Development of Two Dimensional Measures of Restricted and Repetitive Behavior in Parents and Children

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Objective: Restricted and repetitive behaviors (RRBs) are a heterogeneous set of behaviors common across a wide range of neurodevelopmental disorders (NDDs) and neuropsychiatric disorders (NPDs) that extend well into the general population. This study introduces 2 dimensional measurements of RRBs for use in typical and clinical populations from infancy to adulthood.

Method: The Childhood Routines Inventory–Revised (CRI-R) and the Adult Routines Inventory (ARI) were created and administered online to a nationally representative cohort of 3,108 parents with 3,032 children (range 12 months to 17 years 11 months). Twenty-six percent of children and 36% of adults had at least 1 NDD or NPD.

Results: Principal axis factoring exploratory analysis showed a 2-factor structure for the 2 instruments (motor behaviors/compulsions and rigidity/insistence on sameness). Analyses for convergent and discriminant validity, internal consistency (Cronbach $\alpha \geq 0.94$), and test-retest reliability ($r \geq 0.87$) indicated strong psychometric properties. Item response theory analyses indicated strong reliability across the score range for the 2 instruments. RRB rates varied across development, peaking between the preschool and school years. Children with NDDs or NPDs (particularly those with autism spectrum disorder, schizophrenia/bipolar disorder, or obsessive-compulsive disorder/tic disorders) had increased RRBs compared with those with no diagnosis. Parent–child (0.69–0.84) and sibling–sibling (0.76–0.87) intraclass correlations indicated high heritability. Children of parents with an NDD or an NPD exhibited more RRBs compared with children of parents without NDDs or NPDs.

Conclusion: The CRI-R and ARI are open-source instruments with excellent psychometric properties and will be useful for developmental, clinical, and family genetic studies and for the identification of prodromal conditions involving RRBs.

Key words: repetitive behavior, dimensional assessment, parents and children

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Restricted and repetitive behaviors (RRBs) constitute a broad range of behaviors, including simple motor stereotypies and tics and more complex ritualized and rigid behaviors, compulsions, and restricted interests that vary in frequency, intensity, and duration. RRBs are core diagnostic features of autism spectrum disorder (ASD; *DSM-5*)[1] but also appear in a range of other neurodevelopmental disorders (NDDs) and neuropsychiatric disorders (NPDs), such as intellectual and developmental disabilities, schizophrenia, obsessive-compulsive disorder (OCD), and tic disorders (e.g., Tourette's syndrome).[2,3] Contrary to traditional categorical nosologic boundaries, recent findings have indicated that most of these NDDs and NPDs are overlapping syndromes[4] that are best represented as a collection of dimensional traits that extend into the general population.[5,6]

RRBs also are common throughout typical development.[7,8] Strong preferences for sameness in the environment, lining objects in straight lines, rigid routines, and an acute perceptual awareness of minute details are frequently observed in typically developing children 2 to 7 years of age.[9,10] These typical RRBs can serve different adaptive roles, including motor[11,12] and nervous system[13] maturation and emotional and arousal regulation.[10,14] However, in the context of NDDs and NPDs, RRBs adversely affect multiple aspects of functioning[15] and therefore are important targets for clinical intervention.[16] Distinguishing clinically significant behavior from typical behavior requires a clearer understanding of the normal variability of a broader range of RRBs across the lifespan.

In addition to the high rates of comorbidity in individuals with NDDs and NPDs,[17,18] such disorders frequently co-occur within families.[19] In some instances, family members can share a common diagnosis, or different symptoms might manifest as alternative phenotypes of a common genotype.[20] Therefore, it is important to understand the familial context of RRBs, to determine whether children of parents with at least 1 NDD or NPD show increased levels of RRBs that might signify prodromal states, and to enable early identification and intervention of problem behaviors.

Several reliable measurements of RRBs exist, including the Repetitive Behavior Questionnaire–2 (RBQ-2),[8] the

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Repetitive Behavior Scale–Revised (RBS-R),[21] the Childhood Routines Inventory (CRI),[7] and the Yale-Brown Obsessive Compulsive Scale (Y-BOCS).[22] However, most of these measurements (the CRI and RBQ-2 being the exceptions) were designed to assess RRBs in clinical populations and in consequence result in near floor effects when used in typical populations. Furthermore, because most of these existing scales were designed to assess behaviors specific to particular disorders, such as ASD and ID (RBS-R) or OCD (Y-BOCS), they do not assess the full range of RRBs that present across different clinical disorders. Moreover, none of these measurements captures RRBs across the entire range of development, precluding direct comparison of RRBs in adults (e.g., parents) and children.

