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ORIGINAL CONTRIBUTION

Validation of the repetitive and restricted behaviour scale in autism spectrum disorders

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Abstract Repetitive and restricted behaviours represent a common problem for various psychiatric syndromes, especially in autistic spectrum disorders, and they include a wide range of heterogeneous behavioural manifestations. An accurate and standardized description of these behaviours is needed to advance the understanding of this complex and heterogeneous clinical dimension of autism. The present article reports the reliability and validity studies of a new assessment scale: the repetitive and restricted behaviour scale. 145 subjects with autism spectrum disorders were assessed using the RRB scale. The RRB scale has good interrater reliability, internal consistency and content validity. Factorial analysis produced four clinically meaningful factors, i.e. "sensorimotor stereotypies", "reaction to change", "restricted behaviours" and "modulation insufficiency". The RRB scale has good psychometric qualities and constitutes a real breakthrough towards a neurofunctional approach to autistic disorders. It should be valuable for research and treatment, and in clinical practice.

Keywords Autism spectrum disorder · Stereotyped behaviour · Assessment scale · Validation

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Introduction

Repetitive behaviours, stereotyped activities and restricted interests, summarized as repetitive and restricted behaviours (RRB), can occur in a number of psychiatric disorders, particularly in autism spectrum disorders (ASD), obsessive compulsive disorders (OCD), mental retardation, and Tics and Tourette syndrome [6, 13, 23]. In ASD, RRB constitute the third dimension of diagnostic criteria [1, 45] which covers a wide range of heterogeneous behavioural manifestations such as motor stereotypies, sensory-related behaviours, circumscribed interests, rituals, excessive sensitivity to change, echolalia, and selfinjurious behaviours [10, 13, 23, 25, 39, 40]. Their physiopathological mechanisms still remain unsolved [10, 13, 23, 25, 40]. All these behaviours point to a lack of flexibility that results in major difficulties in daily life, both for the individual and his or her relatives [16, 25, 26]. Most of the previous research has divided RRB into low-level and high-level behaviours [4, 7, 10, 13, 17, 27, 30, 39, 40]. Low-level RRB correspond to repetitive sensorimotor behaviours and are said to be linked to younger age and associated with mental retardation [4, 10, 25]. Conversely, high-level RRB refer to more complex repetitive activities (rituals) and insistence on sameness. They seem to develop with age and to be more strongly expressed in high-functioning individuals [4, 17, 39]. High-level RRB might also be more specific of autism than sensorimotor RRB [10, 40].

However, studies dealing with RRB in ASD to date have not taken into account all the forms of RRB and have rarely used specific tools.

The repetitive behaviour scale (RBS and its revised version the RBS-R) has been the most complete and the only specific tool for the assessment of RRB in autism



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[5, 6]. The RBS-R is based on a descriptive approach to the behaviours observed and was recently re-validated [21]. It contains 43 items but lacks some aspects of RRB, such as echolalia, mannerisms and stereotyped emotional manifestations.

We have developed a new assessment scale based not only on a purely descriptive evaluation, but also on a neurofunctional approach to behaviours, i.e. the RRB scale. It covers the whole range of repetitive and restricted behaviours observed in ASD. This new scale should allow first the definition of more objective and homogeneous subsets of RRB that might be related to different psychophysiological mechanisms, and then the improvement of therapeutic strategies and evaluation of their efficacy. The present article reports the reliability and validity studies of the RRB scale.

Methods

The RRB scale

The construction of the RRB scale was based on clinical observations and on an extensive review of the literature on RRB in both autism and mental retardation [2, 5, 9, 20, 21, 23–25, 33, 34, 36, 40]. An initial list of 43 items, corresponding to the most frequent or characteristic RRB was tested on a small sample of children with ASD. This list gave a satisfactory outline of RRB and confirmed their heterogeneity [7]. This list was then submitted to clinical experts (child psychiatrists, psychologists, speech therapists, nurses) in order to make it more accurate and comprehensible, and thus to confirm its face validity.

The present version of the RRB scale comprises 35 items (items detailed in Table 2) explained in a glossary (available on request) and evaluated according to a fivelevel Likert scale (0 = "the behaviour is never expressed")by the person", 1 = "weakly expressed", 2 = "moderately expressed", 3 = "severely expressed" and 4 = "the behaviour is very characteristic of the person and very severely expressed"). Since RRB constitute a complex and heterogeneous dimension, a sound understanding of the person is necessary to complete the scale with accuracy. The RRB scale is therefore filled out by professional caregivers after they have observed the person in various situations of daily life (during free and structured activities, alone and in a group, during mealtimes...). Since some behaviours, such as restricted interests and resistance to change, can be difficult to rate, direct observation can be complemented by information collected from a relative of the subject. Moreover, the rater does not have to take into account RRB that were typical of the person but are no longer observed.



