

Subtypes of Pervasive Developmental Disorder: Clinical Characteristics*

Deborah Fein^{1,2}, Michael Stevens¹, Michelle Dunn³, Lynn Waterhouse⁴, Doris Allen³, Isabelle Rapin³, and Carl Feinstein⁵

¹University of Connecticut, Storrs, ²Boston University School of Medicine, ³Albert Einstein College of Medicine, Bronx, NY, ⁴The College of New Jersey, Trenton, and ⁵Kennedy Krieger Institute, Baltimore, MD

ABSTRACT

Previous analysis of data from 505 preschool children with disordered communication, falling into the a priori categories of Pervasive Developmental Disorder (PDD), Developmental Language Disorder, and Nonautistic Low IQ, showed that PDD is statistically distinct from nonPDD. Current analyses of the 194 children with PDD also showed the existence of two subgroups within PDD. Children in these two subgroups are sharply divided on the basis of overall cognitive level; children in both groups share major symptom areas, but specific behavioral manifestations differ. Differing developmental trajectories into school age validate the distinction. A clinically usable algorithm for classifying PDD children into the two subgroups, based on social developmental level and degree of social abnormality, is provided. The findings suggest that high- and low-functioning individuals with PDD should be conceptualized as essentially distinct and should be studied separately for etiology, pathophysiology, course, and treatment.

Classification of Autistic Spectrum Disorders

Since the mid 1970s, a variety of researchers have called for exploration of possible subgroups within the autistic spectrum (Lotter, 1974; Reiss & Freund, 1990; Szatmari, 1992; Volkmar & Cohen, 1986). It has been repeatedly pointed out that identification of possibly distinct subtypes will be a necessary step in delineating the etiology, pathophysiology, course, and treatment options for children on the autistic spectrum (Roux, Garreau, Barthelemy, & Hameury, 1994). Following DSM-IV usage, we

will hereafter refer to the broad autistic spectrum as 'Pervasive Developmental Disorder' (PDD) and restrict the term 'autistic' to the more tightly defined Autistic Disorder as per DSM-IV (APA, 1994). Many studies done prior to the publication of DSM-IV have subject samples that were termed 'autistic', but that were defined more broadly or with different criteria than the current 'Autistic Disorder'. When describing these studies, therefore, we will refer to their samples as having PDD.

At present there are two general forms of subgrouping within the Pervasive Developmen-

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Address correspondence to: Deborah Fein, Dept. of Psychology, University of Connecticut, U-20, Storrs, CT 06269-1020, USA. E-mail: fein@uconnvm.uconn.edu.

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tal Disorders, as follows: (1) empirical classification conducted on varying subsets of features of the disorder, and (2) clinical consensus and field trial studies conducted on the complete set of diagnostic symptoms.

Empirical subgrouping

Some empirical classifications have focussed on behaviors and symptoms: Wing and Gould (1979) classified children with PDD on the basis of sociability (aloof, passive, active-but-odd), a classification which has been supported recently (Borden & Ollendick, 1994; Castelloe & Dawson, 1993; Volkmar, Cohen, Bregman, & Hooks, 1989); Allen (1988) classified children on the basis of communication and play.

Other studies have attempted classification on the basis of cognitive differences, such as overall cognitive strengths and weaknesses (Fein, Waterhouse, Lucci, & Snyder, 1985) or specific language deficits (Allen & Rapin, 1992; Simmons & Baltaxe, 1975). Other attempts at classification have focussed on presence or absence of EEG abnormalities (Tsai, Tsai, & August, 1985) age of onset, or developmental course (Burd, Fisher, & Kerbeshian, 1989; Percy, Gillberg, & Hagberg, 1990; Prior, Perry, & Gajzago, 1975; Volkmar, 1992), although no firm consistent findings have yet emerged relating onset or course to final behavioral outcome.

