Factor Analysis of Restricted and Repetitive Behaviors in Autism Using the Autism Diagnostic Interview-R

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ABSTRACT: The current study examined the factor structure of restricted and repetitive behaviors (RRB) in children with autism. Factor extraction procedures of 12 items from the Autism Diagnostic Interview-Revised (ADI-R) were applied in N = 207 individuals with autism. Two interpretable factors were identified: Factor 1—*repetitive sensory motor actions* and Factor 2—*resistance to change*. There was a significant negative correlation between an index of level of adaptive functioning and Factor 1. Intraclass correlations were not significant for either factor in a subset of families with two or more siblings with autism (multiplex). No differences in scores were apparent for either factor when multiplex families and families containing only one affected individual with autism (singleton) were compared. RRB in autism are represented by two distinct factors which may reflect two separate groups within autism. Defining subgroups within au-

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tism will allow for reduction of clinical heterogeneity and enhance our ability to dissect the genetic etiology of this complex disorder

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Autism is a complex neurodevelopmental disorder characterized by disturbances in social relations, impaired communicative abilities, and the presence of repetitive behaviors or stereotyped interests.[1] A formal diagnosis of autism requires the presence of difficulties in each of these three areas. However, restricted, repetitive and stereotyped patterns of behavior, interests, and activities (RRB) are not considered central to autism. Not surprisingly, there has been limited study of RRB in autism. Several reasons underlie the limited attention to this category. First, RRB are not specific to autism and occur in a number of developmental, neurologic, and psychiatric disorders.[2] Second, there is substantial variability in RRB among individuals. There are limited data to indicate the stability of RRB in individuals with autism over time. Third, measurement of RRB lacks a well-developed, validated methodology, which is surprising since RRB appear more quantifiable or measurable than social and language functions. Finally, there are few studies which provide normative data with respect to the development of restricted and repetitive behaviors in typically developing children.[3]

Bodfish and colleagues suggest that different forms of repetition are not well defined and lack behavioral referents.[2,4] For instance, many standardized measures of RRB are hampered by item overlap (i.e., the measures contain multiple items which reference the same behaviors). When overlapping items were removed and rating categories were behaviorally defined, Bodfish and colleagues were better able to identify independent RRB in individuals with developmental disabilities.[5] Bodfish and colleagues suggest that application of similar methods to autism will expand understanding of RRB with implications for classification and intervention and, in fact, did apply similar methods to autism.[6] In this study, they examined specific RRB in adults with autism and mental retardation relative to a group of individuals diagnosed with mental retardation alone and matched for age, gender, and IQ. Bodfish et al.[6] found that the autism group exhibited a significantly higher number of stereotypies, compulsions, and more severe compulsions compared with the mental retardation group. In addition, there was a positive relationship between the number of stereotypies and compulsions with severity of autism. These findings represent one of the few efforts to understand repetitive behaviors in autism using

a standardized methodology. However, generalizability to the wider range of individuals with autism is unclear as the participants in this study were limited to only autistic adults with severe and profound mental retardation.

Turner[7] also reviewed RRB in autism and noted that motor behaviors—stereotyped movements in particular—are the most widely studied form of RRB in autism. According to Turner, stereotyped movements are generally believed to index low cognitive ability. In contrast, "higher level repetitive behaviors" such as circumscribed interests are more commonly observed in higher functioning individuals. Turner's review suggests that RRB are not unidimensional and may serve as an index of ability or severity of autism.

Efforts to delineate the role of RRB in autism are important for two reasons: 1) to further studies of the genetic and neurobiologic bases of autism and 2) to help establish subtypes useful for clinical and research purposes. The relationship between RRB and neurobiological underpinnings has been examined in several other disorders. For instance, studies of brain-behavior relationships in Tourette's Disorders have implicated the basal ganglia.[8] In a study of individuals with autism, Sears and colleagues[9] examined the relationship between structural abnormalities based on MRI findings and RRB in 35 individuals with autism. Basal ganglia involvement—in particular the caudate nuclei—was evident and correlated with ADI-R items (compulsions/rituals, difficulties with minor changes, and complex motor mannerisms). This study was the first to identify a clear relationship between a neuroanatomic abnormality and RRB in autism. This study highlights the role of neurobiological correlates in RRB with implications for disorders where RRB are present such as autism.

