clinical assessment. The relationship between RRB and sex, age, nonverbal IQ, autism severity, as well as the diagnostic accuracy of the RBS-R were explored. Stereotyped and Ritualistic/Sameness behaviors were the most common RRB in preschoolers with ASD, without widespread differences between males and females. No significant correlations between RRB and chronological age, or non-verbal IQ were detected. The expressiveness of ritualistic/sameness behaviors positively correlated with autism severity, assessed through the Calibrated Severity Score (CSS) derived from the Autism Diagnostic Observation Schedule (ADOS). Receiver Operator Characteristic (ROC) analysis showed high diagnostic accuracy using the Global Rating Score, which represents the judgment of the parents of as the RRB affect the child's life. However, while the Global Rating Score performed well, the remaining subscales did not. This investigation extends the limited research on early pattern and associated features of RRB in young children with ASD. The use of the RBS-R may increase the knowledge of the RRB complexity and variability and in turn improve the diagnostic and therapeutic procedures within the autistic spectrum.

Keywords: Restricted and repetitive behaviors (RRB), Autism spectrum disorders (ASD), preschoolers, Repetitive Behaviors Scale-Revised (RBS-R), diagnostic accuracy

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1. Introduction

Restricted repetitive and stereotyped patterns of behavior, interests, and activities (RRB) are mandatory features for a diagnosis of Autism Spectrum Disorder (ASD) according to the Diagnostic and Statistical Manual of mental disorders-fifth edition (DSM-5; American Psychiatric Association, 2013), along with the social-communicative impairments. Despite their strong diagnostic significance, most of the research on core symptoms of ASD has been directed to explore the socialcommunicative impairment rather than RRB and therefore RRB profiles and associated features within ASD have not yet been fully elucidated.

The lower interest for RRB could be due primarily to their supposed poor specificity and underestimated diagnostic role in ASD. Indeed, RRB are not unique to ASD, but evident both in other clinical populations and in typical development (TD) (for recent reviews, see Langen, Durston, Kas, van Engeland, & Staal, 2011; Leekam, Prior, & Uljarevic, 2011). Moreover, according to the previous DSM (DSM-IV-TR; American Psychiatric Association, 2000), RRB were not mandatory, but just ‘‘possible’’ feature in the large Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS) category.

Second, the assumption that RRB appeared after the onset of the social-communicative impairment (Charman & Baird, 2002; Osterling, Dawson, & Munson, 2002) might have contributed to minimize their role in programs for early detection of ASD. However, children with ASD, since a very early age, demonstrate RRB with objects, body, and sensory behaviors more frequently and for longer periods of time than children with TD matched on chronological age (Watt, Wetherby, Barber, & Morgan, 2008). Moreover, a higher frequency and a greater diversity of RRB in young children with ASD than age-matched TD have been recently detected (Harrop, McConachie, Emsley, Leadbitter, & Green,

2013). Repetitive actions with objects and repetitive movements of the body/arms/hands have been described as warning symptoms for a diagnosis of ASD in the second year of life (Wetherby et al., 2004). More recent studies supported that RRB may be among the earliest behavioral manifestations of ASD (Kim & Lord, 2010; Ozonoff et al., 2008). In particular, Ozonoff et al. (2008) reported an atypical way of object exploration in one year old children subsequently diagnosed with ASD, whereas Wolff et al. (2014) highlighted that as early as 12 months of age a broad range of repetitive behaviors frequently occurred in toddlers who will receive an ASD diagnosis. Although data should be interpreted with caution and require further investigations, some RRB could be ASDspecific, since children with TD rarely roll or wobble objects, or demonstrate careful placement of objects or spinning objects (Barber, Wetherby, & Chambers, 2012; Watt et al., 2008). Therefore, even if RRB have been neglected for a long time, the interest for these symptoms is growing for the support in early identification of ASD behavioral markers.

Usually, RRB refer to a broad range of behaviors. Turner (1999) categorized this wide spectrum of behaviors into ‘‘lower level’’ characterized by repetition of movement including stereotyped movements, repetitive manipulation of objects, and repetitive forms of self-injurious behavior, and ‘‘higher level’’ including object attachments, insistence on sameness, repetitive language, and circumscribed interests. Lower level behaviors have been found associated with lower cognitive abilities, poorer adaptive skills and younger chronological age whereas higher level behaviors have been shown either no relationship or positive relationships with the same variables (Leekam et al., 2011).