Some investigators have used a variety of empirical and statistical methods to define PDD subgroups. In an early study, Prior, Perry, and Gajzago (1975) found two clusters, one representing early onset and Kanner's type symptomatology, and the other representing later onset and more varied symptoms. Siegel, Anders, Ciaranello, Bienenstock, and Kramer (1986) performed a cluster analysis and identified four groups of children with PDD; although no measure of IQ was included, one of these types seems to correspond to high-functioning autism, and one to retarded autism, whereas the other two were marked by schizotypal features, and by negativism and anxiety. Bagley and McGeein (1989) found four clusters, related to mutism, speech pathology, and social responsiveness, with the mute and unresponsive subjects showing particularly poor outcome after 4 years.

Overall and Campbell (1988) identified anger, hyperactivity, speech deviance, and autism as the behaviors most able to discriminate subtypes of autism. Eaves, Ho, and Eaves (1994) identified four clusters of children with PDD, differing in behavioral profiles and cognitive level. Fein, Waterhouse, Lucci, and Snyder (1985), also using cluster analysis, identified eight cognitive profiles accounting for 51/54 children with PDD; these patterns were related to handedness, but not to autistic symptomatology.

Important beginnings have been made in identifying etiologies that may correspond to certain behavioral or cognitive profiles. Gillberg (1992) addressed this most directly, comparing subgroups of children with PDD with a defined etiology on a set of behaviors and symptoms, cautioning that etiological classification does not necessarily map well onto behavioral subgroups. Specific behavioral features that seemed to occur disproportionately with different biological conditions (although sample sizes per condition were too small for tests of significance) included IQ level, a somewhat remitting course after age 7, and a variety of specific symptoms. Other investigators have suggested subgroups marked by a variety of etiologies and biological markers (Coleman, 1990), although others present evidence that only a small minority of individuals with PDD have known medical conditions (Rutter, Bailey, Bolton, & LeCouteur, 1994).

Level of intellectual function has been found to be significantly correlated with degree of symptomatology in all three domains of autistic impairment (Volkmar, Cicchetti, Cohen, & Bregman, 1992), and most clustering or subtyping solutions identify children who differ in overall level of functioning (Dihoff, 1993). Consequently, some researchers have emphasized the overriding importance of IQ in defining subtypes, particularly with regard to outcome (Rutter & Garmezy, 1983). Cohen, Paul, and Volkmar (1987) make a strong case for a primary division between high- and low-functioning autism, pointing out the different patterns of communication, educational needs, outcome, and likelihood of neurological signs in high- and low-functioning autism. Tsai (1992) also con-

cludes that research supports the division into high- and low-functioning autism, and proposes a set of criteria for the distinction that would be usable within the ICD-10 system.

Indeed, level of intellectual function does seem to play a very important role in many of the subtyping findings mentioned above. The Wing and Gould subtypes have been found to be strongly related to IQ (Castelloe & Dawson, 1993; Volkmar, Cohen, Bregman, & Hooks, 1989). Two of the subtypes identified by Siegel seemed to represent high- and low-functioning autism (Siegel et al., 1986). Dahl, Cohen, and Provence (1986) identified two clusters of children with PDD who appear from the graphic data (although they did not apply statistical tests) to be similar in severity of behavioral impairment, but possibly to differ in intellectual functioning, family pathology, and age of onset. DeLong (1994) found a higher prevalence of neurological findings in low-functioning children with PDD and a higher prevalence of family psychiatric histories in high-functioning children with PDD. Others have also found different family histories in high- and low-functioning PDD, or autism and Asperger's syndrome (Gillberg, 1989; Piven et al., 1990).

Different levels of intellectual function have also been found to mark subgroups that differ in pharmacological response (August, Raz, & Baird, 1987), in comorbidity with Tourette's syndrome (Burd, Fisher, Kerbeshian, & Arnold, 1987) and with schizophrenia (Petty, Ornitz, Michelman, & Zimmerman, 1985), in minor physical anomalies (Links, 1980), in handedness (Fein, Waterhouse, Lucci, Pennington, & Humes, 1985; Soper, 1986), in more developmental regressions and unevenness (Kurita, Kita, & Miyake, 1992), in presence of a definable biological syndrome (Gillberg, 1992), in the development of seizures (Bartak & Rutter, 1976), and in seasonal birth differences (Konstantareas, Hauser, Lennox, & Homatidis, 1986).

