

A Principal Components Analysis of the Autism Diagnostic Interview-Revised

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ABSTRACT

Objective: To develop factors based on the Autism Diagnostic Interview-Revised (ADI-R) that index separate components of the autism phenotype that are genetically relevant and validated against standard measures of the constructs. **Method:** ADIs and ADI-Rs of 292 individuals with autism were subjected to a principal components analysis using VARCLUS. The resulting variable clusters were validated against standard measures. **Results:** Six clusters of variables emerged: spoken language, social intent, compulsions, developmental milestones, savant skills and sensory aversions. Five of the factors were significantly correlated with the validating measures and had good internal consistency, face validity, and discriminant and construct validity. Most intraclass correlations between siblings were adequate for use in genetic studies. **Conclusion:** The ADI-R contains correlated clusters of variables that are valid, genetically relevant, and that can be used in a variety of studies. *J. Am. Acad. Child Adolesc. Psychiatry*, 2003, 42(7):864–872. **Key Words:** autism, Autism Diagnostic Interview-Revised, principal components analysis.

The Autism Diagnostic Interview-Revised (ADI-R; Lord et al., 1994), along with its companion Autism Diagnostic Observation Schedule-Generic (ADOS-G; Lord et al., 1989), is the primary research instrument used for diagnosing autism. The ADI-R is a semistructured, standardized interview, conducted with a caregiver, that assesses the presence and severity of various behaviors commonly found in autism. The interview contains over 100 items that solicit information about a child's language, communication, social development, play, unusual behaviors and interests, and developmental milestones. A key feature of the ADI-R is the diagnostic algorithm, which includes a subset of the items. In order to meet the ADI-R diagnostic criteria, it is necessary to score above a spec-

ified threshold on each of the four algorithm domains: communication, social interaction, repetitive behaviors, and age at onset of some symptom. The ADI-R items chosen for inclusion in the domains are those thought by the developers to best exemplify *DSM-IV* and *ICD-10* criteria for autism and to best discriminate clinically diagnosed cases of autism from cases with abnormal cognitive development without autism. The algorithm domains were not constructed on the basis of any psychometric analyses and were intended to make a categorical diagnosis, rather than to be used as measures of severity.

Several studies have explored the factor structure of some ADI-R items. Tanguay et al. (1998) factor analyzed 28 items they judged to relate to social communication and identified three factors within this group. The scores for all three factors were well correlated with the social domain score on the ADI-R algorithm, but less with the communication domain and even less with the repetitive behaviors domain. They pointed out that the communication domain contained a mixture of items, some relating to pragmatics (social communication) and others to structural language (vocabulary, grammar, and syntax). Lord (1990) performed a principal components analysis on 32 items that exemplified the then-new *ICD-10* diagnostic criteria. The two strongest factors were both mixtures of items assigned to either the social or communication domains. She concluded that social and

Accepted December 27, 2002.

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This work was supported by grant MH55135 to Dr. Folstein and PO1DC03610 to Dr. Tager-Flusberg.

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DOI: 10.1097/01.CHL.0000046870.56865.90

communicative behaviors might represent a single concept that could be more meaningfully divided by context. Silverman et al. (2002) used ADI-R algorithm domains to assess their familiarity. They showed, in a large sample of multiplex families, that some domains and subdomains were familial. Parts of the communication and repetitive behavior domains, but not the social domain, were more similar within sibships than expected.

We report here a principal components analysis that includes most of the ADI-R items, regardless of their inclusion in the diagnostic algorithm. We hoped to develop a set of factors that indexed separate components of the autism phenotype that were genetically relevant, could be validated against standard measures of the constructs, and could be used as approximations to continuously distributed measures for these components. Such ADI-R-based scales would have uses in a variety of studies.