This study presents 2 novel companion instruments inspired by the original CRI[7]: the Childhood Routines Inventory–Revised (CRI-R) and the Adult Routines Inventory (ARI). The original CRI focused on habits and compulsive behavior, whereas the CRI-R and ARI capture a wider range of RRBs, including stereotypies, tics, compulsions, habits, sensory sensitivities, and focused interests, in the context of typical and atypical development in children and adolescents (CRI-R) and adults (ARI) across the entire lifespan. This study examined the factor structure and psychometric properties of these instruments and age-related differences from infancy to adulthood and derived age- and gender-normed *t* scores on a population-based cohort of more than 3,000 families. This study tested the discriminant validity of the scales by comparing individuals with and without NDDs or NPDs, explored the familial pattern of RRBs through an examination of shared variance between children and their parents, and conducted item response theory (IRT) analyses to assess reliability and sensitivity across the scale range.

METHOD

Participants

Participants were recruited through Survey Sampling International (Shelton, CT), which specializes in recruiting demographically representative samples for scientific research in the United States. Eligible participants were sent a link to a Qualtrics survey for completing the questionnaires online. Data were collected on at least 1 parent with at least 1 child. All participants received monetary compensation based on the median time to complete surveys across the entire sample. Complete data on the ARI were collected from 3,108 adults (966 men; mean age 38.15 years, standard deviation [SD] 9.85) and complete data on the CRI-R were collected from 3,032 children (1,574 boys; mean age 9.25 years, SD 4.82, range 1 year 0 months to 17 years 11 months). When available, data on siblings also were collected (n = 844, 51.5% boys, 48.5% girls; mean age 7.99 years, SD 4.14). Where possible, data also were collected from the second parent (n = 217, 55.8% men, 44.2% women; mean age 38.30 years, SD 9.79). Biological parents reported on their children for 89.6% of the dyads (1,914 mothers and 804 fathers), 10.4% were guardians (3.2% stepmother or stepfather, 2.1% adoptive mother or father, 3% grandmother or grandfather, and 1.7% legal guardian). Analyses on the familial nature of RRBs were explored only for those dyads constituted by the biological parent and child. Demographics were representative of the US population for race, income, education, and rural and urban populations,[23] albeit with slight but statistically significant skewing toward lower economic classes (Table 1). Because we recruited a representative sample of the general population, the cohort included families with a lifetime presence of NDD and NPD in 35.8% of adult respondents and 25.6% of children, which is consistent with previous epidemiologic reports on the frequency of NDDs and NPDs in the United States[24,25] (Supplement 1, Table S1, available online).

TABLE 1 Comparison of Socioeconomic Demographics Between Survey Participants and National Statistics

Race/Ethnicity	Survey, %	2010 US Census, %
White	70.2	72.4
African American	14.5	12.6
Hispanic/Latino	11.2	16.4
Asian	6.8	4.8
Asian or Pacific Islander	2.2	0.2
Native American	2.6	0.9
Other	1.2	
Total Household Income	**Survey, %**	**2014 Congressional Research, %**
<$10,000	24.3	7.3
$10,000–$19,999	10.0	11.5
$20,000–$29,999	10.5	10.9
$30,000–$39,999	13.1	10.0
$40,000–$49,999	9.3	8.9
$50,000–$59,999	8.5	7.6
$60,000–$69,999	5.5	6.8
$70,000–$79,999	5.5	5.9
$80,000–$89,999	2.8	4.9
$90,000–$99,999	3.2	4.0
$100,000–$149,999	4.9	12.4
≥$150,000	2.1	9.5

Measurements

The CRI-R is a 62-item parent-report measurement rated on a 5-point Likert scale. The original CRI[7] consisted of 19 items assessing routines, habits, and "compulsive-like" RRBs consistent with symptoms associated with ASD and OCD. However, the original CRI did not encompass certain RRBs that are associated with ASD (e.g., stereotypies) and other NDDs and NPDs. As was the case with the original CRI, items constituting the CRI-R were first derived conceptually by the 2 lead authors after a systematic literature search on RRBs because they present in a range of NDDs and NPDs and of *DSM* criteria for specific disorders while also considering their manifestations in the context of typical development. The final items were retained based on consensus decision and after independent confirmation by a neurodevelopmental pediatrician. Items were chosen to reflect the full range of RRBs seen in normative development and across NDDs and NPDs such as ASD, OCD, tic disorders, schizophrenia, and intellectual disability and included compulsions, motor stereotypies, tics, sensory sensitivities, difficulties with and resistance to minor changes in routine or personal environment, and rituals. When possible, items were cast in a developmentally appropriate context and worded to avoid technical and stigmatizing terms associated with clinical pathology. The same procedure was followed for developing the ARI, an adult self-report measurement that serves as a companion measurement to the CRI-R

for use in family studies or as a stand-alone measurement for research on RRBs in adults. This resulted in 55 items that reflect RRBs associated with the same class of disorders but phrased to be appropriate for adults. A complete list of items is provided in Supplements 2 and 3 (available online).