Validation of the RRB scale required a large and varied population in terms of age, intellectual capacities, symptom severity, and residential settings. Thirteen centres took part in the validation study (for details see "Acknowledgments"). Informed consent was obtained from the parents, and anonymous data were collected and analysed in an INSERM research centre authorized by the Direction Générale de la Santé (No 06032).

The participants were 145 children, adolescents and adults (38 female, 107 male) aged from 3 to 33 years: there were 49 young children (aged from 3 to 7 years), 40 children (8–12 years), 32 teenagers (13–18 years), and 24 adults (19–33 years).

Disorders were diagnosed by expert clinicians according to DSM-IV-R criteria [1] as follows: autistic disorder (AD, n=99), pervasive developmental disorder—not otherwise specified (PDD-NOS, n=41) and Asperger Syndrome (AS, n=5). The ADI-R [24] was collected for 32% of the sample to confirm the diagnoses. No major neurological syndromes were diagnosed. Intellectual abilities [development quotient (DQ) or intellectual quotient (IQ)] was evaluated with different tests depending on age and ability and on centre (BL-R, EDEI-R, WISC-III, WISC-IV, PEP-R, WPPSI-III, Nemi, and Leiter-R [8, 28, 32, 35, 41–43, 47]). According to DSM-IV-R criteria, 24 subjects had profound mental retardation, 33 had severe mental retardation, 37 had moderate mental retardation, 25 had slight mental retardation, and 26 had no mental retardation.

The severity of autistic symptoms was assessed with the CARS [29, 36]; data were available for 121 subjects.

Participants' characteristics are presented in Table 1.

The ages of males and females were similar. Males had higher DQ-IQ than females (t = 2.53, df = 143, p = 0.012); however, the difference was no longer significant when considering only participants with DQ-IQ below 80. Individuals with AD and PDD-NOS did not differ in age or DQ-IQ.

Subpopulation to assess interrater reliability

A subgroup of 21 children and 8 adults (7 female, 22 male), aged 3–33 years (M = 12, SD = 9) was assessed for

Table 1 Participants' characteristics

	Number of subjects	Mean	Standard deviation	Minimum	Maximum
Age (years)	145	12.2	7.3	3	33
DQ-IQ	145	45.0	25.4	6	126
CARS	121	34.9	7.0	20	55.5
Covi	88	7.1	2.7	3	14



interrater reliability. The children were individuals from the Child Psychiatry Day Unit of the "Centre Hospitalier Universitaire" in Tours and the adults were residents of "les Maisonnées", a centre at Azay le Rideau. Diagnoses in this subpopulation were AD (n=16) and PDD-NOS (n=13). Overall DQ-IQ ranged from 10 to 114 (M=55.0; SD=23.9).

Procedures

Validity study

The factor structure of the RRB scale was explored using principal component analysis (PCA) with varimax rotation. The number of factors to be retained was defined by the scree-test criterion and complemented by screeplots of simulated random data. Items were included in the factor structure if they loaded |0.35| or higher on a factor, and if the loading was at least |0.10| higher than the loading on any other factor. All the items loading highly on a factor were referred to as a subscale. They were combined to produce an average score for each subscale. The correlations between each item and the overall score of each subscale were computed to confirm the factor structure. Finally, the Cronbach α coefficient was used to assess internal consistency of each subscale.

Relationships between subscale scores and participant characteristics (i.e. age, DQ-IQ, severity of autistic symptoms (measured with the CARS) and anxiety) were assessed using Pearson's product moment correlation. Anxiety is not a core feature of ASD, but it can explain the expression of RRB [38, 40]. We, therefore, used the Covi anxiety scale which contains three items rated on a five-point scale and provides easy and rapid assessment of anxiety based on observation of the individual [12, 22, 31] (see Table 1). Standard multiple regression analyses were then performed to complement the interpretation of the relationships between the variables (subscale score as dependent variable).

Additional statistical analyses were also carried out to complement the main results (t tests, χ^2 tests, and ANOVA with Newman–Keuls post hoc tests). STATISTICA v8 (StatSoft, Inc.) was used.