Repetitive behaviors may also help to define subtypes for autism, a disorder that encompasses a wide range of clinical manifestations. Tanguay and colleagues[10,11] developed social communicative subtypes derived from behavioral phenomena common to autism. In these studies, Tanguay and colleagues identified items relevant to social-communication skills from the ADI-R and ADOS-G. Using samples of individuals with autism spectrum disorders, Tanguay et al. reported consistent social-communication factor-derived scores from both instruments. While a broader spectrum of individuals with pervasive developmental disorders was included in the studies, the convergence of the two instruments suggests that both parent-reported and clinician observed sources of information are valid methods for construction of subtypes in autism. The construction of empirically derived

RRB factors in autism follows directly from the work of Tanguay. Further, there appears to be a wide range of variability in RRB among individuals with autism that may allow for derivation of distinct groupings of items (or factors). These factors can be used to index behavioral heterogeneity of individuals with autism and allow for potential subgroups or subtypes.

The specific objectives of this study were:

1. To identify restricted and repetitive behaviors factors based on items from the ADI-R.
2. To examine the concordance for the derived factors in affected siblings from multiplex families.
3. To determine the relationship between identified factors and developmental level.

Methods

Participants

The participants in the study consist of 292 individuals with autism who were drawn from a larger ongoing study of the genetics of autism. Study participants ranged in age from 3 to 21 years at time of ADI-R. The participants were from both singleton (only 1 affected individual/family with no other family history of autism) and multiplex (> 1 affected individual/family) families. We performed the factor analysis and assessment of the correlation between factor groupings and developmental level using the probands (N = 207) from the singleton and multiplex families. This strategy was adopted to ensure the use of independent observations (i.e., we used only one individual from the MP families in the factor extraction procedures). Sixty-four of the individuals in these analyses came from families containing two or more affected individuals; these individuals plus their affected siblings were examined for concordance (N = 120 individuals from 60 families with two affected individuals and 12 individuals from 4 families with three affected individuals). The participants are described in Table 1.

Measures

Diagnostic and phenotype assessments were conducted on all study participants. Confirmation of autism diagnosis was completed using the Autism Diagnostic Interview-R (ADI-R).[12] Clinical assessment of adaptive and behavior functioning was completed using the Vineland Adaptive Behavior Scale-Survey edition[13] and the Aberrant Behavior Checklist-Community Version.[14] These are well-established multidimensional instruments that permit the assessment of various domains of functioning.

Table 1
Descriptive Statistics

Characteristic	*N*	*%*	*Mean (Std. Dev.)*	*Range*
Sex				
Male	153	73.9	—	—
Female	54	26.1		
Race/Ethnicity				
Caucasian	166	80.6	—	—
African American	27	13.1		
Hispanic	8	3.9		
Asian	4	1.9		
American Indian	1	0.5		
Missing	1			
Age at ADI exam (months)	207	—	108.7 (54.8)	29–254
Age (months) of first appearance of developmental abnormalities				
As reported by parents ADI-R q2)	207*	—	17.1 (10.6)	0.5–72
As estimated by interviewer (ADI-R q94)	205	16.1 (7.3)	1.0–42	

*Values of "0" months and "since birth" were converted to 0.5. For ADI-R q2, parent report of *can't recall but before three years* was converted to 18 months.

Autism Diagnostic Interview-Revised: The ADI-R[12] was used to confirm the clinical diagnosis of Autistic Disorder based on DSMIV/ICD-10 criteria. The ADI-R is a standardized semi-structured interview that consists of a hierarchical series of probes and questions that allow caretakers to provide detailed descriptions of the individual's behaviors. Select groups of items are summed into area scores that form the basis for the diagnostic algorithm. The classification of autism requires that an individual exceed cutoff scores in each of the three areas: social, communication (nonverbal or verbal), and restricted, repetitive behaviors.[15] All individuals met research diagnosis for autism. Individual items are scored for two time points: current and past (if the behavior ever occurred or if it occurred between the ages of 4 to 5 years).