A subset of parents also completed the Social Responsiveness Scale–2 (SRS-2).[26] The SRS-2 is a widely used measurement that captures dimensions of ASD traits, including 2 subscales tapping RRBs associated with ASD. The SRS was chosen because it is a quantitative measurement of ASD-related behaviors as they manifest in the general population.

Procedures

This study was approved by an institutional review board. All participants reviewed an information document and agreed to participate in the study before completing the surveys.

Four weeks after the initial surveys were administered, a subset of participants was randomly selected for recontact to recomplete the CRI-R and ARI (counterbalanced 1 week apart) or the SRS-2. Completed SRS-2 data were received for 412 parents and 225 children, and retest CRI-R and ARI data were received for 318 parents and 231 children. The CRI-R and ARI scores between the full sample and the respondents who completed the SRS were comparable. Median response time for the ARI and CRI-R was 17 minutes.

Data Analysis

The CRI-R and ARI were examined for latent constructs using principal axis factoring exploratory factor analysis (PAF-EFA). Internal consistency and test-retest reliability were examined. To provide a fine-grained analysis of reliability as a function of score level (conditional reliability across the latent trait), IRT analyses were conducted using graded response models implemented in Mplus 7.3.[27] Separate analyses were conducted for the full parent and child scales and for each EFA-derived subscale. IRT-derived information values can be converted to reliability coefficients, with information values of 3, 5, and 10 reflecting reliability coefficients of 0.67, 0.80, and 0.90, respectively. Convergent validity was examined by comparing the CRI-R and ARI with the SRS-2, and discriminant validity was established by comparing scale scores across various diagnostic groups. Probability density curves and intraclass correlations (ICCs) illustrate the parity of parent–child RRBs (only biological parents were included in this analysis).

RESULTS

Factor Analysis

No more than 2.5% CRI-R or ARI data were missing for any questionnaire item, and there were no systematic differences in missing items. Assumptions of non-multicollinearity, sampling adequacy, and factorability were met. The Kaiser-Meyer-Olkin measurement verified the sampling adequacy for the analysis for the 2 measurements (0.98 for the CRI-R and 0.97 for the ARI). The Bartlett test of sphericity indicated that correlations were sufficiently large for EFA (CRI-R $\chi^2_{1770} = 112{,}503.352$, $p < .001$; ARI $\chi^2_{1378} = 83{,}952.295$, $p < .001$).

Initial PAF-EFA with direct oblimin rotation indicated that 8 components had eigenvalues higher than the Kaiser criterion of 1 for the 2 measurements. Parallel analysis[28,29] was run using the SAS-based code developed by O'Connor[29] to determine the number of factors that should be extracted. Parallel analysis indicated that a 2-factor solution be retained in the final analysis; thus, PAF-EFA was rerun specifying a 2-factor solution. Two factors were interpretable as repetitive sensory motor behaviors/compulsions (RSMBC) and rigidity/insistence on sameness (RIS), accounting for 45.40% (RIS 39.48, RSMBC 5.91) and 42.18% (RSMBC 34.35, RIS 7.82) of the variance for the CRI-R and ARI, respectively (Supplement 1, Tables S2 and S3, available online).

Psychometric Properties

The CRI-R and ARI demonstrated excellent internal consistency (Cronbach α range 0.94–0.96). Four-week test-retest reliability was high for the CRI-R and ARI (ICC ≥ 0.87). The CRI-R total score correlated with the SRS-2 score (Pearson $r = 0.67$) as did the ARI and parent self-report SRS, indicating convergent validity ($r = 0.57$; Table 2).

IRT analyses indicated good to excellent reliability across the full score range for the CRI-R (Figure 1) and ARI (Figure 2), with only slight trailing off of reliability for very low levels of repetitive behavior. Parent and child subscales showed good to excellent reliability (>0.70) across the entire range of IS and RSM scores. The only exception was the

TABLE 2 Psychometric Properties

	ARI MBC	ARI RIS	ARI Total	CRI-R MBC	CRI-R RIS	CRI-R Total
Internal consistency (Cronbach α)	0.94	0.94	0.96	0.95	0.96	0.97
Test-retest reliability ICC (4 wk)[a]	0.88	0.90	0.89	0.88	0.87	0.88
Convergent validity (r; $p < .001$ for all comparisons)[b]						
SRS-2 total	0.59	0.46	0.57	0.66	0.59	0.66
SRS-2 emotion recognition	0.39	0.26	0.36	0.43	0.41	0.44
SRS-2 social avoidance	0.54	0.43	0.53	0.56	0.48	0.54
SRS-2 interpersonal relatedness	0.54	0.40	0.51	0.59	0.51	0.57
SRS-2 insistence on sameness	0.61	0.50	0.61	0.68	0.64	0.69
SRS-2 repetitive mannerisms	0.62	0.440	0.58	0.66	0.51	0.60

Note: ARI = Adult Routines Inventory; CRI-R = Childhood Routines Inventory–Revised; ICC = intercorrelation coefficient; MBC = motor behaviors/compulsions; RIS = rigidity/insistence on sameness; SRS-2 = Social Responsiveness Scale–2.