Interrater reliability study

To check the reliability of the RRB scale, two raters independently filled out the RRB scale for each subject, according to the above-mentioned principles. Reliability was then ascertained by calculating the weighted kappa statistic (Kw) for each of the 35 items [11, 14, 15] and the intraclass correlation coefficient (ICC) on the score of each subscale.

Results

Validity study

Factor structure

Frequency of occurrence of each behaviour was calculated on the basis of present (ratings 1, 2, 3, and 4) versus absent (rating 0) scores. All items occurred in more than 10% and less than 90% of the sample. The 35 items were thus retained for subsequent analysis.

PCA was performed on the 145 subjects and produced four factors that cumulatively accounted for 43% of total variance (screeplots are presented on Fig. 1 and factor loadings are reported in Table 2). These four factors were fairly similar in terms of number of items (11, 7, 8, and 7, respectively) and percentage of explained variance (12, 11, 11, and 9, respectively).

Factor 1 (F1) comprised 11 items corresponding to repetitive motor behaviours, motor mannerisms, repetitive cries, body-focused behaviours and atypical sensory behaviours. This factor was labelled "sensorimotor stereotypies". Factor 2 (F2) comprised seven items that reflected adoption of rituals for various activities, reaction to small changes in the environment and echolalia. It was labelled "reaction to change". Factor 3 (F3) comprised eight items dealing with repetitive or ritualized use of objects, interest in or attachment to objects or details of objects, circumscribed subjects of interest and lack of interest in novelty. It was labelled "restricted behaviours". Factor 4 (F4) comprised seven items that included aggressiveness towards self and others, need to control the progress of activities, stereotyped emotional manifestations and agitation. This factor was labelled "modulation insufficiency".

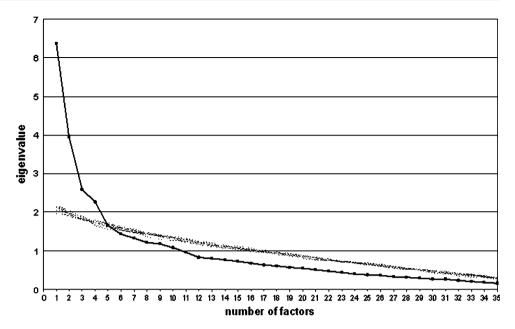
All items of the RRB scale correlated more highly with their subscale (from 0.59 to 0.66) than with the other subscales (less than 0.21). The correlations between the four factors ranged from 0.05 to 0.32. The Cronbach α coefficient showed good internal consistency for each subscale (0.81, 0.79, 0.75, and 0.72, respectively).

No difference was found between males and females for the four subscale scores. In terms of diagnosis, subjects with AD had higher scores than those with PDD-NOS on three subscale scores: F1 (t = 2.79, df = 138, p = 0.006), F2 (t = 3.36, df = 138, p < 0.001), and F3 (t = 4.86, df = 138, p < 0.001).

Significant negative correlations were found between F1 and F3 scores and DQ-IQ. Since CARS score has been known to be linked to level of mental retardation [25] (in this study, r = 0.51, p < 0.001), partial correlations were computed between subscale scores and CARS score: F1, F3, and F4 scores showed significant correlations. For the



Fig. 1 Screeplots produced by PCA of the RRB scale and of ten simulated random datasets



Dotted lines: simulated random data

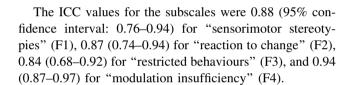
Covi score, a slight negative correlation was found with the F3 score. Conversely, F4 was positively correlated with anxiety (see Table 3).

Standard multiple regression analyses were then performed to clarify the relationships previously shown between F1, F3 and F4 subscale scores on the one hand and CARS score, DQ-IQ, and Covi score on the other. These analyses made it possible to examine the relative importance of these three independent variables in the explanation of F1, F3, and F4 subscale scores. Only the CARS score made a statistically significant contribution to the explanation of the F1 score $[R^2 = 0.44, adjusted R^2 =$ 0.42, F(3.76) = 20.27, p < 0.001]. All three independent variables contributed statistically to the explanation of the F3 score $[R^2 = 0.29, adjusted R^2 = 0.26, F(3,76) = 10.37,$ p < 0.001]. Finally, CARS and Covi scores made a statistically significant contribution to the explanation of the F4 score $[R^2 = 0.26, adjusted R^2 = 0.23, F(3,76) = 8.87,$ p < 0.001], and the Covi score made the greatest contribution (see Table 4).