Outcome also differs by initial IQ level. DeMyer et al. (1973) reported that children with PDD with high IQ tended to show some degree of behavioral and cognitive improvement over time, whereas children with lower IQs showed

relative intellectual declines. Bartak and Rutter (1976) also found poorer outcome with greater degrees of retardation, as well as more severely abnormal behaviors; similar results were reported by Waterhouse and Fein (1984), who found two patterns of development in adolescence. Cognitive level has been shown to be quite stable over time, and more high-functioning children with PDD, except for those with limited language, may more often show increases than decreases in cognitive scores (Lord & Schopler, 1989a, 1989b); this stability extends even into adolescence and adulthood (Venter, Lord, & Schopler, 1991). Lord and Venter (1992), Tsai (1992) and Cohen, Paul, and Volkmar (1987) are in agreement in placing a reasonable a priori cutoff for high functioning autism at an IQ level of 70.

Clinical consensus and field trial subgrouping

Clinical subgroups of PDD presently rest on two standard psychiatric systems, the DSM-IV (APA, 1994), and the ICD-10 (WHO, 1990). ICD-10 subclassifies PDD into six groups: Childhood Autism, Atypical Autism, Rett Syndrome, Childhood Disintegrative Disorder, Overactive Disorder associated with Mental Retardation and Stereotyped Movements, and Asperger's Syndrome. DSM-IV has retained Autistic Disorder (AD) and Pervasive Developmental Disorder Not Otherwise Specified (PDD-NOS) from the DSM-III-R (APA, 1987), but has added three of the ICD-10 groups: Rett Disorder, Childhood Disintegrative Disorder, and Asperger's Disorder.

In field trials for the DSM-IV (Volkmar, Klin, Siegel, & Szatmari, 1994), of 649 subjects with PDD, 454 had been previously diagnosed with AD, forming the clinical "gold standard" sample. From the remaining 240 cases with PDD, field trial clinicians diagnosed the following: Rett's Disorder = 15; Childhood Disintegrative Disorder = 16; Asperger's Disorder = 48; and PDD-NOS = 116. In this method of subgroup validation, diagnoses by the field trial clinicians are compared with "gold standard" diagnoses for sensitivity (the correct inclusion of cases), and specificity (the correct exclusion of cases). The diagnostic criteria used in field

trials that offer maximum sensitivity and specificity are deemed the most valid.

One difficulty with this method is that the gold-standard clinicians have been explicitly trained to recognize previously accepted diagnostic categories; their clinical judgment cannot be construed as necessarily representing the best means of “cutting nature at her joints” (Szatmari, 1992, p. 595).

In an effort to circumvent the problems of a clinical gold standard, Szatmari, Volkmar, and Walter (1995) applied latent class model (LCM) methodology to DSM-III, DSM-III-R, and ICD-10 diagnostic symptoms determined by medical file review for 342 children and adults with developmental disabilities. LCM analysis of the symptoms was used to split the sample into autistic and nonautistic groups. The researchers concluded that ICD-10 showed the best fit with the LCM-derived autistic and nonautistic groups. This methodology eliminates the circularity of a clinical gold standard, although coded symptoms may still reflect prior training in the diagnostic systems being evaluated. This procedure also assumes the existence of a single core group (autism), an assumption that should be empirically explored.

In a recent comparison of four systems of clinical classification (DSM-III, DSM-III-R, DSM-IV, and ICD-10) in a sample of 194 children with clinically diagnosed idiopathic PDD (Waterhouse et al., 1996), we found no difference between the four systems in the patterns of diagnostic features for AD and nonAD groups. Moreover, for all four diagnostic systems, AD cases had significantly lower adaptive functioning than did nonAD PDD cases. Our results suggested that a primary basis for isolating Autistic Disorder in all four clinical systems is severity of developmental impairment.