[a]*For ARI, n = 318; for CRI-R, n = 231.*

[b]*For ARI, n = 418; for CRI-R, n = 418.*

FIGURE 1 Childhood Routines Inventory–Revised (CRI-R) reliability across the full score range. Note: IS = insistence on sameness; RSM = repetitive sensory motor.

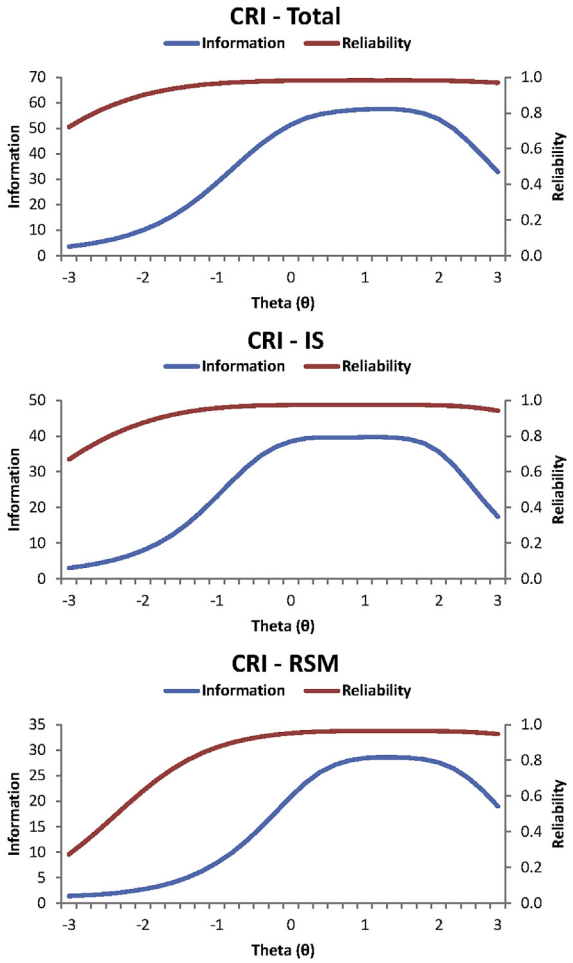
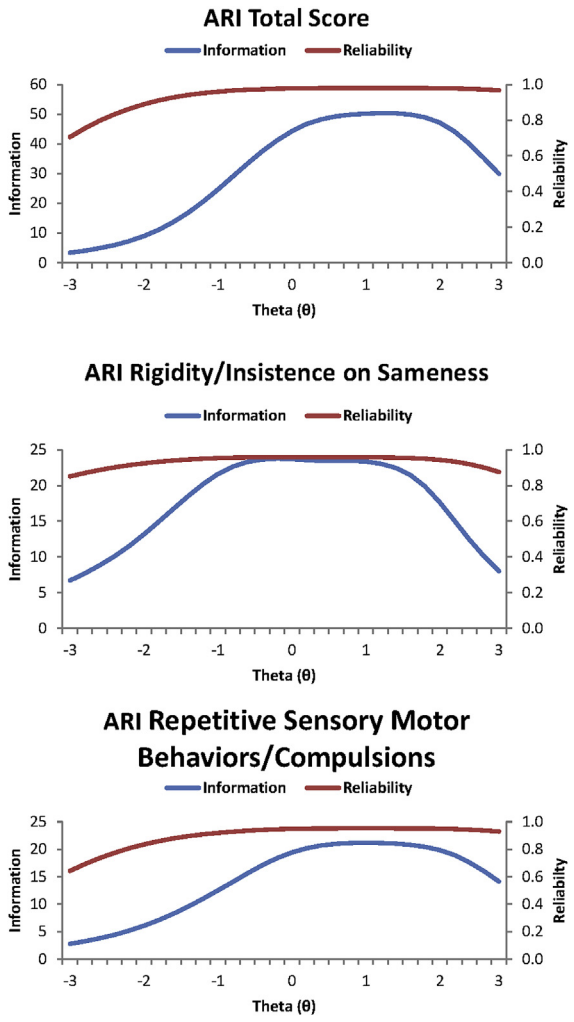


FIGURE 2 Adult Routines Inventory (ARI) reliability across the full score range.



lower reliability for the extreme low end of the score range for adult and child RSM levels. These results suggest that total and subscale scores have very good reliability for clinical and research purposes.