METHOD

Sample

The principal components analysis used ADIs and ADI-Rs of 292 individuals with autism. Ninety ADIs were from the Baltimore Family Study (Piven et al., 1991, 1994), 107 ADI-Rs were from the New England families of the Collaborative Linkage Study of Autism (CLSA)

(e.g., Barrett et al., 1999; CLSA, 2001; Nurmi et al., 2001; and others), and 95 ADI-Rs were purchased from the Autism Genetic Research Exchange (AGRE) database. All cases had a clinical diagnosis of an autism spectrum condition, which was confirmed by the ADI/ADI-R for the Baltimore and AGRE samples and by ADI-R and ADOS-G for the CLSA sample. Reliability checks between examiners were conducted for the three samples. The interviewers at the three sites were trained by certified ADI trainers. For the multiplex families, one proband was chosen at random for inclusion in the analysis in order to ensure unbiased estimates of the coefficients of the correlation matrix. For the validation of the factors, we analyzed an independent sample of 68 children and adolescents with autism who had participated in a longitudinal study of language in autism and who were diagnosed with the ADI-R, ADOS-G, and a clinical evaluation. Most subjects in the validation sample had some useful language. Table 1 shows demographics of the samples.

Methods for Principal Components Analysis

We developed a data set composed of the 98 items that were either identical or closely comparable in both the ADI and the ADI-R. Items that were not common to both versions were omitted. We recoded several language-related items, replacing a score of 8 (item not applicable because of insufficient language) with a 3, the most severe rating. We replaced missing data by the mean score of the variable across the sample so that missing values would not contribute to the principal components. For items that had both “ever” and “current” ratings, both were entered into the analysis to compensate for missing items and to provide some redundancy necessary for scale construction. Inclusion of both types of items also provided a means of differentiating cases that did and did not improve over time. When the age 4 to 5 ever and current scores were run separately, no meaningful factors emerged.

TABLE 1
Demographic Characteristics of the Samples

Sample (N)	Age, yr Mean (SD)	Age Range	Sex Ratio (Male:Female)	Ethnicity	IQ Groups	n
Baltimore (90) ^a	15.60 (7.90)	5–37	68:22	91% White; 6% Hispanic, African American; Asian; 3% other	<30 30–49 50–69 >69	18 24 15 33
AGRE (95) ^b	7.44 (4.39)	4–38	66:19	80% White; 5.26% Asian; 3.16% Hispanic; 3.16% other; 8.42% unknown	<30 35–49 50–69 70–89 >89	0 6 4 7 18
CLSA (107) ^c	18.50 (4.95)	2–47	86:21	92.51% White; 4.67% African American; 1.87% Asian; 0.93% Hispanic		
Tager-Flusberg’s longitudinal language study (68) ^d	6.75 (2.29)	4–13	59:9	94.12% White; 5.88% Hispanic, African American, Asian	<30 30–49 50–69 70–89	0 3 12 23

^a Nonverbal IQ based on tests from medical records.

^b IQ defined as the performance score on Raven Progressive Matrices test, available for 35 participants, 7 unable to test, and 3 unable to score. AGRE = Autism Genetic Research Exchange.

^c No IQ data are available for the Collaborative Linkage Study of Autism (CLSA) sample.

^d Nonverbal IQ derived from Differential Abilities Scales, available for 64 participants.

The 98 variables common to the ADI and ADI-R were entered into VARCLUS (SAS, 2000). The VARCLUS procedure utilizes an oblique principal component analysis (Harman, 1976). For any group of variables, the principal components are the “directions” (each given by some linear combination of the variables) in which most of the variation of the data is explained. The first principal component is the one that accounts for more variation than any other linear combination of the items. VARCLUS attempts to split the variables into clusters (or factors) in such a way as to maximize the amount of variation in the data explained by the totality of all the first principal components. Because the variance explained by the components is maximized, variables loaded onto a cluster will tend to be correlated, whereas variables in distinct clusters will tend to be uncorrelated. The optimal number of clusters was determined to be six. With fewer than six clusters, there remained many variables that did not correlate well with the rest of their group; with more than six, the procedure split up clusters of variables that were fairly well correlated with each other. Additional evidence that six was an appropriate number of factors is provided by examining the analogy of a scree plot for this method of clustering variables. There was a large drop-off in the amount of variation explained in going from two to three clusters, but only about 6.5% of the variance was explained by two clusters. The next relatively large drop-off occurred between six and seven clusters. Six clusters accounted for about 41% of the variation.