Vineland Adaptive Behavior Scales (VABS): The VABS[13] is a measure of adaptive behavior based on an extended interview with an informant familiar with the individual's day-to-day functioning. The VABS assesses adaptive behavior in domains of communication (receptive, expressive, and written), daily living skills (personal, domestic, and community), and socialization (interpersonal relations, play and leisure time, and coping skills). Scores from domains and subdomains permit the comparison of specific profiles of adaptive behaviors in these groups. We used a composite score, the Adaptive Be-

havior Composite, which is considered the most reliable index of adaptive functioning in our analyses. The VABS serves as a general measure of developmental functioning with an emphasis on adaptive behavior. The VABS has been used in this capacity in a number of studies as a means of indexing level of functioning.[16] However, it is important to recognize that the VABS is not used to index cognitive functioning. Individuals with VABS Adaptive Behavior Composite age equivalents ≤ 18 months were excluded from the analysis. This cutoff is based on evidence that the ADI-R is less reliable in discrimination of autism from nonautism in individuals with developmental ages ≤ 18 months.

Procedure

All study subjects were ascertained for participation in a genetic study of autism. Caregivers were administered the ADI-R, and VABS, without modifications. We did not directly assess cognitive functioning in our participants but abstracted available results from medical records. These data are not reported here due to the variable measures used and the broad range of intervals between the ADI-R administration and test dates for many of the participants. Parents of all participants supplied informed consent. Participant assent was solicited. All participating families were also administered a standardized family history questionnaire that queries the family with respect to the diagnosis of or symptoms of autism in siblings, cousins and avuncular relationships. Verification of diagnosis was obtained through clinical examination of participating individuals as outlined below and/or through medical records.

Data Analysis

The ADI-R consists of 111 items. Twelve items were chosen for analysis on the basis of clinical relevance and grouping within the ADI-R (see Table 2). These items were chosen from a larger group of questions that are contained within the Restricted and Repetitive Behaviors or Interests section of the ADI-R. We eliminated items that were administered only to individuals within certain age groups. Note that while the items from the Restricted and Repetitive Behaviors or Interests section of the ADI-R items are believed to represent a homogenous grouping, no factor extraction procedures have been applied. Most items are scored from 0 to 3. Note that in the diagnostic algorithm, scores of 3 are recoded as 2 to limit the impact of severity in the overall diagnostic process. The full range of item scores (0–3) was used to provide maximal information[17] and to allow severity to exert its effect. The few variables with codes > 3 were recoded to fit the 0–3 scale.

All data analyses were conducted using SAS.[18] The twelve items were initially analyzed among the proband data set (N = 207) using principal component analysis (PCA) in order to identify potential factors. We subsequently performed factor analysis (FA) to test our hypothesis that the items had shared or common elements. We calculated standardized Cronbach's alphas to measure the reliability of each identified factor using the ALPHA option in PROC CORR in SAS. Both PCA and FA were run using promax rotation. A two-factor solution was selected on the basis of interpretability. To investi-

Table 2
ADI-R Items Selected for Use in Factor Extraction

Variables	*N*	*Mean (Std. Dev.)*	*Range*
Hand and finger mannerisms	207	0.93 (0.88)	0–3
Unusual sensory interests	207	1.02 (0.65)	0–2
Repetitive use of objects or parts of objects	207	0.97 (0.95)	0–3
Other complex mannerisms or stereotyped body movements	207	0.79 (0.89)	0–3
Rocking	207	0.35 (0.68)	0–3
Unusual preoccupations	207	0.61 (0.85)	0–3
Unusual attachment to objects	207	0.40 (0.72)	0–3
Abnormal idiosyncratic negative responses to specific sensory stimuli	207	0.78 (0.82)	0–3
Undue general sensitivity to noise	207	1.21 (0.93)	0–3
Difficulties with minor changes in routine or personal environment	207	1.04 (0.89)	0–3
Resistance to trivial changes in the environment	207	0.40 (0.69)	0–3
Compulsions/rituals	207	0.89 (0.95)	0–3

gate the role of developmental level as measured by the VABS as a mediating variable in RRB we tested the same data set for correlation between level of functioning (VABS Adaptive Behavior Composite) and each identified factor. We calculated intraclass correlations (ICC)[19] using affected siblings from the multiplex families (N = 132 individuals from 64 families) to determine if familial clustering existed for each of the identified factors. The majority of our affected siblings consisted of sibpairs (N = 120 from 60 families). However, four families contained three or more affected individuals. The probands from the previous analyses were included in the ICC analyses when they had affected siblings with complete data. Finally, to determine whether different processes may be at work in sporadic versus familial autism in our dataset, we tested for differences in each factor score between the singleton and multiplex families using the Wilcoxon rank sum test (2-sided). Both groups were restricted to probands to ensure independence of observations.

