

Open access · Journal Article · DOI:10.1016/J.JAAC.2016.10.014

Development of Two Dimensional Measures of Restricted and Repetitive Behavior in Parents and Children. — Source link 🖸

David W. Evans, Mirko Uljarević, Laina G. Lusk, Eva Loth ...+1 more authors

Institutions: Bucknell University, Cooperative Research Centre, King's College London, Boston Children's Hospital Published on: 01 Jan 2017 - Journal of the American Academy of Child and Adolescent Psychiatry (Elsevier) Topics: Population, Autism spectrum disorder and Stereotypic movement disorder

Related papers:

- Ritual, habit, and perfectionism : The prevalence and development of compulsive-like behavior in normal young children
- Restricted and repetitive behaviors in autism spectrum disorders: a review of research in the last decade.
- Varieties of repetitive behavior in autism: comparisons to mental retardation.
- Subcategories of restricted and repetitive behaviors in children with autism spectrum disorders.
- · Repetitive behaviours in typically developing 2-year-olds







King's Research Portal

DOI: 10.1016/j.jaac.2016.10.014

Document Version Peer reviewed version

Link to publication record in King's Research Portal

Citation for published version (APA):

Evans, D. W., Uljarevic, M., Lusk, L. G., Loth, E., & Frazier, T. (2016). Development of Two New Dimensional Measures of Restricted and Repetitive Behaviors in Parents and Children. *Journal of the American Academy of Child and Adolescent Psychiatry*. https://doi.org/10.1016/j.jaac.2016.10.014

Citing this paper

Please note that where the full-text provided on King's Research Portal is the Author Accepted Manuscript or Post-Print version this may differ from the final Published version. If citing, it is advised that you check and use the publisher's definitive version for pagination, volume/issue, and date of publication details. And where the final published version is provided on the Research Portal, if citing you are again advised to check the publisher's website for any subsequent corrections.

General rights

Copyright and moral rights for the publications made accessible in the Research Portal are retained by the authors and/or other copyright owners and it is a condition of accessing publications that users recognize and abide by the legal requirements associated with these rights.

•Users may download and print one copy of any publication from the Research Portal for the purpose of private study or research. •You may not further distribute the material or use it for any profit-making activity or commercial gain •You may freely distribute the URL identifying the publication in the Research Portal

Take down policy

If you believe that this document breaches copyright please contact librarypure@kcl.ac.uk providing details, and we will remove access to the work immediately and investigate your claim.

Accepted Manuscript

Development of Two New Dimensional Measures of Restricted and Repetitive Behaviors in Parents and Children

David W. Evans, PhD, Mirko Uljarevic, MD, PhD, Laina G. Lusk, BS, Eva Loth, PhD, Thomas Frazier, PhD

PII: S0890-8567(16)31890-1

DOI: 10.1016/j.jaac.2016.10.014

Reference: JAAC 1658

- To appear in: Journal of the American Academy of Child & Adolescent Psychiatry
- Received Date: 13 July 2016
- Revised Date: 29 September 2016
- Accepted Date: 19 October 2016

Please cite this article as: Evans DW, Uljarevic M, Lusk LG, Loth E, Frazier T, Development of Two New Dimensional Measures of Restricted and Repetitive Behaviors in Parents and Children, *Journal of the American Academy of Child & Adolescent Psychiatry* (2016), doi: 10.1016/j.jaac.2016.10.014.

This is a PDF file of an unedited manuscript that has been accepted for publication. As a service to our customers we are providing this early version of the manuscript. The manuscript will undergo copyediting, typesetting, and review of the resulting proof before it is published in its final form. Please note that during the production process errors may be discovered which could affect the content, and all legal disclaimers that apply to the journal pertain.



Development of Two New Dimensional Measures of Restricted and Repetitive Behaviors in Parents and Children

RH: Repetitive Behaviors in Parents and Children

David W. Evans, PhD, Mirko Uljarevic, MD, PhD, Laina G. Lusk, BS, Eva Loth, PhD, Thomas Frazier, PhD

Supplemental material cited in this article is available online.

Accepted October 28, 2016

Dr. Evans is with Bucknell University, Lewisburg, PA. Dr. Uljarevic is with Olga Tennison Autism Research Centre, Cooperative Research Centre for Living with Autism (Autism CRC), School of Psychology and Public Health, La Trobe University, Victoria 3086, Australia. Ms. Lusk is with Geisinger-Bucknell Autism and Developmental Medicine Institute, Lewisburg, PA. Dr. Loth is with Institute of Psychiatry, Psychology and Neuroscience, King's College London, London, UK and Sackler Institute for Translational Neurodevelopment, Institute of Psychiatry, Psychology and Neuroscience, King's College London. Dr. Frazier is with Cleveland Clinic Children's Hospital Center for Pediatric Behavioral Health, Cleveland, Ohio.