We next eliminated variables that correlated at less than $r = 0.40$ with the rest of the variables in its group. Thus, the factors were constructed to maximize item internal consistency. We then eliminated items that also correlated well with another factor to maximize item discriminant validity and thus the extent to which the factors represented independent constructs. After eliminating these variables, VARCLUS was rerun for the 62 remaining variables, and the resulting formulas for the first principal components defined our six scales (Table 2). On the basis of the items loading on the factors we labeled them Spoken Language, Social Intent, Compulsions, Milestones, Savant Skills, and Sensory Aversions. Eighteen of the 62 were algorithm items.

Factor Validation

The factors were validated with an independent sample of 68 autistic subjects. The tests and evaluations used in the factor validation included measures of vocabulary, adaptive behavior, intelligence, executive functioning, and psychiatric diagnosis.

We hypothesized that factor I, Spoken Language, would be correlated with the Expressive Vocabulary Test (EVT; Williams, 1997 #992) and the Peabody Picture Vocabulary Test-Third Edition (PPVT-III; Dunn and Dunn, 1997) combined score. The two tests were standardized on the same normative sample and can thus be averaged to yield an overall measure of vocabulary ability. To validate factor II, Social Intent, we used the standard score of the socialization domain from the Vineland Adaptive Behavior Scales (Sparrow et al., 1991). Factor III, Compulsions, included a subset of the ADI items that code for repetitive behaviors that can strongly resemble compulsions. Psychiatric diagnoses for the subjects in the validation sample had been made with the Schedule for Affective Disorders and Schizophrenia for Developmentally Disabled Children, Adolescents, and Adults-Present and Lifetime Version (K-SADS-DD-PL) (Tadevosyan-Leyfer et al., unpublished), a semistructured diagnostic interview, administered to a caregiver. It is a modification of the Schedule for Affective Disorders and Schizophrenia for School-Age Children-Present and Lifetime Version (K-SADS-PL) (Kaufman et al., 1997) and is designed to assess psychiatric disorders in children and adolescents with developmental

disabilities. The K-SADS was administered to the parents at least 1 year after the ADI-R. From the Obsessive-Compulsive Disorder section, we derived a compulsion score by combining the score for “time consumed by specific compulsions” with the score for “impairment from the specific compulsion.” Factor IV, Milestones, was validated by using the sum of the standard scores from the communication, daily living, and socialization domains of the Vineland Adaptive Behavior Scales, the “total” Vineland score. We also validated this factor with an intelligence test, the Differential Abilities Scales (DAS), which has both Verbal and Performance subscales, and the PPVT/EVT average score. On the basis of work by Treffert (1989) on savants, we hypothesized that factor V, Savant Skills, would be correlated with a measure of working memory. Two working memory tasks were available, the Backward Word Span task, similar to a task used by Russell et al. (1996), and the Backward Block Span task (Isaacs and Vargha-Khadem, 1989). In the Backward Word Span task, participants are presented with a picture plate showing nine items. The experimenter reads sequences of items that increase in length, and the child must point to the items in the reverse order. In the Backward Block Span task, a child is asked to watch the experimenter, who points to an unstructured array of nine identical blocks that increase in length. The child’s task is to repeat this sequence in the reverse order by pointing to the blocks. Like the word span task, the test continues until the child gets two consecutive trials wrong at a given sequence length. To validate factor VI, Sensory Aversions, we used the score for the algorithm item D1 from the ADOS-G, unusual sensory interest in play material/person.

RESULTS

Factor Loading and Constructs

Factor I includes a variety of items pertaining to speech behaviors (Table 2). Four of the items in this factor are included in the Communication section in the ADI-R algorithm, but the factor did not include any aspects of language that could be construed as having social communicative intent. Thus we labeled it Spoken Language rather than Communication. Verbal Rituals loaded on this factor, rather than in the Compulsions factor, even though verbal rituals are often compulsive. Its R^2 with the compulsion factor was very low (0.0025). Factor II, Social Intent, includes 20 items, more than any other factor. It is a mixture of items from the Social and Communication sections of the ADI-R, and of the six algorithm items loaded here, three are from the Communication section and three are from the Social Impairment section. Hand and Finger Mannerisms loaded on this factor; in the ADI-R algorithm it is included in the Repetitive Behaviors section. Factor III, Compulsions, includes repetitive behaviors that often resemble compulsions, in that the child becomes upset when they are interrupted and seems driven to carry them out. It does not include any repetitive motor behaviors, which are not usually compulsive. Factor IV, Developmental Milestones, includes