Results

Initial data analyses were conducted on a sample of 207 probands. Descriptive statistics for the individual items are presented in Table 2.

Principal component analysis was run initially to estimate/confirm the presence of at least two factors as hypothesized. Five eigenvalues

exceeded 1 and accounted for 60.7% of the standardized variance. However, there were minimal differences between factors 2 and subsequent factors and only the first two factors were retained. These components accounted for 32% of the variance. The failure to account for a larger proportion of the variance suggests that RRB may represent independent phenomena with only small clusterings of items. A promax rotation yielded adequate loadings (≥0.40) on the two factors. The first factor consisted of 6 items with loadings ranging from 0.40 to 0.70; factor two (3 items) loadings ranged from 0.61 to 0.79. Given that we hypothesized underlying factor structures (i.e., clinically meaningful groups of variables), a principal factor analysis (promax rotation) was conducted. The correlation matrix for the 12 items is presented in Table 3.

A scree plot confirmed the presence of at least two factors with a gradual diminution of slope after factor 2. We used a cutoff of (≥0.30) for inclusion of an item on a respective factor. Again, given that the variance accounted for by these two factors was low, it was decided to lower the threshold for factor loadings. The factor loadings for the two factors are presented in Table 4. Five items loaded on factor 1 (which we labeled *repetitive motor sensory actions*) while three items loaded on factor 2 (*resistance to change*). Note that the two factors were not correlated with each other with a Pearson correlation coefficient of 0.08 ($df = 205$, $p = 0.25$). The standardized Cronbach's alphas for factors 1 and 2, respectively, were 0.62 and 0.52, respectively. The low score for factor 2 is not surprising given that the factor consists of only 3 items. Typically, scales with few items are more likely to have lower coefficients.[20]

For our second objective we examined the concordance between affected siblings in the multiplex families. The ICCs for Factors 1 and 2, respectively, were 0.12 and 0.26. These represent weak relationships. No significant differences in factor scores were found between the multiplex and singleton groups for either factor 1 ($p = 0.76$) or factor 2 ($p = 0.27$). Finally, for our third objective, we checked for concordance between the respective factors with respect to adaptive-developmental level. The Adaptive Behavior Composite (ABC) from the Vineland Adaptive Behavior Scale was used as an index of developmental level. The Pearson correlation coefficient was –0.27 ($df = 128$, $p = 0.002$) for the correlation between Factor 1 and the ABC while Factor 2 showed a much weaker relationship ($r = 0.007$) with the ABC ($df = 128$, $p = 0.94$).

Table 3
Principal Components Analysis Correlation Matrix (Promax Rotation)

Variable	*noise*	*preocc*	*rep_obj*	*per_rout*	*environ*	*comp_rit*	*att2obj*	*sens_int*	*id_resp*	*fing_man*	*comp_man*	*rocking*
Undue general sensitivity to noise	1.000	0.037	0.062	–0.069	0.117	0.009	0.140	0.131	0.061	0.137	0.054	–0.002
Unusual preoccupations	—	1.000	0.249	–0.006	–0.016	0.033	0.285	0.031	0.039	0.166	0.194	0.049
Repetitive use of objects or parts of objects	—	—	1.000	–0.010	0.025	0.013	0.158	0.395	0.109	0.293	0.232	0.173
Difficulties with minor changes in routine or personal environment	—	—	—	1.000	0.354	0.279	0.066	–0.052	0.058	–0.071	0.144	0.058
Resistance to trivial changes in the environment	—	—	—	—	1.000	0.154	0.163	0.081	0.112	0.125	0.089	0.033
Compulsions/rituals	—	—	—	—	—	1.000	0.006	0.247	0.119	0.084	0.048	0.095
Unusual attachment to objects	—	—	—	—	—	—	1.000	–0.017	–0.089	0.165	0.092	0.051
Unusual sensory interests	—	—	—	—	—	—	—	1.000	0.182	0.336	0.209	0.151
Abnormal idiosyncratic negative responses to specific sensory stimuli	—	—	—	—	—	—	—	—	1.000	0.107	0.097	–0.012
Hand and finger mannerisms	—	—	—	—	—	—	—	—	—	1.000	0.261	0.292
Other complex mannerisms or stereotyped body movements	—	—	—	—	—	—	—	—	—	—	1.000	0.112
Rocking	—	—	—	—	—	—	—	—	—	—	—	1.000