Association between RRB and socio-communicative features of ASD is a controversial issue: some researchers have proposed a dissociation between the socio-communicative and RRB (Happe´, Ronald, & Plomin, 2006), whereas others researchers found a link between RRB and social deficits measured by the ADOS-G (Watt et al., 2008).

Also sex differences in RRB expression were explored with some contrasting findings: a greater level of RRB in males (Sipes, Matson, Worley, & Kozlowski, 2011; Szatmari et al., 2012; Mandy et al., 2012), and an absence of sex differences (Andersson, Gillberg, & Miniscalco, 2013; Joseph, Thurm, Farmer, & Shumway, 2013) have been reported. A recent systematic review and metanalysis is that examined the impact of sex on the core ASD symptoms described more RRB in boys only if they were over six years of age (Van Wijngaarden-Cremers et al., 2013).

As far as the RRB assessment tools, several methods of measuring RRB in ASD were used, including caregiver interviews or questionnaires and observational methods. The most widely used and validated measure is the ‘‘Restricted interests and repetitive behaviors’’ section of Autism Diagnostic Interview-Revised (ADI-R; Rutter, Le Couteur, & Lord, 2003), even though other instruments have been used including the Yale-Brown Obsessive-Compulsive Scale (Y-BOCS; Goodman et al., 1989a,b) and its adapted version for Children (CY-BOCS; Scahill et al., 1997), the Yale Special Interests Interview (YSII; South, Klin, & Ozonoff, 1999), the Childhood Routines Inventory (CRI; Evans et al., 1997), the Repetitive Behavior Questionnaire (RBQ; Turner, 1995) and the Repetitive Behavior Interview (RBI; Turner, 1997). Among the parent-report instruments for the evaluation of RRB, the Repetitive Behavior Scale-Revised (RBS-R) proposed by Bodfish, Symons, Parker, and Lewis (2000) is a questionnaire that provides a detailed assessment of RRB. To our knowledge, to date the RBS-R scale has not yet been translated into Italian language.

Previous studies that seek to substantiate the relationship between RRB and other clinical features in children with ASD through the RBS-R found contrasting results. In a large sample of children and adolescents aged 4–18 years with ASD, Bishop et al. (2013) reported an inverse correlation between ‘‘lower level’’ RRB and both NVIQ and chronological age, whereas ‘‘higher level’’ behaviors showed no relationship with IQ. Mirenda et al. (2010) did

not observe any significant relationship between RRB and both non-verbal IQ and chronological age in a sample of 287 preschool-aged children with ASD. Joseph et al. (2013) failed to find significant relationship between RRB and non-verbal Developmental Quotient, chronological age, social communication and sex in an autistic sample of preschoolers. In a sample of ASD toddlers, Wolff et al. (2014) observed that ‘‘higher level’’ RRB increased with chronological age and restricted behaviors were modestly negatively correlated with non-verbal Developmental Quotient at 12 months of age, suggesting that the relationship between RRB and cognitive measures develops over time.

It is worth noting that the different composition of ASD samples across studies in terms of age, sex, cognitive function and ASD symptom severity may contribute to obtain different and sometimes contradictory results, and thus may have significantly interfered with a clear understanding of the RRB profile in ASD patients.

1.1. Study purposes

The first purpose of this study was to apply, for the first time, the Italian version of the Repetitive Behavior Scale-Revised (RBS-R: Bodfish et al., 2000) in a sample of preschool-aged children with ASD and to investigate the relationship between RRB and demographic/clinical variables (i.e. chronological age, gender, autism severity and non-verbal cognitive abilities).

The second aim of this study was to explore the diagnostic accuracy of the RBS-R – Italian version – in a sample of preschool-aged children with ASD compared to peers with typical development.