TABLE 2
ADI Factors

	<i>R</i> ² w/Own Factor		<i>R</i> ² w/Own Factor
Factor 1: Spoken Language		Factor 2: Social Intent	
Complexity of nonechoed utterances—current	0.27	Conventional/instrumental gestures at 4–5 ^a	0.32
Overall level of language	0.74	Nodding at 4–5 ^a	0.22
Pronominal reversal—current	0.56	Head shaking at 4–5 ^a	0.21
Pronominal reversal—ever ^a	0.41	Attention to voice at 4–5	0.21
Neologisms/idiosyncratic language—current	0.74	Affection at 4–5	0.29
Neologisms/idiosyncratic language—ever ^a	0.62	Quality of social overtures at 4–5 ^a	0.38
Inappropriate questions or statements—current	0.70	Offers comfort at 4–5 ^a	0.39
Inappropriate questions or statements—ever ^a	0.66	Greeting at 4–5	0.33
Verbal ritual—current	0.69	Coming for comfort 4–5	0.40
Verbal rituals—ever ^a	0.62	Imitative social play 4–5 ^a	0.37
Reciprocal conversation—current ^a	0.57	Conventional/instrumental gestures—current	0.36
Intonation/volume/rhythm/rate—current	0.61	Coming for comfort—current	0.29
Vocal expression—current	0.52	Greeting—current	0.37
		Quality of social overtures—current	0.28
		Appropriateness of social response—current	0.23
		Range of facial expression used to communicate—current ^a	0.32
		Sharing others' pleasure and excitement—current	0.25
		Curiosity—current	0.28
		Hand and finger mannerisms—current	0.22
		Hand and finger mannerisms—ever ^a	0.26
Factor 3: Compulsions	<i>R</i> ² w/Own Factor	Factor 4: Developmental Milestones	<i>R</i> ² w/Own Factor
Stereotyped utterances—current	0.28	Walked unaided	0.29
Stereotyped utterances—ever ^a	0.28	Acquisition of bladder control: daytime	0.63
Unusual preoccupations—current	0.38	Acquisition of bowel control	0.76
Unusual preoccupations—ever ^a	0.46	Acquisition of bladder control: night	0.55
Compulsions/rituals—current	0.28	Age of first single words ^a	0.19
Compulsions/rituals—ever ^a	0.26	Age of first phrases ^a	0.24
Resistance to trivial changes in the environment—current	0.39	Sat unaided on flat surface	0.29
Resistance to trivial changes in the environment—ever	0.46		
Unusual attachment to objects—ever	0.15		
Factor 5: Savant Skills	<i>R</i> ² w/Own Factor	Factor 6: Sensory Aversions	<i>R</i> ² w/Own Factor
Visuospatial ability—current	0.39	Undue general sensitivity to noise—ever	0.19
Visuospatial ability—ever	0.44	Abnormal idiosyncratic negative response to specific sensory stimuli—current	0.70
Computational ability—current	0.26	Abnormal idiosyncratic negative response to specific sensory stimuli—ever	0.73
Computational ability—ever	0.24	Gait—current	0.45
Memory skill—current	0.50	Gait—ever	0.45
Memory skill—ever	0.50		
Musical ability—current	0.39		

^a Autism Diagnostic Interview-Revised (ADI-R) algorithm items.

a variety of milestones including bowel and bladder control, as well as motor and language skills. Factor V, Savant Skills, includes four of the five isolated skills we entered. The ADI-R includes six; the Reading Skills item was not entered because it is not a part of the original ADI, and

drawing ability did not load on this factor. Factor VI, Sensory Aversions, includes the general item from the ADI-R that codes for a variety of sensory sensitivities, as well as the specific item for coding sensitivity to noise. Abnormal Gait also loaded on this factor.