Although it is difficult to draw simple conclusions from this wide range of clinical and statistical subtyping studies, one common thread that does emerge is that PDD subgroups that differ in level of intellectual function may differ in many other domains of measurement. It is also clear that despite the utility of currently defined PDD syndromes, empirical efforts at ‘cutting nature at

its joints’ must still be pursued; because currently defined PDD syndromes do not yet have fully established reliability and validity, empirical subtyping efforts should include the entire PDD spectrum.

The purpose of the present study was to explore empirical subtypes in a large sample of preschool children with PDD. Specifically, we were interested in the following:

(a) whether statistically distinct subgroups could be identified within the PDD population, and if so: (b) what behaviors best discriminate the PDD subtypes, (c) what is the correspondence between the empirically derived PDD subtypes and clinically defined syndromes, (d) could a clinically usable algorithm satisfactorily classify the children into subgroups, and (e) would the outcomes of children in the subgroups converge or diverge?

Statistical evidence for the discontinuity between PDD and other disorders such as language disorder and mental retardation, as well as for two subtypes within the PDD spectrum, is briefly presented in Rapin (1996). Current results will focus on the correspondence between the statistically derived PDD subgroups and clinical diagnoses, and on characterizing the behaviors of children in the derived groups.

METHODS

Subjects

Detailed description of subjects including demographic information is found in Rapin (1996). The initial pool of subjects comprised 633 preschool children with disordered communication. Subjects were divided into those with Pervasive Developmental Disorder (PDD), Developmental Language Disorder (DLN), and Nonautistic subjects with low-IQ (LoIQ).

Children were recruited by (1) clinical referral to the clinician/researchers among the investigators, and by (2) solicited participation of schools and programs for children with special needs. Recruitment occurred at five geographically separated sites where the investigators were located: Albert Einstein College of Medicine (Bronx, NY), North Shore Children’s Hospital (Manhasset, NY), Case Western Reserve Medical Center (Cleveland, OH),

Boston University School of Medicine (Boston, MA), and Trenton State College (now The College of New Jersey, Trenton, NJ).

The sites were charged with recruiting different types of children: Children with PDD were recruited from the Boston, Trenton, and Bronx sites, children with DLD from Cleveland, Manhasset, and the Bronx, and each of the five sites recruited a share of the LoIQ cohort.

Because the goal of recruitment efforts was to insure an adequate number of children in low base-rate conditions, no effort was made to sample randomly or consecutively from referral sources. Therefore the present project should not be regarded as epidemiological.

General inclusion and exclusion criteria were as follows: age between 36 and 60 months (for DLD and normal children) and between 36 and 84 months (for PDD and LoIQ children); language of family: English; hearing: 25 Db or better (binaural). Because the goal of the project was to investigate classification of developmental disorders of higher cerebral function affecting communication, children were excluded if they had known and defined brain lesions, or diseases such as tuberous sclerosis or neurofibromatosis, or frequent and uncontrolled seizures. Children were also excluded if they had significant sensory or motor disabilities that would create substantial assessment problems, or if they were on high doses of anticonvulsant drugs.

For children who passed the general inclusion and exclusion criteria, completeness of their data was examined, and children lacking key measures ($n = 36$), or who had dropped out of the study ($n = 41$), were eliminated. These 77 children did not differ significantly in their demographic characteristics from other children recruited at the same site. Criteria for membership in clinical groups were applied to the remaining 556 children. (A flow chart of how children were referred, screened, and diagnosed is shown in Rapin (1996) p. 32).

Children were selected for the PDD sample as follows:

(1) Children had to have been identified by a clinical professional as having significant impairment in social relatedness. The initial screening to document the presence of the claimed social impairment was by means of the brief three-part Wing Autistic Disorder Checklist (WADIC), an interview questionnaire developed by Wing (1985; items listed in Table 5) in the development of DSM-III-R. The child's mother or teacher was interviewed with this instrument and the child was excluded from the PDD group if the child's mother or teacher did not endorse (1) at least two items in

the A section (impairment in social relatedness) or, (2) at least one item from each of the three sections: (A) social impairment, (B) impairment in social communication, and (C) repetitive or restricted activities. Two hundred and twenty-one children recruited as potential PDD subjects passed the initial screening.