2. Methods

2.1. Participants

2.1.1. Study 1: Application of the RBS-R – Italian version – to preschoolers with ASD. A total of 79 preschoolers including 64 males and 15 females affected by ASD were

included in the study. Participants were between 23 and 71 months (mean [*SD*] age = 51.8 [10.8] months) and were recruited from the ASD Unit at the IRCCS Stella Maris Foundation (Pisa, Italy), a tertiary care university hospital, between September 2009 and June 2013. These children were selected among individuals who performed the first diagnostic evaluation for a suspicion of ASD and received a final diagnosis of ASD based on DSM-IV-TR criteria (American Psychiatric Association, 2000). ASD diagnosis was performed by a multidisciplinary team (a senior child psychiatrist, an experienced clinically trained research child psychologist and a speech-language pathologist) during 5–7 days of extensive evaluation and confirmed by the administration of Autism Diagnostic Observation Schedule-Generic (ADOS-G; Lord et al., 2000). Exclusion criteria were: (a) neurological syndromes or focal neurological signs; (b) significant sensory impairment (e.g., blindness, deafness); (c) anamnesis of severe birth asphyxia, head injury or epilepsy; (d) potential secondary causes of ASD revealed by high-resolution karyotyping, DNA analysis of Fragile-X, or screening tests for inborn errors of metabolism and (e) use of any psychotropic medication.

2.1.2. Study 2: Diagnostic Accuracy of the RBS-R-Italian version. Scores on RBS-R of the ASD sample were compared with those of 79 typically developing (TD) controls. The TD sample, consisting of 64 males and 15 females (mean [*SD*] age = 51.6 [10.6] months), was recruited from two Pisa area kindergartens. Children with TD were not developmentally assessed via standardized test. The inclusion criteria for this group were: (a) frequency of regular kindergarten without support teacher (the Italian law provides teacher support for children with developmental/clinical problems); (b) no parent or teacher concern about child development, as indicated in both of the two following descriptive questions of the Child Behaviour Checklist (CBCL; Achenbach & Rescorla, 2000): ‘Does the child have any illness or disability (either physical or mental)?’ and ‘What concerns you most about the child?’; (c) a CBCL Total Problems T score lower than 60 (considered indicative of not clinical case); (d)

absence of a CBCL profile suggesting the presence of an ASD (Muratori et al., 2011). Good psychometric properties of the CBCL (validity and reliability) have been described (Frigerio et al., 2009).

Demographic and clinical characteristics of ASD and TD samples are reported in Table 1.

<Insert Table 1 here>

2.2. Instruments

2.2.1. Restrictive and repetitive behaviors. RRB were assessed through the RBS-R, an empirically derived clinical rating scale fulfilled by parents which provide a quantitative, continuous measure of the full spectrum of RRB. Items are rated on a four-point Likert scale (ranging from 0 = behavior does not occur to 3 = behavior occurs often and is a severe problem). Items on the RBS-R are grouped into six subscales: (1) Stereotyped Behavior; (2) Self-Injurious Behavior (SIB); (3) Compulsive Behavior; (4) Ritualistic Behavior; (5) Sameness Behavior; and (6) Restricted Interests Behaviors. Two raw scores can be calculated for each subscale: one based on the summed item scores within each subscale named ‘‘Score’’—and one based on the number of items endorsed (i.e. number of items with non-zero score) named ‘‘Endorsement’’. Total Score (the sum of all Subscales-Scores), Total Number-Endorsed Score (the sum of all Number-Endorsed–Subscale-Scores), Global Rating Score (the judgment of the parents of as the RRB affect the child’s life summarized in a score that ranges from 1 to 100) were also calculated.

A five-factor solution (Lam & Aman, 2007) and a two-factor solution (Georgiades, Papageorgiou, & Anagnostou, 2010; Mirenda et al., 2010) scoring of RBS-R were adopted for this study. Specifically, in the five-factor solution scoring, the original Ritualistic Behavior and Sameness Behavior subscales were combined in the Ritualistic/Sameness Behavior subscale, whereas in the two-factor solution scoring, a Low-Level Index (composed of items

pertaining to Stereotyped, and SelfInjurious subscales) and a High-Level Index (composed of items related to Compulsive, Ritualistic, Sameness and Restricted Interests Behaviors subscales) were evaluated.

Forward and backward translations were conducted according to the guidelines for cross-cultural adaptation of measures provided by Guillemin, Bombardier, and Beaton (1993). Two native Italian speakers who were also fluent in English and had extensive clinical experience in autism produced independent forward translations of the scale from English to Italian. The two forward translations were reviewed by a ‘‘consensus committee’’ of Italian speaking clinicians until agreement was reached on all items. The final version of the forward translation was backward translated to English by a native English speaker researcher who was also fluent in Italian. The backward translation was item-to-item compared with the original instrument by an English-speaking “consensus committee” of clinicians and no differences were discovered.