Interrater reliability study

The RRB scale showed good reliability

Using the criteria defined by Cicchetti [11], 9 items (items 1, 6, 16, 18, 22, 29, 30, 32, 34) had excellent (Kw = 0.75–1) reliability, 17 items (items 2, 3, 5, 7, 8, 9, 11, 13, 17, 19, 21, 23, 24, 25, 26, 31, 33) had good (0.60–0.74) reliability and 9 items (items 4, 10, 12, 14, 15, 20, 27, 28, 35) had moderate (0.40-0.59) reliability (see Table 2).



Discussion

The validation study showed the sound psychometric qualities of the RRB scale and its ability to describe four clinically meaningful components within the repetitive and restricted behaviours of ASD, i.e. sensorimotor stereotypies, reaction to change, restricted behaviours, and modulation insufficiency. Of these four factors, three were more severely expressed in AD compared to PDD-NOS. Moreover, sensorimotor stereotypies and reaction to change were not influenced by level of mental retardation or by anxiety, and they remained stable over time. These two dimensions, therefore, appear to be integral parts of autistic core symptoms. On the other hand, restricted behaviours, which were particularly related to level of mental retardation, and modulation insufficiency, mainly linked to the expression of anxiety, are probably less specific of ASD.

Features and content of the RRB scale

The four dimensions of the RRB scale did not appear to be influenced by age. This is consistent with studies emphasising the persistence with age of this dimension of autistic disorder [18, 37].



Table 2 Factor loadings and weighted kappa statistic Kw of the 35 items of the RRB scale

	Item		Factor loadings (% of variance)			
			F1 (12%)	F2 (11%)	F3 (11%)	F4 (9%)
1	Repetitive body rocking	0.76	0.65	0.09	0.06	-0.01
2	Repetitive and atypical movements of mouth	0.64	0.61	0.02	-0.09	0.05
3	Repetitive movements of head	0.70	0.67	0.08	0.14	-0.12
4	Repetitive movements of arms and hands	0.56	0.56	-0.18	0.14	0.08
5	Bizarre gait	0.64	0.53	0.10	-0.11	0.09
6	Repetitive pacing	0.75	0.31	0.03	0.40	0.40
7	Mannerism or rigidity of posture	0.69	0.59	0.20	-0.17	0.01
8	Quick and complex movements of fingers in front of eyes, with or without objects	0.67	0.59	-0.20	0.37	0.12
9	Repetitive and non-functional use of objects	0.65	0.34	-0.05	0.66	-0.01
10	Interest in a detail of objects	0.46	0.36	0.21	0.60	0.06
11	Attachment to certain objects	0.64	-0.03	0.15	0.63	0.23
12	Circumscribed interests	0.57	-0.04	0.26	0.50	0.09
13	Exact repetition of words, sentences or tunes	0.72	-0.03	0.53	-0.24	-0.03
14	Repetitive uttering of cries or sounds	0.56	0.54	-0.05	0.27	0.26
15	Aggressive behaviours towards others or objects	0.51	-0.01	-0.06	0.07	0.71
16	Self-injurious behaviours	0.75	0.28	-0.08	0.21	0.48
17	Body-focused behaviours	0.73	0.51	-0.22	0.07	0.30
18	Rituals for daily living activities	0.76	0.03	0.69	0.17	0.13
19	Play and leisure rituals	0.73	0.02	0.68	0.41	-0.15
20	Communication rituals	0.55	0.05	0.67	-0.06	0.16
21	Route and exploration rituals	0.74	0.06	0.62	0.27	0.16
22	Alignment rituals	0.75	-0.14	0.24	0.58	0.17
23	Reaction when progress of a ritual or activity is disturbed	0.61	0.07	0.44	0.29	0.52
24	Repetitive tendency to hoard objects	0.63	-0.15	-0.11	0.58	0.29
25	Tendency to try to control activities or conversation	0.66	-0.20	0.38	-0.15	0.60
26	Atypical sensory behaviours	0.72	0.53	0.12	0.12	0.29
27	Attraction to things that move	0.56	0.15	-0.15	0.59	-0.04
28	Attraction to certain sounds or noises	0.55	0.42	-0.02	0.28	0.10
29	Interest in a part of the body of others	0.90	0.33	0.01	-0.14	0.47
30	Need that things be laid out in a specific pattern	0.79	0.03	0.45	0.48	0.00
31	Reaction to changes in material environment	0.65	-0.04	0.65	0.29	-0.16
32	Reaction to changes in appearance or behaviours of relatives	0.85	0.06	0.61	-0.05	0.09
33	Lack of interest in novelty	0.60	0.08	0.07	0.37	-0.11
34	Stereotyped emotional manifestations	0.77	0.17	0.20	0.00	0.59
35	Difficulty in remaining still and inactive	0.59	0.13	0.03	0.16	0.57