(2) All children who passed the initial PDD screening were seen by a child psychiatrist at each site who conducted a structured comprehensive psychiatric evaluation. The psychiatrist rated each child for: (a) DSM-III (1980) diagnosis; (b) DSM-III-R (1987) diagnosis; (c) a symptom checklist of social abnormalities (items listed in Table 2); and (d) level of social relatedness on the basis of Wing's categorical system (Wing & Gould, 1979; see last item in Table 8). One hundred and ninety-four children were diagnosed with Pervasive Developmental Disorder; 176 of these children were diagnosed by the child psychiatrists with DSM-III-R Autistic Disorder (AD), and 98 were diagnosed with DSM-III Infantile Autism (details of the diagnosis rates and concordance between diagnostic systems, including later development of an algorithm for diagnosing DSM-IV autism, are presented in a separate paper by Waterhouse et al., 1996).

Many analyses were carried out with the 176 children with DSM-III-R AD (excluding the 18 children with PDD-NOS). These include analyses reported in the monograph (Rapin, 1996) that describes complete demographics and group data from all domains examined in the project. For the subgrouping analysis, however, it was considered important to include all 194 children with PDD. Because the 18 children with PDD-NOS are on the PDD spectrum, and cannot be assumed a priori to be reliably and validly distinct from the other children with PDD, they were included in the subgrouping analyses.

Assignment to the LoIQ and DLD groups was made as follows: All children referred to the project were screened with the WADIC (see above). The 283 children whose parent or teacher did not endorse at least two items from the social area, or one from each of the other three areas, were not seen by a study psychiatrist, and were considered for inclusion in the DLD or LoIQ group. If the psychiatrist saw a child but judged the child not to meet criteria for a PDD disorder, the child was placed back in their DLD or LoIQ group.

All children with no PDD who had nonverbal IQs lower than 80, and who met the general selection criteria, were placed in the LoIQ group. Placement in the DLD group was based on (1) no PDD diagnosis, (2) nonverbal IQ greater than or equal to

80, and (3) a significant deficiency on language measures, defined as a score on the Test of Early Language Development (Hresko et al., 1981) that was 15 points below their nonverbal IQ, or a mean length of utterance that was 1 SD below the mean for the child's chronological age (for details, see Rapin, 1996).

Demographic and IQ data are presented in Table 1. Nonverbal IQs were calculated from scores on the Stanford-Binet (4th ed.; Thorndike, Hagen, & Sattler, 1986), and for children who could not score at basal levels on the Stanford-Binet, scores were calculated from the Kent scoring of the Bayley Scales of Infant Development (Reuter, Stancin, & Craig, 1981). Nonverbal IQs were calculated as ratio IQs (CA/MA x 100), because use of standardized deviation IQs would have assigned a bottom score to many of the children. Mean nonverbal IQ score was 102.3 ($SD = 17.1$) for the children with DLD and 55.5 ($SD = 19.9$) for the children with LoIQ. Children in the PDD sample were divided into those with nonverbal IQs in the average range, that is, at or above 80, and those with below average nonverbal IQs, that is, below 80. Of the 194 children with PDD, 60 obtained normal nonverbal IQ scores ($M = 102.9$; $SD = 23.1$); the remaining 134 obtained subnormal nonverbal IQ scores ($M = 45.6$; $SD = 19.4$).

Measures

All subjects were given a comprehensive set of cognitive, behavioral, historical, neurological, and psychiatric measures. Characteristics by clinical group are described in detail in the monograph (Rapin, 1996). From the available preschool tests, ratings, and measurements, we identified those with the most promising discriminant validities for the PDD-nonPDD distinction and for possible PDD subgroups, as follows: (a) Items having clinical face validity for diagnosing autism or PDD, and evidence of statistical validity were selected; (b) Items with a common format were combined,

using unweighted sums, to form one or more subscales; (c) These subscales, gender, and SES were used as predictors in logistic regression prediction of diagnosis (age was not included, as the DLD sample was slightly younger than the other two samples); and (d) The weighted composite, in logit form, was used as a candidate indicator.