2.2.2. Autism diagnostic measures. The Autism Diagnostic Observation Schedule-Generic (ADOS-G; Lord et al., 2000) is a semi-structured instrument wherein the examiner attempts to engage the child with a series of social presses and rates the child’s social-communicative impairment and restricted and repetitive behaviors. ADOS-G comprises four modules tailored to an individual’s language ability. In the current study, participants completed Module 1 or Module 2 of the ADOS-G; the assessment was performed by two clinical psychologists (F.A. and A.N.) who met standard requirements for research reliability. A calibrated ASD severity score (CSS) was calculated based on ADOS raw scores (Gotham, Pickles, & Lord, 2009). The CSS range is 1–10; it provides a measure of autism symptoms that is independent of age and language ability and thus is better suited than the ADOS for assessing the severity of ASD (Shumway et al., 2012).

2.2.3. Non-verbal cognitive/developmental abilities. A number of standardized tests

were used to assess intellectual abilities due to differences in the age, verbal skills and functioning level of children. These included: the Leiter International Performance Scale-Revised (LIPS-R; Roid & Miller, 1997), the Griffiths Mental Developmental Scales-Extended-Revised (GMDS-ER; Luiz et al., 2006), and the Italian version of Wechsler Preschool and Primary Scale of Intelligence (WPPSI, Wechsler, 2008). When the tool provides a mental age (MA), IQ was estimated dividing MA by the child's chronological age (CA): ([MA/CA] 100). For this study we consider the non-verbal IQ scores.

2.3. Procedures

The study was carried out according to the standards for good ethical practice of the IRCCS Stella Maris Foundation and in accordance with the guidelines of the Declaration of Helsinki. Written informed consent from a parent or guardian of each participant was obtained. After a brief explanation of the RBS-R, parents of ASD children filled out the RBS-R at the beginning of the comprehensive clinical evaluation. Analogously, after a brief explanation of the questionnaires, parents in the TD group filled out the RBS-R and the CBCL in an anonymous way at kindergarten.

2.4. Data analyses

The Spearman correlation was used to explore the relationships between RBS-R scores and age, NVIQ, and CSS within the ASD sample.

The Mann–Whitney test was performed to evaluate differences on RBS-R scores between all females with ASD (15 subjects; there was only one eligible match per girl) and a subgroup of 15 males with ASD matched with females for NVIQ and chronological age.

The Receiver operating curve (ROC) analysis was performed to check the diagnostic accuracy of the RBS-R in ASD versus TD groups. The area under the curve (AUC) represents the accuracy of the instrument in predicting children who will or will not have ASD. The interpretation of the AUC values is traditionally as follows: an AUC < .7 suggests "low"

diagnostic accuracy; an AUC from .7 to .9 suggests "moderate" diagnostic accuracy, and an AUC .9 suggests "high" diagnostic accuracy. Analyses were carried out using SPSS version 15.0 for Windows (SPSS Inc., Chicago, IL, USA).

3. Results

3.1. Study 1

3.1.1. RRB profile in Italian preschoolers with ASD. Preschoolers with ASD showed a greater frequency of Stereotyped (SB) and Ritualistic (RSB) behaviors followed by Restricted Interest (RIB), Compulsive (CB) and Self-Injurius behaviors (SIB) (see Table 2). Moreover ASD showed a greater frequency of atypical behaviors than TD (see Table 3). RBS-R mean scores are summarized in Table 4.

<Insert Tables 2, 3, 4 here>

3.1.2. Correlation between RBS-R scores and age, non-verbal IQ and CSS scores. No significant correlations between RBS-R scores, chronological age and non-verbal IQ were found. The CSS was weakly and significantly correlated with Ritualistic/Sameness Behaviors endorsement scale. Correlation data are summarized in Tables 5.

<Insert Table 5 here>

3.1.3. Sex differences on RBS-R scores within ASD sample. A preliminary analysis between females with ASD and a subgroup of males with ASD matched on non-verbal IQ and chronological age did not reveal any significant differences on CCS ($p = .357$), chronological age ($p = .838$) and non-verbal IQ ($p = .775$).