Research reported in this study was supported by the National Institute of Mental Health of the National Institutes of Health under award number RO1MH074090. The content is solely the responsibility of the authors and does not necessarily represent the official views of the National Institutes of Health. M.U. was supported by an Autism CRC postdoctoral fellowship. E.L. was supported by EU-AIMS (European Autism Interventions), which receives support from the Innovative Medicines Initiative Joint Undertaking under grant agreement no. 115300, the resources of which are composed of financial contributions from the European Union's Seventh Framework Programme (grant FP7/2007-2013), from the European Federation of Pharmaceutical Industries and Associations companies' in-kind contributions, and from Autism Speaks. Drs. Frazier, Uljarevic, and Evans served as the statistical experts for this research. The authors would like to thank James F. Leckman, MD, Yale School of Medicine, Child Study Center, for his feedback and insightful comments, and to Scott M. Myers, MD, of the Gesinger Health System, for his feedback on the items.

Disclosure: Drs. Evans, Uljarevic, Loth, Frazier, and Ms. Lusk report no biomedical financial interests or potential conflicts of interest.

Correspondence to David W. Evans, PhD, Department of Psychology, Bucknell University, Lewisburg, PA, USA; email: <u>dwevans@bucknell.edu</u>.

ABSTRACT

Objective: Restricted and repetitive behaviors (RRBs) are a heterogeneous set of behaviors common across a wide range of neurodevelopmental (NDD) and neuropsychiatric disorders (NPD) that extend well into the general population. We introduce two dimensional measures of RRBs for use in both 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 nationality representative cohort of 3,108 parents with 3,032 children (range 12 months – 17 years; 11 months). Twenty-six percent of children and thirty six percent of adults had one or more NDDs/NPDs.

Results: Principal axis factoring exploratory analysis revealed a two-factor structure for both instruments (Motor Behaviors/Compulsions and Rigidity/Insistence on Sameness). Analyses for convergent and discriminant validity, internal consistency (Cronbach $\alpha \ge .94$), and testretest reliability (r $\ge .87$) all indicated strong psychometric properties. Item response theory (IRT) analyses indicated strong reliability across the score range for both instruments. RRB rates varied across development, peaking between the preschool and school years. Children with NDD/NPDs (particularly those with autism spectrum disorder [ASD], schizophrenia/bipolar, obsessive-compulsive disorder [OCD]/tic disorders) had elevated RRBs relative to those with no diagnosis. Parent–child (.69-.84) and sibling–sibling (.76-.87) intraclass correlations indicate high heritability. Children of parents with an NDD/NPD exhibited more RRBs relative to children of parents without NDD/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, as well as for the identification of prodromal conditions involving RRB.

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

1

INTRODUCTION

Restricted and repetitive behaviors (RRBs) comprise a broad range of behaviors including simple motor stereotypies and tics as well as 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 (NDD) and neuropsychiatric disorders (NPD), such as intellectual (ID) and developmental disabilities (DD), schizophrenia,, obsessive-compulsive disorder (OCD), and tic disorders (e.g., Tourette syndrome).^{2,3} Contrary to traditional categorical nosological boundaries, recent findings indicate that the majority of these NDD/NPD are overlapping syndromes⁴ that are best represented as a collection of dimensional traits that extend into the general population.^{5,6}

RRBs are also 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 between 2-7 years of age.^{9,10} These typical RRBs may serve a variety of adaptive roles including motor^{11,12} and nervous system maturation,¹³ and emotional and arousal regulation.^{10,14} In the context of NDDs/NPDs, however, RRBs adversely impact multiple aspects of functioning¹⁵ and are therefore 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 among individuals with NDDs/NPDs,^{17,18} such disorders also frequently co-occur within families.¹⁹ In some instances, family members may share a common diagnosis, or different symptoms may manifest as alternative phenotypes of a common genotype.²⁰ It is important, therefore, to understand the familial context of RRBs, to determine whether children of parents with one or more NDDs/NPDs

show increased levels of RRBs that may signify prodromal states, and to enable early identification and intervention of problem behaviors.