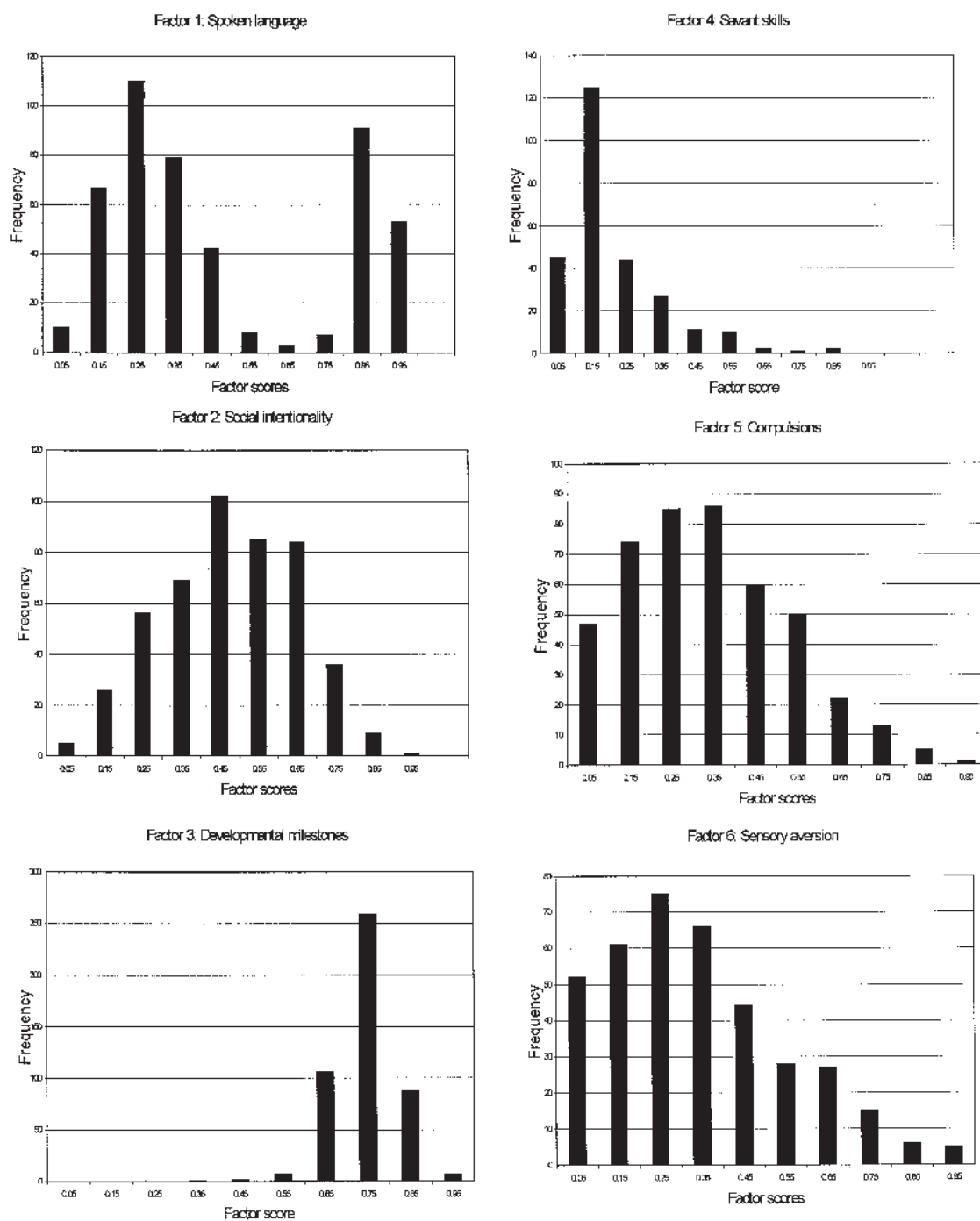


Fig. 1 Distribution scores of each factor.