The Mann–Whitney test did not detect significant differences on RBS-R scores between the two subgroups, with the exception of Stereotyped Endorsement subscale mean score that was higher in males than in females ($p = .033$).

3.2 Study 2: Diagnostic Accuracy of the RBS-R - Italian version

ROC analysis indicated that the diagnostic accuracy in discriminating ASD from TD

was high for the Global Rating Score (AUC = .903; 95% CI .850–.955), whereas it was moderate for the Stereotyped (AUC = .804; 95% CI .742–.866), Ritualistic/ Sameness (AUC = .755; 95% CI .676–.835), Restricted Interest (AUC = .736; 95% CI .653–.820), Total scales (AUC = .814; 95% CI .744–.885), Low (AUC = .806; 95% CI .732–.879), and High Index (AUC = .771; 95% CI .694–.848). Finally, the diagnostic accuracy was low for the Self-Injurious (AUC = .610; 95% CI .516–.703), and Compulsive scales (AUC = .677; 95% CI .589–.765) (see Table 6).

<Insert Table 6 here>

4. Discussion

In a group of carefully assessed preschoolers with ASD we investigated the presence and the severity of different patterns of restricted and repetitive behaviors (RRB), as well as their correlation with clinical and demographic features. Moreover, the diagnostic accuracy of the RBS-R in discriminating ASD from TD was examined.

Consistently with previous methodologically similar literature (Joseph et al., 2013; Mirenda et al., 2010) preschoolers with ASD showed various types of RRB, but Stereotyped and Ritualistic/Sameness behaviors were the most frequent symptoms, followed by Restricted Interest and Compulsive behaviors. By contrast, the lowest frequency on RBS-R subscales was found for Self-Injurious behaviors; therefore they constituted the less frequent RRB in pre-schoolers with ASD. This result is not surprising as self-injurious behaviors are not ASD-specific compared to stereotyped behaviors, ritualistic/ sameness behaviors and restricted interests that are expected in participants with ASD. The low frequency of self-injurious RRB in our sample (41.7%) appears in line with previous literature. For example, a 50% occurrence rate of self-injurious behaviors was found in a French population of ASD patients, and these invalidating symptoms were particularly common in children under five years of age, with a greater severity of autism and delayed adaptive skills (Baghdadli, Pascal, Grisi, & Aussiloux,

2003).

The current investigation did not provide support for the existence of a relationship between RRB and chronological age. Differently, several previous studies detected the effect of age on the expressiveness of the RRB (Bishop, Richler, & Lord, 2006; Lam & Aman, 2007; Militerni, Bravaccio, Falco, Fico, & Palermo, 2002) and specifically, motor behaviors have been found more frequently in younger children, whereas complex behaviors in older participants (Bishop et al., 2013; Militerni et al., 2002; Richler, Huerta, Bishop, & Lord, 2010). However, it is possible that the narrow age range investigated in this study prevented highlighting changes in expression of RRB over time. However, albeit the pre-school years cover only a narrow age range, this period is a time of rapid and dramatic brain development associated with enormous maturation processes in social, cognitive, linguistic and adaptive functioning. So, even if our results did not detect differences in the RRB profile in younger versus older preschoolers with ASD, longitudinal studies are needed in order to clarify the issue of quantitatively and qualitatively changes of RRB over time in pre-school age.

In line with some investigations (Joseph et al., 2013; Mirenda et al., 2010), we did not observe that the cognitive development may affect the pattern of RRB in preschoolers with ASD.

Specifically, high-level RRB are thought to be more ASD-specific than low-level RRB, since their expressiveness can frequently be independent of NVIQ (Bishop et al., 2013; Richler et al., 2010; Szatmari et al., 2006). Failing to identify any significant relationship between high-level RRB and non-verbal IQ, our study supported that high-level RRB may be a possible distinctive feature of the autistic disorder rather than being related to the cognitive functioning per se. However, contrary to replicated results of literature (Bishop et al., 2013; Cuccaro et al., 2003; Lam, Bodfish, & Piven, 2008) we did not report a negative correlation between lower-order RRB and non-verbal IQ, but it is possible that the early age of our

participants contributed to the failure to find such a relationship. According to this view, a previous work on 830 ASD children (age range: 15 months to 11 years) reported that stereotypic behaviors and repetitive use of objects are common in participants with ASD below five years of age independently of the IQ level, whereas in children over five years of age, the frequency of these low-order RRB persisted in participants with intellectual disabilities, but declined in those with normal IQ range (Szatmari et al., 2006). Other reports confirmed these data (Esbensen, Seltzer, Lam, & Bodfish, 2009; Rojahn, Matlock, & Tasse, 2000), suggesting that a high prevalence of low-level RRB in ASD participants is strongly associated with intellectual disabilities only at an older age.