Factor loadings in bold indicate the factor on which item loaded

"Sensorimotor stereotypies" and "restricted behaviours" were significantly associated with severity of autistic symptoms assessed with the CARS and were more severely expressed in AD subjects compared to PDD-NOS subjects. However, while sensorimotor stereotypies appeared to be mainly associated with severe forms of AD, the restricted behaviours seemed to constitute a more complex dimension. In fact, "restricted behaviours" were

related to autism but were also influenced by mental retardation and anxiety.

On the other hand, "reaction to change" was linked neither to severity of autistic symptoms nor to intellectual abilities. However, reactivity to change was greater in AD subjects than in PDD-NOS subjects. We can thus hypothesize that "reaction to change" is a specific marker of AD, independent of the severity of symptoms or mental



Table 3 Correlations between subscale scores and participants' characteristics

Subscale	Age	DQ-IQ	CARS ^a	Covi	
F1—sensorimotor stereotypies	0.17*	-0.37***	0.47***	0.16	
F2—reaction to change	0.11	-0.05	-0.05	0.05	
F3—restricted behaviours	-0.05	-0.26**	0.31***	-0.25*	
F4—modulation insufficiency	0.14	-0.14	0.29**	0.40***	

^a Partial correlation controlled for DQ-IQ

Table 4 Standard multiple regression analyses of variables explaining the subscale scores (N=80)

Variable	β	SE β	t value
DQ-IQ	-0.05	0.10	-0.53
CARS	0.63	0.10	6.22***
Covi	0.06	0.08	0.73
DQ-IQ	-0.29	0.11	-2.57*
CARS	0.26	0.11	2.30*
Covi	-0.28	0.10	-2.87**
DQ-IQ	-0.04	0.12	-0.37
CARS	0.30	0.12	2.54*
Covi	0.37	0.10	3.69***
	DQ-IQ CARS Covi DQ-IQ CARS Covi DQ-IQ CARS	DQ-IQ -0.05 CARS 0.63 Covi 0.06 DQ-IQ -0.29 CARS 0.26 Covi -0.28 DQ-IQ -0.04 CARS 0.30	DQ-IQ -0.05 0.10 CARS 0.63 0.10 Covi 0.06 0.08 DQ-IQ -0.29 0.11 CARS 0.26 0.11 Covi -0.28 0.10 DQ-IQ -0.04 0.12 CARS 0.30 0.12

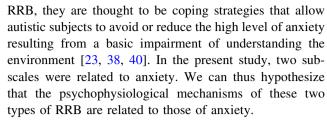
 β = standardized regression coefficients, SE β = standard error of β * P < 0.05; ** P < 0.01; *** P < 0.001

retardation [10, 40]. Previous studies have also proposed that these RRB are a feature of high-functioning autism and AS [17, 39]. The sample in the present study did not comprise enough participants with good intellectual abilities to confirm this. Finally, it could be hypothesised that this dimension of the RRB scale might reflect an obsessive-compulsive trait that can be observed in some autistic subjects [46].

The fourth factor, "modulation insufficiency", is the only factor which did not differentiate AD from PDD-NOS, despite being linked to severity of autistic symptoms. The strong relationship between modulation insufficiency and anxiety suggests that such RRB might be related to anxiety disorders, which are frequently observed in ASD, rather than to autistic core symptoms [38]. Impulsiveness, agitation and emotional instability could also be explained by associated disorders such as ADHD or oppositional disorder.

Anxiety and RRB

Subjects with autism are known to have difficulties in communicating their emotional states; however, several studies have shown a high prevalence of anxiety in autistic disorder [19, 44]. Among the functions hypothesized for



On the one hand, a high level of expression of "restricted behaviours" is associated with fewer manifestations of anxiety. Indeed, focusing on a restricted subject of interest or attraction to details of objects can correspond to avoiding strategies. By focusing on a restricted range of known and reassuring stimuli, the person avoids external stimuli that are difficult to integrate and that are stressful.