Derivation of Factor Scores

The coefficients for items in each factor were very similar. Therefore, to simplify scoring, we summed the face value of each item when computing the raw factor score. Each factor score was then transformed to a proportion

of the total possible score for that factor, so that possible scores for each factor range from 0 to 1. This allows each factor to have the same range of scores (even though each factor contains different numbers of items) and provides a means for dealing with missing values without imput-

TABLE 3
Correlation of the Autism Diagnostic Interview Factors With Each Other, Two-Tailed Tests (*N* = 68)

	Spoken Language	Social Intent	Compulsions	Developmental Milestones	Savant Skills
Spoken Language	—				
Social Intent	$\rho = 0.45^{**}$	—			
Compulsions	$\rho = 0.33^{**}$	$\rho = 0.25^*$	—		
Dev. Milestones	$\rho = 0.17$	$\rho = 0.38^{**}$	$\rho = 0.07$	—	
Savant Skills	$\rho = 0.02$	$\rho = 0.04$	$\rho = 0.22$	$\rho = -0.15$	—
Sensory Aversions	$\rho = -0.01$	$\rho = 0.06$	$\rho = 0.21$	$\rho = 0.01$	$\rho = 0.09$

* $p < .05$; ** $p < .01$.

ing scores for missing variables. The distribution of scores for each factor in the sample used to construct the factors is shown in Figure 1.

Factor Intercorrelation

We constructed the factors so that items included in each factor had very little correlation with other factors. However, there were small but statistically significant correlations between the Social Intent factor and three of the remaining six factors (Spoken Language, Compulsions, and Developmental Milestones) (Table 3). Also, a negative correlation was found between the Spoken Language and Compulsions factors (more severe compulsions were associated with better language).

Use of the Factors as Measures of Severity

The factors were constructed so that the items are highly intercorrelated. Thus, they form scales, and the factor scores can be used as ordinal approximations to continuously distributed variables. Evidence for their relationship to the severity of the constructs comes from some of the validation analyses shown below. The EVT,

PPVT, DAS, and Vineland are standard measures used to estimate severity of the constructs they represent.

Factor Validation

Face Validity. Most of the items that loaded on specific factors resulted in constructs with good face validity, reflecting well-known aspects of autism. However, the inclusion of a few specific items onto particular factors was unexpected, as mentioned above.

Construct Validity. Factor I, Spoken Language, was predicted to correlate with the EVT and PPVT combined score ($\rho = -0.30, p = .014$) (Table 4). The highest correlation was found between factor I and the total Vineland standard score ($\rho = -0.39; p = .001$). Factor II, Social Intent, correlated with the Vineland socialization standard score ($\rho = -0.37, p = .002$), but it correlated equally well with the Vineland total standard score ($\rho = -0.38, p = .002$). Factor III, Compulsions, correlated only with the compulsion score derived from KSADS-DD-PL ($\rho = 0.57, p = .000$). Factor IV, Developmental Milestones, correlated as predicted with the Vineland total score ($\rho = -0.27, p = .026$) and Full Scale IQ ($\rho = -0.26, p = .029$), but it correlated best with verbal measures, the PPVT and EVT combined score ($\rho = -0.43,$

TABLE 4
Correlation of the Autism Diagnostic Interview Factors With Validation Measures, Two-Tailed Tests (*N* = 68)

Factor	Validating Tests	Correlation
I. Spoken Language	EVT and PPVT combined score	$\rho = -0.30; p = .014$
II. Social Intent	Vineland socialization domain standard score	$\rho = -0.37, p = .002$
III. Compulsions	K-SADS Impairment and time spent on compulsions	$\rho = 0.57, p < .001$
IV. Developmental Milestones	Vineland total standard score	$\rho = -.27, p = .026$
	Full Scale IQ	$\rho = -.26, p = .029$
	PPVT and EVT combined score	$\rho = -.43, p < .001$
V. Savant Skills	Composite score of performance on Backward Word and Block Span	$\rho = .38, p = .017$
VI. Sensory Aversions	Phobia score derived from K-SADS	$\rho = 0.26, p = .054$

Note: EVT = Expressive Vocabulary Test; PPVT = Peabody Picture Vocabulary Test; K-SADS = Schedule for Affective Disorders and Schizophrenia for School-Age Children.

$p < .001$). Factor V, Savant Skills, correlated with a composite score of working memory, performance on Backward Word and Block Span ($\rho = 0.38, p = .017$), as predicted from the work of Treffert. Factor VI, Sensory Aversions, correlated with the “unusual sensory interests” score from the ADOS-G ($\rho = -0.27, p = .028$).