Our data did not reveal a strong relationship between ASD severity and RRB since the Calibrated Severity Score is observed to only be correlated with Ritualistic/Sameness Behaviors endorsement subscale. Previous reports investigating the association between core symptoms severity and RRB in preschoolers with ASD have shown inconsistent results (Joseph et al., 2013; Bodfish et al., 2000; Szatmari et al., 2006; Watt et al., 2008), possibly due to different measures used to define the social and communicative impairment, as well as to the different phenotypic features of the samples.

With respect to sex differences on RBS-R scores, our study did not detect statistically significant results, with the exception of values in Stereotypic endorsement subscale. In fact, males with ASD exhibit a greater variability of Stereotypic symptoms than females, suggesting that a more homogeneous phenotype in this domain may characterize pre-school girls with ASD. Several reports observed milder RRB amongst females with ASD compared to ASD males (Hartley and Sikora, 2009; Mandy et al., 2012; Sipes et al., 2011), even if others failed to find any sex differences in RRB (Andersson et al., 2013; Joseph et al., 2013). A possible explanation that concurs to the higher prevalence of RRB in ASD males than ASD females might be that the items of RBS-R are, almost in part, male-oriented (for example, the

restricted interests behaviors subscale frequently refer to cars, trains, dinosaurs), and thus they may under-detect RRB in females with ASD. This possible gender bias should be adequately considered in the interpretation of results and would benefit from including female-specific items in the questionnaires addressed for ASD participants.

Finally, in order to shed light on the diagnostic accuracy of the RBS-R in discriminating ASD from TD, ROC analysis was performed. A high diagnostic accuracy was detected only when the Global Rating Score of the RBS-R was used, a score that reflects the judgment of the parents of as the RRB affect the child's life. Therefore, the impact of RRB on child's daily functioning – neither the type nor the frequency of the RRB – seems to be the ''true'' factor able to discriminate between ASD and TD children. This result is consistent with research indicating the interference of RRB with level of functioning (Cuccaro et al., 2003) or adaptive behavior (Gabriels, Cuccaro, Hill, Ivers, & Goldson, 2005), and suggests that the degree of interference of RRB should be carefully assessed in clinical practice as an ASD typical sign.

5. Conclusions and future directions

This study takes advantage of a relatively large sample of preschoolers with ASD who underwent a thorough clinical assessment. These accurate phenotypic data allowed us to investigate the relationships between subscale scores of RBS-R and demographic/clinical measures. However, four limitations pertain to the current research. First, the lack of a broad representation of females with ASD in our sample, although in line with the epidemiology of the ASD, reduces the overall impact of our results. Since sex differences in ASD have important clinical implications, additional research using a large sample of females with ASD is needed (Mandic-Maravic et al., 2015).

Second, since the cognitive assessment tools of ASD patients vary according to child's level of ability (see Section 3), an unavoidable limitation in this study was the combined use

of NVIQ and ratio IQ, across several types of tests. In order to mitigate this bias, future investigations may opt to calculate the ratio IQ for every subject.

Third, it is important to note that the RBS-R scale is a parent report measure. Thus, the possibility of parental bias in interpreting the questions and quantifying the behaviors cannot be ruled out. In particular, both an overestimation that a low insight of maladaptive behaviors of their child should be considered, depending on mood/anxiety and core personality traits of primary caregivers. However, parents might have more opportunities to observe RRB and generally provide reliable information on symptoms of their own child (Glascoe, 1999).

Fourth, despite the new diagnostic classification (DSM-5, 2013) highlights the importance of RRB as a group of symptoms necessary for an ASD diagnosis, an accurate instrument that assesses all range of behaviors included under the broad category of RRB has not been developed yet. Future validated parent report interviews designed to comprise the evaluation of aberrant sensory features and repetitive speech, two RRB categories included in the current version of DSM are therefore warranted.