The psych scale consisted of all items from the Wing (1985) Autism Diagnostic Checklist; item list can be found in Table 5. The parent scale consisted of all items from the parent report of autistic type behaviors in the child's development. Items from this scale that discriminated the two PDD subgroups are listed in Table 7 and the entire instrument is described in Rapin (1996).

The teacher scale consisted of selected items from the Schedule of Handicaps, Behaviors and Skills teacher report instrument (Wing, 1982). Items from this scale that discriminated the two PDD subgroups are found in Table 8 and the entire schedule is reprinted in Rapin.

In addition to these scales, individual scores and items were used from the following: *Peabody Picture Vocabulary Test - R* (Dunn & Dunn, 1981), a test of single word receptive vocabulary, *Nonverbal IQ*, as described above, *Expressive One-Word Picture Vocabulary Test* (Gardiner, 1979), a test of single word expressive vocabulary, *Vineland Adaptive Behavior Scales* measures of *Communication*, *Daily Living*, and *Socialization* (Sparrow, Balla, & Cicchetti, 1984), and *Social Abnormalities Scales*, developed for the present project, described in Rapin (1996) and items listed in Table 2. Items are scored 0 (behavior not present), 1 (mild or occasional), or 2 (frequent or marked).

Measures that proved most successful in discriminating PDD subgroups are described below; descriptive statistics for the subgroups at both preschool and school age are provided for the other measures (Tables 5–11).

Table 1. Sample Characteristics.

	Group			
	PDD	DLD	LoIQ	Total
<i>n</i>	194	201	110	505
% Male	84	74	54	73
Mean age (months)	59.1	49.0	59.6	55.19
Mean nonverbal ratio IQ	63.32	102.3	55.5	77.13

Note. PDD=Pervasive Developmental Disorder; DLD=Developmental Language Disorder; LoIQ=Nonautistic Low IQ.

Table 2. Items Comprising Social Abnormalities Scales.

Social Abnormalities I	Social Abnormalities II
1. seem very hard to reach or 'in a shell'	1. exhibit excessive and/or unusual fears
2. walk through or over people as if they do not exist	2. seem unaware of social rules of interpersonal physical contact
3. respond to affection or touching by withdrawing, or by becoming rigid or limp	3. exhibit or repeatedly discuss unusual preoccupations
4. show aversion to people, preferring to engage in solitary activity	4. masturbate or undress in public with no apparent awareness of inappropriateness
5. acquire things by directing person's hand	5. tend to be excessively clingy and overdependent, unable to tend to own basic needs or become socially engaged without constant support from a familiar adult
6. have difficulty maintaining eye contact; avoid looking at people or 'looking through them'	6. repeat words or phrases from the past which have no discernible connection with the present social context
7. lack empathy for other people's needs, interests, or ideas; continue to pursue his/her own favorite preoccupations despite all attempts to dissuade	7. while interacting, have a 'not really there' quality
8. fail to initiate interaction with adults solely for social purposes (aside from need fulfillment)	8. become excessively excited or overstimulated by social contact or interactive play
9. react to strangers and familiar persons indiscriminately	9. seem unable to become fully engaged in reciprocal dialogue initiated by a conversational partner
10. shows any attachment behaviors to mother (and possibly a few other familiar people), but no social behavior in relation to most adults and children	10. engage in obsessive rituals without apparent awareness of possible reactions of others
11. Have intense prolonged temper outbursts or panic attacks with no visible motivation and inability to be soothed or consoled by familiar persons	

Note. Items are scored as 0 (not present), 1 (mild or occasional), 2 (marked, severe, or frequent). Social Abnormalities I score ranges from 0–22, Social Abnormalities II from 0–20.