Relationship of ADI-R Factor Scores to Age

There has been some concern about the relationship between scores on some ADI-R items and age of the subject when the interview with the parent is administered. There has been no suggestion that parents’ memories of whether symptoms have ever occurred depend on age, but rather that memory of timing or severity may be affected by the age of the autistic child at the time the ADI-R was administered. We tested the correlation of factor scores with the age at the time when the ADI-R was administered, using the Pearson correlation coefficient. In this sample, only factor II, Social Interactions, was slightly correlated with age, with older children having higher (worse) scores ($r = 0.19, p = .009$).

Sib-Sib Correlations

Because we want to use the factors for genetic analyses, we tested the within-sib pair correlation of scores for each factor using intraclass correlations (Table 5). To develop the factors, we used only one sibling from multiplex families. This analysis included both siblings in these same families ($N = 192$ families). Significant correlations were found between siblings for five out of six factors; Social Intent was not significantly correlated but became so when age was covaried.

DISCUSSION

In a principal components analysis of items common to the original ADI and its revised version, the ADI-R, six factors were constructed that contain items with good

internal consistency and thus form scales, with good face validity, discriminant validity, and construct validity. Our primary goal was to “dissect” the autism phenotype into genetically relevant components. The sib-sib correlations are highly significant for five of the six factors, suggesting that they are appropriate for use in genetic analyses. Social Intent showed little correlation between affected siblings, similar to findings of Szatmari et al. (2002) and Silverman et al. (2002). The sib-sib correlation became significant for the Social Intent factor once the subject’s age at the time of ADI administration was covaried. The magnitude of the sib-sib correlations, while modest, is typically found for behavioral or physical traits.

The items that loaded on the first three factors, Spoken Language, Social Intent, and Compulsions, illustrate an emerging theme in discussions of the diagnostic criteria for autism. Several authors recently pointed out that the first two criteria, social interaction and communication, are so closely related that they should perhaps be considered a single criterion (Lord, 1990; Tanguay et al., 1998). Our factor II, Social Intent, included both social-affective items such as quality of social overtures, as well as communication items such as greetings, gestures, and nonverbal communication. The inclusion of both social and communicative items in this factor underscores the overlap between these diagnostic criteria.

In contrast, the language-related items that loaded on Spoken Language were related to verbal output rather than to communicative or pragmatic aspects of language. There is significant variability in the acquisition of spoken language among children with autism (e.g., Lord and Paul, 1997), with a significant minority of children remaining mute, while others achieve normal grammar and vocabulary. Kjelgaard and Tager-Flusberg (2001) recently investigated the variability in linguistic abilities among verbal children with autism, using a range of standardized language measures. Their study highlighted the pres-

TABLE 5

Intraclass Correlations (ICCs)

Factors	ICC	<i>P</i> Values for Associate <i>F</i> Statistics	Covarying for Age	
			ICC	<i>p</i>
Factor I: Language	0.20	.0002	0.16	.0185
Factor II: Social Intent	0.12	.0593		
Factor III: Compulsions	0.24	.0005		
Factor IV: Milestones	0.15	.0188		
Factor V: Savant Skills	0.29	.0001		
Factor VI: Sensory Aversions	0.30	.0001		

ence of a distinct subgroup of children with autism, representing about half of the sample, whose developmental language disorder was considered similar to specific language impairment (Leonard, 1998). The bimodal distribution of the scores on the spoken language factor (Fig. 1) illustrates the subgroups within the autism population of children with normal spoken language (factor scores .80 and higher), and those with impaired language or specific language impairment (factor scores below .5).

There is increasing evidence that abnormal language genes may be one of the risk factors for autism. Developmental delays in language and reading are more common in family members of autistic children than in families of controls (Cox et al., 1975), and the family members with a history of such delays have some features of reading impairment later in life (Folstein et al., 1999). Also, in three genetic linkage studies, subsetting the families on the basis of delayed onset of speech identified intervals of interest on chromosomes 2q (Buxbaum et al., 2001), 7q (Alarcon et al., 2002; CLSA, 2001), and 13q (CLSA, 2001). Two of the intervals on chromosomes 7q (Lai et al., 2001) and 13q (Bartlett et al., 2002) have been identified in studies of language-disordered families.