Cross-Sectional Developmental Trajectories

CRI-R raw scores were compared across 4 age groups that represent key developmental phases relevant to RRBs[4]: children no older than 2 years (age group 1, n = 423), 3 to 7 years old (age group 2, n = 784), 8 to 13 years old (age group 3, n = 1,202), and 14 to 17 years old (age group 4, n = 623). A 4 (age group) by 2 (gender) analysis of variance was performed for total CRI-R scores and showed a significant age effect ($F = 20.93$, $p < .0001$, partial eta-squared [η^2] $p = .02$), whereas the effect of sex or age-by-sex interaction term was not significant. Age groups 2 and 3 (who did not differ from each other) exhibited significantly more RRBs than age groups 1 and 4 ($p < .0001$ for all comparisons). Age groups 1 and 4 did not differ. For RSMBC ($F = 4.89$, $p = .002$, η^2 $p = .005$), age groups 1 and 4 had the lowest scores and age groups 2 and 3 had the highest scores. For RIS score, the age main effect was significant ($F = 33.21$, $p < .00001$, η^2 $p = .03$), whereas the sex or sex-by-age group interaction was not. Group 1 had the lowest scores. Significant increases in RIS scores were seen in age groups 2 and 3 (who did not differ from each other), with a decrease in age group 4. Conversions of raw to *t* score were generated so that the authors could validly compare standardized scores across ages and by sex (*t* scores), whereas raw scores allow for the study of developmental changes in absolute levels of RRB (for conversion of raw to *t* score, see Supplement 1, Table S4, available online).

For the ARI, a 5 (18–25 years old, n = 267; 26–35 years old, n = 1,092; 36–45 years old, n = 1,065; 46–55 years old, n = 527; ≥56 years old, n = 157) by 2 (sex) analysis of variance showed a main effect for sex, with men exhibiting higher total ARI raw scores ($F = 7.36$, $p = 0.007$, η^2 $p = .02$) and RSMBC scores ($F = 18.41$, $p < .0001$, η^2 $p = .006$) than women. No significant differences emerged on the RIS factor score. The age group main effect also was significant for total ARI score

($F = 50.58$, $p < .0001$, η^2 $p = .061$), RSMBC score ($F = 71.08$, $p < .0001$, η^2 $p = .084$), and RIS score ($F = 18.91$, $p < .0001$, η^2 $p = .024$). ARI scores decreased with age, albeit cross-sectionally (for conversion of raw to t score, see Supplement 1, Table S5, available online).

Discriminant Validity

Given the high rates of comorbidity across diagnoses, we next created diagnostic clusters that represented groupings of diagnoses. This allowed us to prioritize analysis of rarer and more severe diagnoses (e.g., bipolar and schizophrenia) that are often comorbid with more common diagnoses (e.g., attention-deficit/hyperactivity disorder) and that have been associated with more severe RRBs. The clusters were created along a hierarchy based on prevalence rates and relevance to RRBs. A 1-way between-groups analysis of covariance was conducted to compare total CRI-R (or ARI) scores and each of the subscales across NDD and NPD clusters with chronological age as the covariate (Tables 2 and 3 present descriptive statistics across groups).

Childhood Routines Inventory–Revised. Analyses of covariance (after adjusting for chronological age) were performed for CRI-R total score ($F = 63.14$, $p < .0001$, η^2 $p = .147$), RSMBC score ($F = 68.51$, $p < .0001$, η^2 $p = .158$), and RIS score ($F = 40.91$, $p < .0001$, η^2 $p = .101$). Post hoc comparisons showed that total and subscale scores provided excellent discrimination across diagnostic clusters, with the no-diagnosis group (NDx) having significantly lower scores than the other groups for CRI-R total score ($p < .00001$, Cohen's d ≥ 0.52), RSMBC score ($p < .00001$, Cohen's d ≥ 0.52), and RIS score ($p < .00001$, Cohen's d ≥ 0.50), apart from NDx versus attention deficit disorder/attention-deficit/hyperactivity disorder (Cohen's d = 0.27). For the CRI-R total scores, the highest scores were the OCD/tic disorder and ASD diagnostic clusters, followed by the bipolar/schizophrenia cluster. The ASD and OCD/tic disorder clusters had significantly higher total CRI-R scores (but did not differ significantly from each other) than all other groups ($p < .01$ for all comparisons; for ASD versus other clusters, Cohen's d ≥ 0.48 for all comparisons apart from ASD versus bipolar/schizophrenia, where Cohen's d = 0.24; for OCD versus other clusters, Cohen's d ≥ 0.33 for all comparisons). The RSMBC post hoc tests showed that the OCD/tic disorder, bipolar/schizophrenia, and ASD clusters had significantly higher scores than all other clusters ($p \leq .004$ and Cohen's d ≥ 0.28 for all comparisons). The OCD/tic disorder group was significantly higher than the ASD group ($p = .006$, Cohen's d = 0.36) but not higher than the bipolar/schizophrenia group, which in turn was not significantly higher than the ASD group. For RIS, the ASD cluster had significantly higher scores compared with all other clusters ($p \leq .001$ and Cohen's d ≥ 0.59 for all comparisons), apart from OCD/tic disorders clusters, where the difference did not reach statistical significance (Table 3 presents descriptive statistics).