Several reliable measures of RRBs exist, including the Repetitive Behavior Questionnaire-2 (RBQ-2),⁸ the 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 measures (the CRI and RBQ-2 being the exceptions) were designed to assess RRBs in clinical populations, and consequently result in near floor-effects when used in typical populations. Furthermore, because most of these existing scales were designed to assess behaviours specific to particular disorders, such as ASD and intellectual disability (ID; RBS-R) or OCD (Y-BOCS), they also do not assess the full range of RRBs that present across different clinical disorders. Finally, none of these measures captures RRBs across the entire range of development, precluding direct comparison of RRBs in adults (e.g., parents) and children.

Here we present two novel companion instruments inspired by the original CRI⁷: the Childhood Routines Inventory-Revised (CRI-R), and the Adult Routines Inventory (ARI). Whereas the original CRI focused on habits and compulsive behavior, 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 both typical and atypical development, in children and adolescents (CRI-R) and adults (ARI), across the entire lifespan. We examine the factor structure and psychometric properties of these instruments, age-related differences from infancy to adulthood, and derive age- and gender-normed t-scores on a population-based cohort of over 3,000 families. We test the discriminant validity of the scales by comparing individuals with and without NDDs/NPDs, explore the familial pattern of RRBs through an examination of shared variance between children and their parents, and conduct item response theory (IRT) analyses to assess reliability and sensitivity across the scale range.

METHOD

Participants

Participants were recruited through Survey Sampling International (SSI), who specialize 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 one parent with at least one 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 (Mage= 38.15 years, SD= 9.85, 966 males) and on the CRI-R from 3,032 children (M_{age} = 9.25 years, SD= 4.82, range: 1 year, 0 months to 17 years, 11 months, 1,574 males). When available, data on siblings was also collected (n=844; mean age= 7.99 years; SD= 4.14; 51.5% male, 48.5% female). Where possible, data were also collected from the second parent as well (n= 217; mean age= 38.30 years; SD= 9.79; 55.8% male, 44.2% female). Biological parents reported on their children for 89.6% of the dyads (1,914 mothers and 804 fathers), 10.4% guardians (3.2%) stepmother/stepfather, 2.1% adoptive mother/father, 3% were grandmother/grandfather, and 1.7% legal guardian). Analyses on the familiality of RRBs were explored only for those dyads comprising the biological parent and child. Demographics were representative of the US population in terms of race, income, education, and rural/ urban populations³⁰, albeit with slight but statistically significant skewing toward lower economic classes (see 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 epidemiological reports on the frequency of NDDs/NPDs in the United States^{31,32} (see Supplement 1, Table S1, available online).

Table 1 here

Measures

The CRI-R is a 62-item, parent-report measure rated on a five-point Likert scale. The original CRI⁷ comprised 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/NPDs. As was the case with the original CRI, items comprising the CRI-R were first derived conceptually by the two lead authors, following a systematic literature search on RRBs as they present in a range of NDDs/NPDs, as well as DSM criteria for specific disorders, while also considering their manifestations in the context of typical development. The final items were retained based on the 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/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. Where possible, items were cast in a developmentally appropriate context, and were worded to avoid technical/stigmatizing terms associated with clinical pathology. The same procedure was followed for developing the ARI, an adult self-report measure that serves as a companion measure to the CRI-R for use in family studies, or as a stand-alone measure 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).²³ The SRS-2 is a widely used measure that captures dimensions of ASD traits, including two

subscales tapping RRBs associated with ASD. The SRS was chosen as it is a quantitative measure 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 again complete the CRI-R and ARI (counter-balanced one week apart), or the SRS-2. Completed SRS-2 data were received for 412 parents and 225 children, and re-test CRI-R/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 v7.3²⁴. Separate analyses were conducted for the full parent and child scales and for each of the exploratory factor analysis-derived subscales. 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/ARI to the SRS-2, and discriminant validity was established by comparing scale scores across various diagnostic groups. Probability density curves and intraclass correlations 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 all met. The Kaiser-Meyer-Olkin (KMO) measure verified the sampling adequacy for the analysis for both measures: KMO =.98 for the CRI-R and .97 for the ARI. Bartlett's test of sphericity indicated that correlations were sufficiently large for exploratory factor analysis (CRI-R: χ^2 [1770] = 112503.352, *p*<.001; ARI: χ^2 [1378] = 83952.295, *p*<.001).

Initial principal axis factoring exploratory factor analysis (PAF-EFA) with direct oblimin rotation indicated that eight components had eigenvalues over Kaiser's criterion of 1 for both measures. Parallel analysis (PA^{25,26}) was run using the SAS-based code developed by O'Connor²⁶ to determine the number of factors that should be extracted. PA indicated that a two-factor solution be retained in the final analysis and thus PAF-EFA was rerun specifying a two-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 (see Supplement 1, Tables S2 and S3, available online).