Fifth, the lack of a sample with other developmental concerns in order to have comparison data to match with ASD sample. Notwithstanding these limitations, our research confirms the utility of using the RBS-R to define the clinical aspects and the severity of RRB, a core impairment that could strongly interfere with day to day functioning of ASD patients. Moreover, research focused on the effectiveness of ASD treatments could benefit from periodic RRB assessment through the RBS-R, in order to obtain useful information about the presence, the severity and the interference of RRB with daily life. Finally, since the RBS-R could provide a stratification of ASD individuals according to the types of RRB, it may be used to examine the brain-behavior correlation in this domain. This issue is not yet univocal, since neuroimaging studies have consistently reported the involvement of an altered cortico-striato-thalamo-cortical circuitry in the emergence of RRB (Calderoni, Bellani, Hardan,

Muratori, & Brambilla, 2014; Langen et al., 2011), but the corpus callosum and the cerebellum have also been recently implicated (D'Mello, Crocetti, Mostofsky, & Stoodley, 2015; Wolff et al., 2015).

Declaration of interest

The authors report no declarations of interest. The authors alone are responsible for the content and writing of this article.

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Table 1

Demographic and clinical characteristics of ASD and TD groups

	ASD			TD		
	Mean (*SD*)	Median	Range	Mean (*SD*)	Median	Range
Males	64	–	–	64	–	–
Females	15	–	–	15	–	–
Mean age in months	51.8 (10.8)	51.7	23–71	51.6 (10.6)	51.6	25–69
Mean non-verbal IQ	90.9 (20.6)	90	41–129	–	–	–
Mean ADOS CSS	6.13 (1.79)	6	2–10	–	–	–

ASD: subjects with autism spectrum disorders; TD: subjects with typical development; SD: standard deviation; ADOS: Autism Diagnostic Observation Schedule; CSS: Calibrated Severity Score.

Table 2

Frequency of endorsement in a given subscale in ASD and TD sample

Subscales	ASD	TD
Stereotyped behaviors	88.6	49.3
Self injurious behaviors	41.7	21.5
Compulsive behaviors	75.9	36.7
Ritualistic/sameness behaviors	89.8	54.4
Restricted interest behaviors	79.7	43.1

Table 3

Frequency of endorsement in a given behavior in ASD and TD sample

Item no.	ASD	TD
1. Body movements	20.3	7.6
2. Head movements	14.9	8.9
3. Finger movements	51.4	8.9
4. Locomotion	56.8	20.3
5. Object usage	48.6	24.1
6. Sensory	50	10.1
7. Hits w/body	17.6	8.9
8. Hits against surface	10.8	7.6
9. Hits w/object	5.4	1.3
10. Bites self	10.8	10.1
11. Pulls hair/skin	13.5	5.1
12. Rubs/scratches	2.7	2.5
13. Inserts finger/object	12.2	7.6
14. Picks skin	6.8	5.1
15. Ordering	45.9	15.2
16. Completeness	33.8	10.1
17. Washing	13.5	5.1
18. Checking	13.5	6.3
19. Counting	27	21.5
20. Hoarding	12.2	16.5
21. Repeating	23	16.5

22. Needs to touch/tap	23	13.9
23. Eating/mealtime	35.1	25.3
24. Sleeping/bedtime	50	40.5
25. Self care routine	14.9	19
26. Transportation routine	28.4	20.3
27. Play/leisure routine	41.9	21.5
28. Communication	45.9	15.2
29. Placement of objects	21.6	16.5
30. No new places	18.9	10.1
31. No interruption	77	41.8
32. Walks certain way	14.9	6.3
33. Sits certain place	13.5	16.5
34. Appearance/behavior of others	13.5	11.4
35. Usescertain door	10.8	3.8
36. Videotapes	59.5	34.2
37. Difficult transitions	1.4	13.9
38. Insists on routine	39.2	12.7
39. Insists on time	13.5	11.4
40. Preoccupation with subject	55.4	17.7
41. Attached to object	21.6	10.1
42. Preoccupied with part of object	39.2	11.4
43. Preoccupation with movement	54.1	20.3