Adult Routines Inventory. Analyses of covariance (after adjusting for chronological age) were performed for ARI total score ($F = 62.78$, $p < .0001$, η^2 $p = .096$), ARI RSMBC score ($F = 83.79$, $p < .0001$, η^2 $p = .124$), and ARI RIS score ($F = 27.995$ $p < .0001$, η^2 $p = .045$). As presented in Table 3, post hoc tests showed that NDx participants had significantly lower ARI total ($p < .0001$ and Cohen's d ≥ 0.39 for all comparisons), RSMBC ($p < .0001$ and Cohen's d ≥ 0.49 for all comparisons), and RIS ($p < .0001$ and Cohen's d ≥ 0.21 for all comparisons) scores than all other clusters. For the ARI total scores, the ASD cluster had significantly higher scores than all other clusters ($p \leq .02$ and Cohen's d ≥ 0.42 for all comparisons), apart from the OCD/tic disorder group, where the difference did not reach statistical significance. For RSMBC scores, the ASD cluster was significantly higher than all other clusters ($p \leq .01$ and Cohen's d ≥ 0.46 for all comparisons). For RIS, the OCD/tic disorder cluster had significantly higher scores than all other groups ($p \leq .005$ and Cohen's d ≥ 0.45 for all comparisons), apart from ASD, where the difference did not reach statistical significance; in turn ASD was higher than all other clusters ($p \leq .02$ and Cohen's d ≥ 0.43 for all comparisons), apart from bipolar/schizophrenia. Table 4 presents descriptive statistics.

RRBs in Parents and Their Children

The ICCs compared RRBs in biological parent–child dyads (total score ICC = 0.81, $p < .00001$; RMBS score ICC = 0.84, $p < .00001$; RIS score ICC = 0.69, $p < .00001$). Because there is a risk that parents' response patterns for their self-reports and reports of their children could inflate the ICCs, we also

TABLE 3 Childhood Routines Inventory–Revised (CRI-R) Scores Across Cluster Diagnoses

Cluster Diagnoses	n	CRI-R Total, Mean (SD)	CRI-R RSMBC, Mean (SD)	CRI-R RIS, Mean (SD)
No diagnosis	2,183	123.42 (40.77)	42.61 (16.60)	80.81 (26.99)
ADD/ADHD	145	140.44 (45.89)	52.18 (19.45)	88.25 (28.72)
ODD/CD	43	149.76 (52.90)	55.39 (20.88)	94.37 (35.95)
Speech disorder	168	151.97 (53.23)	56.11 (23.41)	95.86 (32.07)
Depression/anxiety	139	156.93 (51.65)	59.21 (23.75)	97.72 (31.97)
ID/DD	65	162.35 (52.46)	58.35 (22.60)	104.00 (32.37)
BD/schizophrenia	42	174.16 (50.27)	70.86 (22.48)	103.31 (31.47)
ASD	92	186.46 (48.51)	64.76 (22.41)	121.69 (30.25)
OCD/tic disorder	54	191.44 (52.56)	73.26 (24.36)	118.18 (31.41)

Note: ADD/ADHD = attention deficit disorder/attention-deficit/hyperactivity disorder; ASD = autism spectrum disorder; BD = bipolar disorder; CD = conduct disorder; DD = developmental disability; ID = intellectual disability; OCD = obsessive-compulsive disorder; ODD = oppositional defiant disorder; RIS = rigidity/insistence on sameness; RSMBC = repetitive sensory motor behaviors/compulsions; SD = standard deviation.

TABLE 4 Adult Routines Inventory (ARI) Scores Across Cluster Diagnoses

Cluster Diagnoses	n	ARI Total, Mean (SD)	ARI RSMBC, Mean (SD)	ARI RIS, Mean (SD)
No diagnosis	1,959	123.24 (33.43)	56.89 (19.09)	66.34 (17.74)
ADD/ADHD	70	147.23 (38.99)	75.26 (22.31)	71.97 (18.47)
Depression/anxiety	692	136.35 (33.01)	66.39 (19.36)	69.96 (17.32)
ASD	24	172.46 (46.48)	91.42 (25.44)	81.04 (22.91)
OCD/tic disorder	71	166.00 (32.15)	80.90 (19.97)	85.09 (15.71)
Bipolar/schizophrenia	157	155.53 (36.75)	78.08 (21.67)	77.44 (18.25)

Note: ADD/ADHD = attention deficit disorder/attention-deficit/hyperactivity disorder; ASD = autism spectrum disorder; OCD = obsessive-compulsive disorder; RIS = rigidity/insistence on sameness; RSMBC = repetitive sensory motor behaviors/compulsions; SD = standard deviation.

examined the parent–child ICCs between the child and the other parent, the parent who did not complete the CRI-R (total score ICC = 0.82, $p < .00001$; RMBS score ICC = 0.87, $p < .00001$; RIS score ICC = 0.68, $p < .00001$). As noted, CRI-R data also were available for 844 sibling pairs (total score sibling–sibling ICC = 0.83, $p < .00001$); RMBS score sibling–sibling ICC = 0.87, $p < .00001$; RIS score sibling–sibling ICC = 0.76, $p < .00001$).