Psychometric Properties

Both the CRI-R and ARI demonstrated excellent internal consistency (Cronbach α ranged from .94 to.96). Four-week test-retest reliability was high for both the CRI-R and ARI (ICC \geq .87). Convergent validity: the CRI-R total score correlated with the SRS-2 (Pearson r=.67) as did the ARI and the parent self-report SRS (r=.57) (See Table 2).

Table 2 here

IRT analyses indicated good to excellent reliability across the full score range for the ARI and CRI-R, with only slight trailing off of reliability for very low levels of repetitive behavior (see Figure 1 for CRI-R and Figure 2 for ARI). Parent and child subscales showed good to excellent reliability (>.70) across the whole range of IS and RSM. The only exception was the 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 both clinical and research purposes.

Figures 1 and 2 here

Cross-Sectional Developmental Trajectories

CRI-R raw scores were compared across four age groups that represent key developmental phases relevant to RRBs⁴: Children ≤ 2 (Age Group 1; n=423); 3 to 7 (Age Group 2; n= 784); 8 to 13 (Age Group 3; n= 1,202); and 14 years to 17 years (Age Group 4; n=623). A 4 (Age Group) X 2 (gender) analysis of variance (ANOVA) was performed for total CRI-R scores and revealed a significant age effect (F= 20.93, p< .0001, partial ηp^2 = .02), while the effect of sex, or the age X 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 (all p< .0001). Groups 1 and 4 did not differ. For RSMBC (F=4.89, p = .002, partial ηp^2 = .005), Age Groups 1 and 4 had the lowest scores, and Age Groups 2 and 3 had the highest scores. For RIS the age main effect was significant (F=33.21, p < .00001, partial $\eta p^2 = .03$) while the sex or the sex X 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. Raw-to-T-score conversions were generated so that investigators could validly compare standardized scores across ages and by sex (T-scores), while raw scores allow for studying developmental changes in absolute levels of RRB (for Raw to T scores conversion, see Supplement 1, Table S4, available online).

For the ARI, a 5 (Age Group: 18-25, n= 267; 26-35, n= 1,092; 36-45, n= 1,065; 46-55, N= 527; 56 and older, N= 157) X 2 (sex) ANOVA revealed a main effect for sex with males exhibiting higher Total ARI raw (F=7.36, p= 0.007, partial ηp^2 = .02), and RSMBC scores (F= 18.41, p< .0001, partial ηp^2 = .006) than females. No significant differences emerged on the RIS factor score. The Age Group main effect was also significant for Total ARI (F=50.58, p< .0001, partial ηp^2 = .061), RSMBC (F= 71.08, p< .0001, partial ηp^2 = .084) and RIS (F= 18.91, p< .0001, partial ηp^2 = .024). ARI scores decrease with age, albeit crosssectionally (for raw to T scores conversion, 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., ADHD) 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 one-way between-groups analysis of covariance (ANCOVA) was conducted to compare total CRI-R (or ARI) scores and each of the subscales across NDD/NPD clusters with chronological age as covariate (see Tables 2 and 3 respectively for descriptive statistics across groups).

CRI-R. ANCOVAs (after adjusting for chronological age) were as follows: CRI-R total score F=63.14, p < .0001, partial ηp^2 = .147; RSMBC F=68.51, p < .0001, partial ηp^2 = .158; RIS F=40.91, p < .0001, partial ηp^2 = .101. Post hoc comparisons demonstrate that both total and subscale scores provided excellent discrimination across diagnostic clusters with the No Diagnosis group (NDx) being significantly lower than all other groups in case of CRI-R total, RSMBC, and RIS scores (all p < .00001 and Cohen's d for all comparisons $\geq .52$ for CRI-R total total score, $\geq .52$ for RSMBC and $\geq .50$ for RIS, apart from NDx vs attention deficit disorder

[ADD]/ADHD comparison where Cohen's d= .27). For the CRI-R total scores, the highest scores were the OCD/tic disorder and ASD diagnostic clusters, followed by the Bipolar/Schizophrenia group. 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 (all p<.01; for ASD vs other clusters, all Cohen's d \geq .48 apart from ASD vs bipolar/schizophrenia comparison, where Cohen's d was .24; for OCD vs other clusters, all Cohen's d \geq .33). The RSMBC post hoc tests revealed that the OCD/tic disorder, bipolar/schizophrenia, and ASD clusters had significantly higher scores than all other clusters (all $p \leq$.004, all Cohen's d \geq .28). The OCD/tic disorder group was significantly higher than the ASD group (p= .006, Cohen's d= .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 (apart from OCD/tic disorders clusters, where the difference did not reach statistical significance) with all $p \leq$.001 and Cohen's d for all comparisons \geq .59 (see Table 3 for descriptive statistics).