Table 4
Factor Analysis of Restricted Behaviors and Repetitive Interests
(Maximum-likelihood with promax rotation standardized regression coefficients)

Variables	*Factor 1: Repetitive Sensory Motor Actions*	*Factor 2: Resistance to Change*
Hand and finger mannerisms	0.615	0.012
Unusual sensory interests	0.576	0.026
Repetitive use of objects or parts of objects	0.562	0.066
Other complex mannerisms or stereotyped body movements	0.395	0.199
Rocking	0.315	0.101
Unusual preoccupations	0.286	0.033
Unusual attachment to objects	0.214	0.096
Abnormal idiosyncratic negative responses to specific sensory stimuli	0.203	0.086
Undue general sensitivity to noise	0.200	–0.042
Difficulties with minor changes in routine or personal environment	–0.080	0.992
Resistance to trivial changes in the environment	0.143	0.375
Compulsions/rituals	0.173	0.303

Factor loadings of those items which exceed .30 are underlined.

Discussion

RRBs are required for a diagnosis of autism. However, unlike social and communication behaviors, there has been limited study of repetitive phenomena. This study examined the factor structure of RRB in a sample of individuals with autism. Principal factor analysis of 12 items from the ADI-R revealed two factors that were labeled *repetitive sensory-motor behaviors* and *resistance to change*. Identification of these two factors demonstrates that the ADI-R is a suitable assessment strategy to document repetitive behaviors and suggests that parent report of these behaviors is accurate. Many of the RRB are typically dramatic and easily observed which may also account for the accuracy of parental responses. Note that while the ADI-R provides information about the presence of such behaviors it is limited in capturing information about more specific aspects of RRB such as frequency or intensity.

The two factors represent different phenomena in autism. Factor 1,

repetitive sensory-motor behaviors, consists of behaviors that are unified along the dimension of motor and sensory behaviors that have no apparent purpose apart from self-stimulation. These behaviors occur in many different developmental disabilities and most likely covary with developmental level. Their relationship with level of autism is not clear. This factor is similar to that described as lower order repetitive phenomena. Factor 2, *resistance to change*, consists of behaviors that reflect insistence on sameness. The individual is imposing an order (or responding to a lack of order) in the environment. These behaviors represent a more clinically coherent group of items and do not necessarily occur among individuals with varied developmental disorders (i.e., they may be specific to autism).

These two factors may serve to index level of functioning. Both Turner[7] and Bodfish et al.[6] propose that there are differences in the quality and nature of RRB in relation to developmental level. Individuals with lower cognitive scores display increased repetitive motor and sensory behaviors while individuals with higher cognitive abilities are more likely to engage in complex actions or show more intense interests. Our results support this bi-dimensional conceptualization when using the VABS as an index of level of functioning. Factor 1 (repetitive sensory motor actions) was significantly negatively correlated with level of functioning as indexed by the Adaptive Behavior Composite. As predicted, with increases in RRB there is a corresponding decrease in developmental functioning. Factor 2 (resistance to change) did not correlate with the ABC. These data support the recent work of Liss and colleagues[21] in which different relationships between autism symptoms and adaptive behaviors were noted as a function of developmental level using the VABS. The extent to which the RRB factors would correlate with cognitive-developmental levels cannot be determined. Also, another consideration is found in the work of Evans and Gray[22] who found that mental age is a critical determinant of the presence of compulsions. Children with developmental problems demonstrate similar patterns of behavior as individuals without developmental problems who are matched for developmental age. A clear pattern of reduced compulsive behavior emerges over time and with changes in development. An important distinction lies in the extent to which developmental level is related to repetitive behaviors in individuals without mental retardation.