To test whether children of parents with an NDD or NPD diagnosis had increased rates of RRBs, we generated "shift plots" comparing parent–child probability density distributions in parents with versus those without an NDD or an NPD. This showed that children whose parents had an NDD or NPD diagnosis were "shifted" 0.37 to 0.50 SD toward greater symptom expression compared with children whose parents did not have a diagnosis (Figure 3), although the parent–child correlations remained high and significant. When comparing the children whose parents had the highest rates of RRBs (i.e., bipolar/schizophrenia, ASD, and OCD/tic disorders groups) with those with any other NDD or NPD and those with NDx, children from the highest parent RRB group were shifted from 0.4 to 0.7 SD toward higher CRI-R total scores from their cohort.

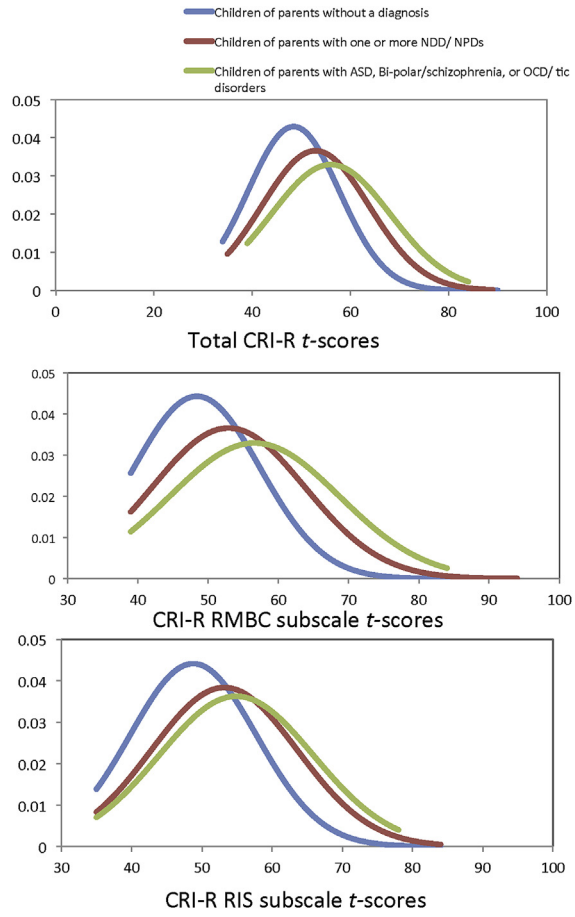
DISCUSSION

The study describes the development, factor structure, and psychometric properties of 2 new dimensional measurements of RRBs across the lifespan. We refined and expanded an existing quantitative measurement of RRB in children, the CRI,[7] to encompass the full spectrum of RRBs, including stereotypies, self-injurious behavior, tics, compulsions, habits, sensory sensitivities, and focused interests. Modeled on the CRI-R but adjusted to reflect the adult period of development, we created and normed the ARI. Our sample was ascertained from a representative population-based cohort and included individuals with and without NDDs and NPDs, consistent with epidemiologic estimates of the frequency of psychiatric disorders in the United States[24,25] and ranging from infancy to older adulthood.

The similar factor structure of the ARI and CRI-R scales, with nearly identical item content in the RSMBC and RIS factors of the CRI-R and ARI, allows for comparisons of RRBs across the lifespan. These factors reported in the present study are consistent with previous research examining the structure of RRB.[10]

The comparability of these measurements also allows for research on RRBs in families—typical families and families that have at least 1 member who has an NDD or NPD—and for examination of the variability of behavioral phenotypes in known genetic syndromes in relation to unaffected family members.[5]

FIGURE 3 Childhood Routines Inventory–Revised (CRI-R) probability density distributions for different parental diagnostic groups. Note: ASD = autism spectrum disorder; NDD = neurodevelopmental disorder; NPD = neuropsychiatric disorder; OCD = obsessive-compulsive disorder; RIS = rigidity/insistence on sameness; RMBC = repetitive motor behaviors/compulsions.