Table 4

RBS-R scores in the ASD sample

		ASD			
		Total (*n* = 79)	Males (*n* = 64)	Females (*n* = 15)	
RBS-R subscale		Mean (*SD*)	Mean (*SD*)	Mean (*SD*)	Pairwise effect Size (*d*)[a]
Stereotyped behaviors	Score	5.43 (4.19)	5.77 (4.04)	4.00 (4.65)	.40
	Endorsement	3.66 (2.40)	3.97 (2.29)	3.66 (2.40)	.13
Self Injurious behaviors	Score	1.06 (1.98)	1.02 (1.98)	1.27 (2.05)	- .12
	Endorsement	.80 (1.29)	.73 (1.18)	1.07 (1.71)	- .23
Compulsive behaviors	Score	2.13 (2.29)	2.28 (2.42)	1.47 (1.55)	.39
	Endorsement	1.52 (1.34)	1.61 (1.39)	1.13 (1.06)	.38
Ritualistic/sameness behaviors	Score	4.99 (4.19)	5.20 (4.34)	4.07 (3.45)	.28
	Endorsement	3.67 (2.69)	3.80 (2.79)	3.13 (2.20)	.26
Restricted interest behaviors	Score	2.41 (2.06)	2.61(2.13)	1.53 (1.50)	.58
	Endorsement	1.42 (.98)	1.50 (.98)	1.07 (.96)	.44

Total behaviors	Score	16.01 (11.05)	16.88 (11.38)	12.33 (9.59)	.43
	Endorsement	11.06 (6.17)	11.61 (6.03)	8.73 (6.43)	.46
Low index[b]		4.38 (9.88)	4.39 (4.49)	3.8 (5.44)	.11
High index[b]		12.62 (4.55)	12.50 (9.78)	8 (7.6)	.51
Global score		37.83 (23.66)	38.38 (22.79)	35.62 (27.86)	.10

ASD: Autism Spectrum Disorders; SD: Standard Deviation; RBS-R: Repetitive Behavior Scale-Revised.

[a]Effect size based on Cohen's d .20 is small, .50 is medium, and .80 is large.

[b]Low Index and High Index were calculated using Mirenda et al. (2010) procedures.

Table 5

Correlations between RBS-R subscales and chronological age, non-verbal IQ, CSS in ASD participants

		ASD		
RBS-R subscale		Chronological age	Non-verbal IQ	CSS
Stereotyped behaviors	Score	r = .003	r = - .059	r = .087
	Endorsement	r = - .057	r = - .111	r = .163
Self injurious behaviors	Score	r = .029	r = - .117	r = .033
	Endorsement	r = - .002	r = - .188	r = - 023
Compulsive behaviors	Score	r = - 074	r = - .058	r = .160
	Endorsement	r = - .067	r = - .134	r = .208
Ritualistic/sameness behaviors	Score	r = .093	r = .080	r = - .228
	Endorsement	r = .088	r = .063	r = - .287*
Restricted interest behaviors	Score	r = .139	r = .112	r = - .128
	Endorsement	r = .089	r = .185	r = - .155
Total behaviors	Score	r = .052	r = - .005	r = - .051
	Endorsement	r = .015	r = - .054	r = - .043

Low index	$r = .009$	$r = -.083$	$r = -.057$
High index	$r = .068$	$r = .071$	$r = -.115$
Global score	$r = -.055$	$r = -.120$	$r = -.036$

ASD: Autism Spectrum Disorders; SD: Standard Deviation; RBS-R: Repetitive Behavior Scale-Revised.

*$p = .019$.

Table 6

AUC (area under the curve) on RBS-R when comparing ASD group versus TD group

	RBS-R subscales ASD versus TD								
	Stereotyped Score	Self Injurious Score	Compulsive Score	Ritualistic/ Sameness Score	Restricted Int. Score	Total Score	Low	High	Global
AUC	.814	.610	.677	.755	.736	.814	.804	.775	.903
S. E.	.37.	.47	.45	.40	.43	.36	.34	.41	27
95% CI	.742 – .866	.516 –.803	.589 – .765	.676 – .835	.653 –.820	.744 –.885	.729 –.872	.687 –.862	.850 – .955
AUC accuracy	Moderate	Low	Low	Moderate	Moderate	Moderate	Moderate	Moderate	High

S.E. = Standard Error.

*Sweet and Picket criteria for AUC accuracy; ASD: subjects with autism spectrum disorders; TD: subjects with typical development; RBS-R: Repetitive Behavior Scale-Revised.