ARI. ANCOVAs (after adjusting for chronological age) were as follows: ARI total score F=62.78, p < .0001, partial $\eta p^2 = .096$; ARI RSMBC F=83.79, p < .0001, partial $\eta p^2 = .124$; ARI RIS F=27.995 p < .0001, partial $\eta p^2 = .045$. As displayed in Table 3, post hoc tests revealed that NDx participants had significantly lower ARI total (all p < .0001, all Cohen's d $\geq .39$), RSMBC (all p < .0001, all Cohen's d $\geq .49$), and RIS (all p < .0001, all Cohen's d $\geq .21$) scores than all other clusters. For the ARI total scores, the ASD cluster had significantly higher scores than all other clusters (all $p \leq .02$, all Cohen's d $\geq .42$) 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 (all $p \leq .01$, all Cohen's d $\geq .46$). For RIS, the OCD/tic disorder cluster had significantly higher scores than all other groups (all $p \leq .005$, all Cohen's d $\geq .45$) apart from ASD, where difference did not reach statistical significantly higher scores than all other groups (all $p \leq .005$, all Cohen's d $\geq .45$) apart from ASD, where difference did not reach statistical significantly higher scores than all other groups (all $p \leq .005$, all Cohen's d $\geq .45$) apart from ASD, where difference did not reach statistical significantly higher scores than all other groups (all $p \leq .005$, all Cohen's d $\geq .45$) apart from ASD, where difference did not reach statistical significantly higher scores than all other groups (all $p \leq .005$, all Cohen's d $\geq .45$) apart from ASD, where difference did not reach statistical significantly higher scores than all other groups (all $p \leq .005$, all Cohen's d $\geq .45$) apart from ASD, where difference did not reach statistical significantly higher scores than all other groups (all $p \leq .005$, all Cohen's d $\geq .45$) apart from ASD, where difference did not reach statistical significantly higher scores than all other groups (all $p \leq .005$, all Coh

significance; in turn, ASD was higher than all other clusters (all $p \le .02$, all Cohen's $d \ge .43$) apart from bipolar/schizophrenia. See Table 4 for descriptive statistics.

Tables 3 and 4 here

RRBs in Parents and Their Children

Intraclass correlations (ICC) compared biological parent–child dyads' RRBs. These were as follows: Total Score= .81 (p<.00001); RMBS= .84 (p<.00001); RIS=.69 (p<.00001). Because there is a risk that parents' response patterns for their self-reports and reports of their children may inflate the ICCs, we also examined the parent–child ICCs between the child and the other parent, the parent who did not complete the CRI-R. ICCs were: Total Score= .82 (p<.00001); RMBS= .87 (p<.00001); RIS= .68 (p<.00001). As noted, CRI-R data were also available on 844 sibling pairs. The sibling-sibling ICCs were: Total score= .83 (p<.00001); RMBS = .87 (p<.00001); RIS = .76 (p<.00001).

To test whether children of parents with an NDD/NPD diagnosis have elevated rates of RRBs, we generated "shift plots" comparing parent–child probability density distributions between parents with versus those without an NDD/NPD. This showed that children whose parents have a NDD/NPD diagnosis were "shifted" .37 to .50 SD toward greater symptom expression relative to children whose parents did not have a diagnosis (see Figure 3), while 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) to a) those with any other NDD/NPD and b) those with NDx, children from the highest parent RRB group were shifted from.4 to .7 SD towards higher CRI-R total scores from their cohort.

Figure 3 Here

DISCUSSION

The study describes the development, factor structure, and psychometric properties of two new dimensional measures of RRBs across the lifespan. We have refined and expanded an existing quantitative measure of RRB in children, the Childhood Routines Inventory,⁷ to encompass the full spectrum of RRBs, including stereotypies, self-injurious behavior, tics, compulsions, habits, sensory sensitivities, and focused interests. Modelled on the CRI-R but adjusted to reflect the adult period of development, we created and normed the Adult Routines Inventory (ARI). Our sample was ascertained from a representative population-based cohort and included individuals both with and without NDD/NPDs, consistent with epidemiological estimates of the frequency of psychiatric disorders in the US^{31,32}, 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 Repetitive Sensory-Motor Behavior/Compulsions (RSMBC) and Rigidity/Insistence on Sameness (RIS) factors of the CRI-R and ARI, allows for comparisons of RRBs across the lifespan. These factors reported here are consistent with previous research examining the structure of RRB.¹⁰

The comparability of these measures also allows for research on RRBs in families both typical families, and where one or more family members have NDDs/NPDs—and to examine the variability of behavioral phenotypes in known genetic syndromes relative to unaffected family members⁵.