The CRI-R and ARI demonstrated excellent psychometric properties as evidenced by excellent internal validity (Cronbach $\alpha \geq 0.94$ for total and factor scores) and test-retest reliability (ICC ≥ 0.87 for total and factor scores). IRT results suggest that total and subscale scores for the 2 instruments show very good reliability for clinical and research purposes. Convergent validity was demonstrated by comparing the CRI-R and ARI with the SRS-2, a well-established measurement of social behavior. Findings showed significant overlap between the CRI-R and ARI and the SRS-2 and substantial unshared variance. Although there were moderate to high correlations between the ARI and CRI-R scales and the SRS-2 subscales, these relations were strongest between the RIS and RSMBC subscales of the ARI and CRI-R and the 2 RRB subscales of the SRS-2 compared with the 3 SRS-2 subscales that reflect social-communicative and social-emotional traits. We also found distinct patterns of CRI-R and ARI total scores and subscale scores across different NDDs and NPDs. Together, this suggests that the CRI-R and ARI subscales tap specific patterns of behaviors, rather than simply reflecting global distress or psychopathology per se. The CRI-R and ARI also provide a more comprehensive assessment of a wider range of RRBs that appear in a range of NDDs and NPDs than does the SRS, which primarily measures RRBs linked to ASD. However, future research will help determine clinically relevant thresholds on the CRI-R and ARI subscales and will provide validation of these instruments in different clinical cohorts. Furthermore, in future research, it will be important to explore the performance of the ARI and CRI-R against other RRB measurements, including the RBQ-2 and RBQ-2A[8,33] and the Y-BOCS.[22]

The cross-sectional developmental trajectory of CRI raw scores showed a similar developmental pattern as previously shown for the CRI[7] but extends this approach to a broader range of RRBs and through the adolescent period. RRBs increased from infancy to preschool age, plateaued in early school age, then decreased in later adolescence. This trend appears to continue throughout adulthood, which has not heretofore been reported and merits further exploration using longitudinal designs.

Comparisons between children and adolescents with and without an NDD or NPD showed that the CRI discriminates those with an NDD or an NPD across all ages. Even during the preschool period, children with an NDD showed vastly higher rates of RRBs, indicating a magnified rather than developmentally deviant pattern. The potential clinical utility of the CRI-R and ARI was established by comparing the measurements across clinical diagnostic groups. Children with ASD and OCD/tic disorders were found to have the highest total CRI-R and RIS scores. The OCD/tic disorders group had the highest RSMBC scores followed by children with bipolar/schizophrenia and ASD. On the ARI, participants with a history of ASD had the highest RSMBC scores, and individuals in the ASD and OCD/tic disorder groups had the highest ARI total and RIS scores. These results highlight the fact that many clinical diagnoses represent a host of overlapping dimensional traits, and that arbitrary demarcations are often used to separate generic developmental brain dysfunctions into distinct but artificial entities.[30,31]

When interpreting these results, it is necessary to recognize that diagnostic groups were based on self-reported diagnoses of NDDs and NPDs rather than the use of screening questionnaires or diagnostic interviews. However, the reported lifetime frequency of any NDD or NPD is similar to recent prevalence rates estimated for adults in the United States (35.6%)[24,32] and children and adolescents (30.1%).[24] It will be important for future work to provide further validation of the ARI and CRI-R against well-validated dimensional measurements of NDDs and NPDs and diagnostic clinical interviews and assessments. It is critical for future research to go beyond the categorical approach and explore how variation within the domains of self-regulation and reactivity, negative and positive valence systems, cognitive systems, systems for social processes, and arousal/modulation systems proposed by the Research Domain Criteria framework[6] influences the expression of particular types of RRBs.

The high parent–child ICCs between the ARI and CRI-R extend previous work on familial patterns of RRBs in families in which at least 1 individual is diagnosed with ASD.[34] In psychiatric genomics research, there is increasing emphasis on identifying sources of variable expressivity of genetic syndromes. For example, although only 25% of probands with the de novo 16p11.2 deletion meet diagnostic criteria for ASD, we demonstrated[30] that, as a group, probands were shifted toward greater symptom expression (as measured by the SRS) compared with non-carrier first-degree relatives, whether or not the probands met diagnoses for ASD. We demonstrate a similar pattern in this study by noting a shift in the children of parents with NDD or NPD toward higher RRB scores while preserving the parent–child correlation. Although the high parent–child ICC could be influenced by parental response patterns (where 1 parent completed the self-report ARI and the parent-report CRI), when the parent–child dyads compared the child scores with those of the parent who did not complete the child report, the ICCs showed almost identical values, suggesting heritability of the construct. Nevertheless, heritability should be further validated, for example, in comparisons of monozygotic and dizygotic twin pairs.

Restricted and repetitive behaviors are ubiquitous throughout typical development and reflect core symptoms of different NDDs and NPDs. As current approaches to understanding the nature of NDDs and NPDs move beyond the traditional categorical nosology, it becomes increasingly important to establish and validate dimensional measurements that apply to healthy, unhealthy, and at-risk populations across the lifespan. The present measurements serve different purposes. First, they allow for assessment of prodromal symptom expression of RRBs in children and adults at risk for NDD or NPD; second, they facilitate family studies that compare children with parents on compatible measurements of RRBs; third, their dimensional nature provides a sensitivity that will pave the way for establishing reliable biomarkers of core behavioral traits that span across NDDs and NPDs and typical development and across the lifespan. These are the first measurements of RRB that have been validated in a nationally representative, population-based sample. &

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