On the other hand, anxious participants had high levels of "modulation insufficiency". In fact, the different behavioural manifestations contained in this subscale seem to reflect difficulty with modulation of emotions. Therefore, subjects with a high score on this subscale may have major difficulties in adapting to a situation and controlling their feelings. The behaviours which make up the "modulation insufficiency" subscale can also be viewed as a means by which autistic subjects can exteriorize their internal states. Conversely, individuals with restricted behaviours, who tended to have severe autistic symptoms and associated mental retardation, probably had difficulties in communicating their anxiety.

Comparison with previous findings on RRB

"Sensorimotor stereotypies" (F1) and "reaction to change" (F2) are in accordance with the low/high-level model proposed by several authors [13, 39, 40], whereas "restricted behaviours" (F3) and "modulation insufficiency" (F4) constitute supplementary meaningful dimensions that could provide additional information for the understanding of RRB. "Restricted behaviours" include behaviours that were previously associated with different levels of ability; namely "restricted thoughts" considered as a high-level RRB and "interest in details of objects" placed in low-level RRB [4, 40]. Similarly, "modulation insufficiency" contains behaviours known to be associated with mental retardation (self-injury) [4, 23, 25] and less severe forms (stereotyped emotional manifestations). Except for self-injurious behaviours [23, 25], the behaviours included in the "modulation insufficiency" subscale have not been studied before as RRB. Interestingly, this dimension echoes the second factor of the BSE-R: a standardized scale which evaluates autistic behaviours [2].

Another scale assessing RRB has already been published: the RBS-R [6]. Both scales describe similar subtypes of RRB, i.e. reaction to change, sensorimotor activities and restricted behaviours [21], but there are



^{*} $P \le 0.05$; ** $P \le 0.01$; *** $P \le 0.001$

differences between the two scales in terms of content and approach to behaviours. Although the RRB scale assesses behaviours that are not present in the RBS-R, the main difference is probably in the construction of their items. For the RBS-R each item corresponds to one precise behavioural manifestation (e.g. "insists on sitting in the same place", "insists on using a particular door", "hits self with body part", "hits self with object") whereas in the RRB scale each item includes different behavioural manifestations of the same hypothesized psychophysiological mechanism (e.g. "self-injurious behaviours", "rituals for daily living activities"). We believe that this complementary approach to RRB should provide an additional insight into these behaviours and improve the understanding of the underlying mechanisms of RRB.

Strengths and limitations

First, content validity allowed identification of the hypothesized multidimensional structure of the RRB scale and extraction of four statistically sound factors. Only two of the 35 items of the RRB scale failed to load on a single factor, and they were not included in the four subscales derived by PCA. However, this four-factor solution does not explain all the heterogeneity and complexity of RRB in ASD (43% of variance). It is, nevertheless, in accordance with previous factor analyses of RRB [21].

The sample was not completely representative of the ASD population, since 82% of the participants had mental retardation and the sample comprised only five subjects with AS. The proposed estimates of associated cognitive impairments in ASD populations currently range from 40 to 70% [3]. However, the study reported here included individuals from various centres that reflect the different residential settings offered in France. Further studies should be performed including a larger group of high-level autistic and Asperger individuals.

Finally, this variety of centres and the direct observation method used may have produced variations in the ratings of the RRB scale. In fact, the detailed glossary attached to the scale and the possibility of complementing the observation by information collected from relatives probably contributed to the good reliability and improved the accuracy of the evaluation.

Conclusion

The RRB scale provides a precise and reliable functional description of RRB over the whole autistic spectrum. It supports the multidimensionality of this field of autistic symptomatology. The description of different behavioural profiles should thus be valuable in research and clinical

practice. It also supports the idea that dimensional analysis can be a more fruitful approach to the autistic spectrum than using diagnostic categories. Moreover, this new scale should help to characterize symptoms which are not specific to one psychiatric syndrome and which are an issue in differential diagnosis. Such an approach could also provide treatment indications and hypotheses on physiopathological mechanisms. For example, since RRB are observed in both ASD and in other psychiatric disorders, it would be interesting to evaluate RRB in subjects with mental retardation or OCD and then to compare their profiles to those of ASD individuals. Similarly, since RRB are known to be particularly persistent in autism, it would be interesting to study the potential differential evolution of the four types of RRB described by the scale and to assess their sensitivity to treatment.

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