Both the CRI-R and ARI demonstrated excellent psychometric properties as evidenced by excellent internal validity (Cronbach's $\alpha \ge .94$ for both total and factor scores) and test-retest reliability (ICCs $\ge .87$ for both total and factor scores). Item response theory analyses results suggest that total and subscale scores for both instruments show very good reliability for clinical and research purposes. Convergent validity was demonstrated by comparing the CRI-R and ARI to a well-established measure of social behaviour: the Social

Responsiveness Scale-2 (SRS-2). Findings revealed significant overlap between the CRI-R and ARI with the SRS-2, while also revealing substantial unshared variance. Although there were moderate to high correlations between the ARI and CRI-R scales and the SRS-2 subscales, the strengths of these relationships were highest between the RIS and RSMB ARI/CRI-R subscales and the two RRB subscales of the SRS-2, relative to the three SRS-2 subscales that reflect social-communicative and social-emotional traits. We also found distinct patterns of CRI-R/ARI total scores and subscale scores across different NDDs/ NPDs. Together, this suggests that the CRI-R/ARI subscales tap specific patterns of behaviours, 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 NDD/NPD than does the SRS, which primarily measures RRBs linked to ASD. However, future research will help determine clinically relevant thresholds on the CRI-R/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/CRI-R against other RRB measures, including the RBQ-2 and RBQ-2A⁸ and the Y-BOCS.²²

The cross-sectional developmental trajectory of CRI raw scores revealed a similar developmental pattern as previously shown for the CRI⁷ but extends this approach to a broader range of RRBs and through the adolescent period. RRB increased from infancy to preschool age, plateaued in early school age, then diminished 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/adolescents with and without an NDD/NPD revealed that the CRI discriminates those with an NDD/NPD across all ages. Even during the preschool period, children with an NDD showed vastly elevated 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 measures across clinical diagnostic groups. Children with ASD and OCD/tic disorders were reported 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.^{27,28}

When interpreting these results, it is necessary to recognize that diagnostic groups were based on self-reported diagnoses of NDDs/NPDs rather than the use of screening questionnaires or diagnostic interviews. However, the reported lifetime frequency of any NDD/NPD is similar to recent prevalence rates estimated for adults in the US (35.6%)^{31,33} and children/adolescents (30.1%).³¹ It will be important for future work to provide further validation of the ARI/CRI-R against well-validated dimensional measures of NDDs/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 (RDoC) framework⁶, influence the expression of particular types of RRBs.

The high parent-child intraclass correlations between the ARI and CRI-R extend previous work on familial patterns of RRBs in families where at least one individual is diagnosed with ASD.²⁹ 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 demonstrated²⁷ that, as a group, probands were shifted toward greater symptom expression (as measured by the SRS) relative to non-carrier first degree relatives, whether or not the probands met diagnoses for ASD. We demonstrate a similar pattern here, noting a shift in the children of parents with NDD/NPD towards higher RRB scores while preserving the parent–child correlation. Although the high parent–child ICC could be influenced by parental response patterns (where one parent completed both the self-report ARI and the parent-report CRI), when the parent–child dyads compared the child scores to 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, including comparisons of monozygotic and dizygotic twin pairs, for example.

Restricted and repetitive behaviors are ubiquitous throughout typical development and also reflect core symptoms of a variety of NDDs/NPDs. As current approaches to understanding the nature of NDDs/NPDs move beyond the traditional categorical nosology, it becomes increasingly important to establish and validate dimensional measures that apply to healthy, unhealthy, and at risk populations across the lifespan. The measures presented here serve a variety of purposes. First, they allow for assessment of prodromal symptom expression of RRBs in children and adults at risk for NDD/NPD; secondly, they facilitate family studies that compare children to parents on compatible measures 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 NDD/NPD and typical development, and across the lifespan. These are the first measures of RRB that have been validated in a nationally representative, population-based sample.