While differentiation of these two factors is irrelevant for clinical diagnosis, the identification of distinct RRB factors may help research studies identify the genetic neurobiological components of autism. Au-

tism is a very heterogeneous disorder characterized by a wide range of clinical manifestations. Earlier studies of RRB in autism conceptualized RRB as a broad group of behaviors that represented a single class. However, the identification of distinct factors within RRB demonstrates that there are potential subgroups that in turn may reflect different etiologic mechanisms. These findings are consistent with the work of Berkson and colleagues[23] who argue that repetitive behaviors should not be grouped together in a single class. Differentiation of etiologies may be especially important in efforts to index response to treatments.

Repetition in some form is a part of autism. There is a strong likelihood that the form and extent of the repetition are tied to developmental level. Increasing attention to the etiological mechanisms involved in RRB may have value for understanding the development of other behaviors in autism. Current theories suggest that there are relationships between the RRB seen in autism and neuroanatomic and neuroimmunologic findings. The variability in repetitive phenomena in people with autism may be significant for subgrouping autism in genetic studies. It would seem critical, however, that subsequent studies quantify the role of developmental disability apart from autism in relation to repetitive phenomena. One potential value for use of RRB factors is in genetic studies. The tremendous behavioral variation in clinical features in autism strongly suggests the possibility of subtypes. The addition of an RRB subtype or subset may be of value in segregating genetic analysis. Preliminary efforts[24] have found a potential relationship between Factor 2 and increased detection of linkage signal for chromosome 15. Clearly, increased efforts to understand the role of RRB in autism will be of great value in our attempts to dissect the genetic etiology of this complex disorder.

Limitations

The study of RRB relies solely on parent-report and does not distinguish between current and ever ratings. While these are available from the ADI-R, we chose to use ever ratings as there were no statistically significant differences between current and historical ratings in our sample. Actual observation and recording of RRB in the participants would provide an additional point of reference for understanding RRB. However, many RRB are low rate behaviors and would be difficult to capture without extended observational periods over time.

Another limitation is the lack of information about RRB in individuals with other developmental disorders—in particular mental retardation. The current study was restricted to individuals with autism and the ADI-R is predominantly used for the purpose of classifying individuals with this disorder. Certainly, this study is exploratory regarding the nature of RRB in autism. However, future studies should focus on investigation of RRB in autism in contrast to individuals with other developmental disorders. This would allow for identification of behaviors or factors that are more explicitly associated with developmental disability as well as those behaviors or factors that characterize autism. A third limitation of this study is the use of the VABS only as an index of developmental level. Given the lack of information regarding intervention history and the amenability of adaptive functioning to intervention, this introduces a source of error that was not controlled.

Summary

Autism is a complex neurodevelopmental disorder that is characterized by disturbances in social relations, impaired communicative abilities, and the presence of repetitive behaviors or stereotyped interests. Although restricted, repetitive and stereotyped patterns of behavior, interests, and activities (RRB) are necessary for diagnosis they are not considered central to the disorder. Most efforts to clarify behavior variation in autism have emphasized social communicative dimensions, leaving RRB both understudied and poorly understood. The current study identified two RRB factors in a sample of individuals with autism (N = 207) using select items from the Autism Diagnostic Interview-Revised (ADI-R). Factor extraction methods (promax) of 12 items from the ADI-R yielded two interpretable and distinct factors: Factor 1—*repetitive sensory motor actions* and Factor 2—*insistence on sameness.* Correlational analysis of the relationship between level of functioning and Factors 1 and 2 found a significant negative correlation between our index of developmental level and Factor 1. This is consistent with our hypothesis that repetitive sensory and motor actions are an index of developmental disability. We also assessed factors 1 and 2 in a subset of families who had two or more siblings with autism (multiplex) within our larger sample of participating families. The sibling pairs did not demonstrate meaningful similarities within families. No differences were apparent when the multiplex and families containing only one affected individual with autism (singleton) were

compared on either factor. The findings have significant implications for efforts to dissect the genetic etiology of autism. The use of empirically derived RRB factors represents one strategy to reduce the effects of behavioral heterogeneity in neurobiological and genetic studies of autism.

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