The third factor, Compulsions, included those items included as the third criterion for autism, repetitive and ritualistic behaviors that often seem to be driven. That is to say, the child seems to have to carry out these behaviors in a certain way and the behaviors are difficult to interrupt because the child becomes very upset. However, factor II does not include hand and finger mannerisms or other complex motor mannerisms. This finding confirms the recent work by Cuccaro et al. (in press). We were surprised that repetitive motor behaviors loaded with items related to Social Interaction. This aspect of autism has been little studied, but we have some data (unpublished) from parent interviews suggesting that repetitive motor behaviors occur in a different context than compulsions and are often used to avoid social interactions, whereas compulsions often require the involvement of other people.

The items that loaded on the last three factors—Milestones, Savant Skills, and Sensory Aversions—are not required for the diagnosis of autism and are quite variable from patient to patient. Delayed milestones correlated best with measures of verbal IQ rather than overall level of functioning. Sib-sib correlations for this factor, although significant, were small. In contrast, Szatmari et al. (2002) have shown, using cluster analysis, that very

low functioning, based on the Vineland, is highly correlated between siblings. From a genetic perspective, the Szatmari data suggest that some susceptibility genes for autism may influence severity, rather than any particular symptom. Our data would suggest that this severity factor is not reflected in milestones, which may be more related to low verbal skills rather than low skills overall.

The sib-sib correlation for Savant Skills was the highest and most significant of all the factors. We called the factor “Savant Skills” because the ADI-R question addresses a range of abilities that exceed the overall level of functioning, the definition of savant skills suggested by Treffert (1989). However, in the coding we had to rely solely on the report of the parent; the level of the particular skills displayed by the child was not measured by any psychometric instruments. Savant Skills were not correlated with other factors. Thus, they appear to be independent of other features of autism, IQ, socialization, and language. It is of interest that all these skills loaded on the same factor, even though they are usually thought to be rather different from each other. Again, this suggests some independent genetic risk factor for savant skills in general (see companion paper) and invites the study of parents of autistic persons who have them, as well as savants who are not autistic. Sensory Aversion is another unexplained variable feature of autism that formed its own factor and was correlated in siblings, suggesting the possibility of a separate genetic effect.

Clinical Implications

Our findings, along with those by Lord (1990) and Tanguay et al. (1998), suggest that we should perhaps return to the diagnostic criteria originally proposed by Kanner (1943), of which there were only two. His first criterion was “the inability of the children to relate themselves to others in the usual way.” In this he included both pragmatics and interest in other people. He fully discussed the variable ability of autistic children to speak, but this was not a criterion, probably because it varied so much from patient to patient. His second criterion was “the obsessive desire for the maintenance of sameness.” In this category he emphasized the compulsive nature of the ritualistic and repetitive behavior. While he described other repetitive motor behaviors, such as hand flapping, he emphasized as diagnostic those that were compulsive.

Limitations

Our use of both “current” and “ever” ratings could be questioned, since it gives more weight to those items that

have both kinds of ratings. In all cases, both loaded on the same factor. Both were used because each type of rating taps a somewhat different aspect of autism. A behavior rated only on “ever” but not on “current” will have a different severity than one rated on both time frames. It is variability in developmental trajectory that accounts for some of the variability in phenotype. Also, use of both items somewhat decreases the influence of missing variables.

Another limitation of the study was the relatively low correlations of the factors with the tests used for validation. However, this was expected because all the validating constructs have been designed for individuals with typical development and are not ideally suited for the population with autism spectrum conditions.

Finally, the principal component analysis was run under the assumption that children with autism represent a single population. However, the distribution of the factor scores for the Spoken Language factor was bimodal, suggesting the possibility that two different autism populations may exist, based their language levels. Thus, some bias may have been introduced by including all cases meeting criteria for autism in the derivation of the factors. In future such studies, it will be interesting to divide the sample by verbal ability to see whether the ADI variables form different clusters in these two subgroups.

Conclusion

The ADRI-R contains factors that can be used in numerous types of autism research as approximations to continuously distributed variables that are related to severity. The factor structure also supports recent discussions suggesting that two of the three currently standard criteria for autism, communication and social interaction, are not independent. The next step for a study like this will be to replicate these findings and to see whether they are useful in extracting biological meaning from the complex construct that is autism.

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