References

- American Psychiatric Association. Diagnostic and Statistical Manual of Mental Disorders (5th ed). Arlington: American Psychiatric Publishing; 2013.
- 2. Mahone EM, Bridges D, Prahme C, Singer HS. Repetitive arm and hand movements (complex motor stereotypies) in children. *Journal of Pediatrics* 2004;145:391–395.
- Langen M, Durston S, Kas MJH, van Engeland H, Staal WG. The neurobiology of repetitive behavior: ...and men. Neuroscience and Biobehavioral Reviews 2011;35(3):356–65.
- 4. Hyman E. Introducing a glimmer of light for neuropsychiatric disorders. *Nature* 2008;455:890-893.
- 5. Moreno De Luca A, Myers SM, Challman TD, Moreno-De-Luca D. Evans DW, Ledbetter DH. Developmental brain dysfunction (DBD): Revival and expansion of an old concept based on new genetic evidence. *The Lancet Neurology* 2013;12(4):406-414.
- Insel T. The NIMH Research Domain Criteria (RDoC) Project: Precision Medicine for Psychiatry. Am J Psychiatry. 2014;171:395-397.
- Evans DW, Leckman JF, Carter A, et al. Ritual, habit and perfectionism: The prevalence and development of compulsive-like behavior in normal young children. *Child Development* 1997;68:58–68.
- Leekam SR, Tandos J, McConachie H, et al. Repetitive behaviors in typically developing
 2-year-olds. J Child Psychol Psychiatry. 2007;48:1131–1138.
- Cevikaslan A, Evans DW, Dedeoglu C, Kalaca S, Yazgan Y. A cross-sectional survey of repetitive behaviors and restricted interests in a typically developing turkish child population. *Child Psychiatry Hum Devel.* 2013;45(4):472-82.
- 10. Leekam SR, Prior MR, Uljarević M. Restricted and repetitive behaviors in autism spectrum disorders: A review of research in the last decade. *Psychol Bull*.

2011;137:562-593.

- 11. Wolff PH. Stereotypic behavior and development. *Canadian Psychologist*. 1968;9:474-485.
- 12. Thelen E. Rhythmical stereotypies in normal human infants. *Anim Behav.* 1979;27:699–715.
- 13. Sprague RL, Newell KM (Eds). *Stereotyped movements: Brain and behavior relationships*. Washington, DC: American Psychological Association; 1996.
- 14. Evans DW, Canavera K, Klinepeter FL, Taga K, Maccubbin E. The fears, phobias and anxieties of children with autism spectrum disorders and Down syndrome:
 Comparisons with developmentally and chronologically age matched children. *Child Psychiatry and Human Development* 2005;36:3-26.
- 15. Nuzzolo-Gomez R, Leonard MA, Ortiz E, Rivera CM, Greer DR. Teaching children with autism to prefer books or toys over stereotypy or passivity. *Journal of Positive Behavior Interventions* 2002;4:80–88.
- 16. Grahame V, Brett D, Dixon L, et al. Managing Repetitive Behaviours in Young Children with Autism Spectrum Disorder (ASD): Pilot Randomised Controlled Trial of a New Parent Group Intervention. J Autism Devel Disord. 2015;45:3168-82.
- 17. Simonoff E, Pickles A, Charman T, Chandler S, Loucas T, Baird G. Psychiatric disorders in children with autism spectrum disorders: prevalence, comorbidity, and associated factors in a population-derived sample. *J Am Acad Child Adolesc Psychiatry*. 2008;47:921-9.
- 18. Meier SM, Petersen L, Schendel DE, Mattheisen M., Mortensen PB., Mors O. Obsessive-Compulsive Disorder and Autism Spectrum Disorders: Longitudinal and Offspring Risk. PLOS ONE 2015;10(11):e0141703
- 19. Jokiranta E, Brown AS, Heinimaa M, Cheslack-Postava K, Suominen A, Sourander A.

Parental psychiatric disorders and autism spectrum disorders. *Psychiatry Res.* 2013;207:203-11.

- 20. Lee SH, Ripke S, Neale BM, et al. Genetic relationship between five psychiatric disorders estimated from genome-wide SNPs. *Nature Genetics* 2013;45(9):984-994.
- 21. Bodfish JW, Symons FJ, Lewis MH. *The Repetitive Behavior Scale*. Morgantown, NC: Western Carolina Center Research Reports; 1997.
- 22. Goodman WK, Price LH, Rasmussen SA, et al. The Yale- Brown Obsessive-Compulsive Scale, I: development, use, and reliability. *Arch Gen Psychiatry*. 1989;46:1006–1011.
- 23. Constantino JN, Gruber CP. *The Social Responsiveness Scale Manual, Second Edition* (*SRS-2*). Los Angeles, CA: Western Psychological Services; 2012.
- 24. Muthén LK, Muthén BO. *Mplus User's Guide. Seventh Edition.* Los Angeles, CA: Muthén and Muthén; 1998-2012
- 25. Horn JL. A rationale and test for the number of factors in factor analysis. *Psychometrika* 1965;30;179-185.
- 26. O'Connor BP. SPSS and SAS programs for determining the number of components using parallel analysis and Velicer's MAP test. *Behav Res Methods Instrum Comput.*2000;32:396-402.
- 27. Beauchaine TP, McNulty T. Comorbidities and continuities as ontogenic processes: Towards a developmental spectrum model of externalizing psychopathology. *Devel Psychopathol.* 2013;25:1505-1528.
- 28. Gillberg C. The ESSENCE in child psychiatry: Early Symptomatic Syndromes Eliciting Neurodevelopmental Clinical Examinations. *Res Devel Disabil.* 2010;31:1543-51.
- 29. Uljarević M, Evans DW, Alvares GA, Whitehouse AOJ. Relationship Between Restricted and Repetitive Behaviours in Children with Autism Spectrum Disorder and their Parents. *Molecular Autism* 2016;7:29.

- US Census Bureau. (2010; 2014). State and county Quickfacts, N.Y. Retrieved May, 2016, from <u>http://quickfacts.census.gov</u>.
- 31. Merikangas KR, He J-p, Burstein M, et al. Lifetime Prevalence of Mental Disorders in US Adolescents: Results from the National Comorbidity Survey Replication, Adolescent Supplement (NCS-A). *J Am Acad Child Adolesc Psychiatry*. 2010;49:980–9.
- 32. Kessler RC, Berglund PA, Demler O, Jin R, Walters EE. Lifetime prevalence and age-ofonset distributions of DSM-IV disorders in the National Comorbidity Survey Replication (NCS-R). Arch Gen Psychiatry. 2005;62(6):593-602.
- 33. Boyle CA, Boulet S, Schieve L, et al. Trends in the Prevalence of Developmental Disabilities in US Children, 1997–2008. *Pediatrics*. 2011;127:1034-42.

 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-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

	ACCEPT	TED MANUSCRIPT
\$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 or more	2.1	9.5

Table 2. Psychometric Properties

	ARI	ARI	ARI	CRI-R	CRI-R	CRI-R
	MBC	RIS	Total	MBC	RIS	Total
Internal Consistency (Cronbach's α)	.94	.94	.96	.95	.96	.97
Test-retest Reliability ICC	N= 31	8		N= 231		
(4 weeks)	.88	.90	.89	.88	.87	.88
Convergent Validity	N= 41	8		N=418		
(r; all p values < .001)						
SRS-2 Total	.59	.46	.57	.66	.59	.66
SRS-2 Emotion Recognition	.39	.26	.36	.43	.41	.44
SRS-2 Social Avoidance	.54	.43	.53	.56	.48	.54
SRS-2 Interpersonal Relatedness	.54	.40	.51	.59	.51	.57
SRS-2 Insistence on Sameness	.61	.50	.61	.68	.64	.69
SRS-2 Repetitive Mannerisms	.62	.44.	.58	.66	.51	.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 = Social Responsiveness Scale.

			CRI-R	
		CRI-R Total	RSMBC	CRI-R RIS
		Mean (SD)	Mean (SD)	Mean (SD)
Cluster diagnoses	Ν			
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)

Table 3. Childhood Routines Inventory-Revised (CRI-R) Scores Across Cluster Diagnoses

Note: ADD/ADHD = attention deficit disorder/attention-deficit/hyperactivity 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.

Table 4. Adult Routines Inventory (ARI) Scores Across Cluster Diagnoses

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

Note: ADD/ADHD = attention deficit disorder/attention-deficit/hyperactivity disorder; ASD OCD = obsessive-compulsive disorder; RIS = = autism spectrum disorder; **Rigidity/Insistence** Sameness; Repetitive on RSMBC Sensory Motor = Behaviors/Compulsions.

Figure 1. Childhood Routines Inventory–Revised (CRI-R) reliability across the full score range. Note: RSM = Repetitive Sensory Motor.

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

Figure 3. Child Childhood Routines Inventory (CRI) probability density distributions for

different parental diagnostic groups. Note: ASD = autism spectrum disorder; CRI-R =

Childhood Routines Inventory-Revised; NDD = neurodevelopmental disorder; NPD =

neuropsychiatric disorder; OCD = obsessive-compulsive disorder; RMBC = Repetitive Motor

Behaviors/Compulsions; RIS = Rigidity/Insistence on Sameness.