Testing at School Age

All children were recontacted at age 7 and 9, and those willing to continue in the project were given a comprehensive battery including assessment of behavior, skills, and symptoms, neurological status, educational achievement, play and interaction, and neuropsychological functioning. Detailed analysis of longitudinal trends from preschool to school age is ongoing and will appear elsewhere. A subset of major, completed variables administered at the school age and preschool testings were examined for developmental trajectories in the autism subgroups. These measures included the Peabody Picture Vocabulary Test, the Stanford-Binet verbal and abstract visual standard scores, VABS scores, and the Wing (1985) Autism Diagnostic Checklist. School-age data were available from 121 children with diagnoses of PDD. When preschool and school-age data are presented together, only children who were seen at both time points are included.

Detection of Subgroups

Detection of subgroups depends heavily on statistical methods; problems with earlier methods led to

the adaptation and use of the regression-mixture model proposed by Golden and colleagues. This model originated with early approaches by Meehl and Golden (1982), and resulted in an heuristic version (Golden, 1991) and the present maximum-likelihood model (Golden & Mayer, 1995). The model can be viewed as an extension of the univariate mixture model, where a manifest distribution of a single variable is analyzed for the component distributions (presumed to arise from separate groups) that may comprise it. In the Golden regression-mixture model, the manifest bivariate distribution for pairs of variables is so analyzed. Possible subgroups are indicated by a strongly sigmoidal smoothed regression line. Figures depicting such discontinuity for both PDD-nonPDD, and for the two PDD subgroups can be found in Rapin (1996, Chapter 10).

The following general sequence of tests is performed: (1) Logistic regression and regression classification trees are used to select the best clinical neurobehavioral indicators for the conjectured subgroups. The finding of subgroups should be largely independent of specific measures selected, and therefore replicable across measures. In the current

case, the best indicators were the VABS scores, the number of symptoms on the Wing (1985) Autistic Disorder Checklist, Social Abnormalities I, and nonverbal IQ (as well as derived scales comprised of items from the neurological exam, parent interview, and teacher interview); (2) Multiple regressions using pairs of these variables were analyzed for the presence of distinct subgroups in the PDD sample; (3) Because two groups were detected, the probability of belonging to each subgroup was estimated for each individual child in the sample; (4) The cognitive and behavioral profiles of children so assigned was analyzed; and (5) Algorithms for calculating the probability of a particular child's belonging to the lower functioning group were derived. The cognitive and behavioral profiles and the algorithms are the focus of this paper.

This regression mixture analysis, in addition to allowing estimation of the means and variances on indicator variables for subjects in the group and its complement, also allows estimation of the subgroup base rates in the combined sample, and assessment of model validity by checking goodness of fit. For a more complete mathematical description of the regression-mixture method, and examples of its application to other large data sets, see Golden and Mayer (1995), and Rapin (1996, Chapter 10). It should be emphasized that, unlike cluster analyses, the method outlined by Golden and Mayer does not necessarily detect subgroups. If the sigmoidal bivariate regression line indicates the presence of subgroups, as in the current case, then description of the subgroups can proceed.

Subsequent to the detection of subgroups by the Golden method, traditional hierarchic cluster analyses were used to confirm the subgroups; there was good correspondence between groups detected by the Golden method and by cluster analysis (see below).

RESULTS

Subtypes of PDD

To examine subtypes of PDD, data were examined from all the children placed in the PDD category by empirical analyses described in Rapin (1996, Chapter 10). This group comprised 93% of the children with a priori diagnoses of PDD, plus a small number of children with LoIQ and DLD (see next section). Loess smoothed regression curves of Nonverbal IQ, Social Abnormalities I, Vineland Socialization, Vineland Daily Living, and number of symptoms on the

Wing checklist (Wing, 1985), taken two at a time, all showed the sigmoidal shape characteristic of dichotomous groupicity (see Rapin, 1996). The two groups are referred to as PDD A (higher functioning) and PDD B (lower functioning). Base rates of the two groups are roughly equal in the total PDD group, with PDD A accounting for 49% and PDD B accounting for 51% of the overall PDD group.

Relationship of Empirical Assignment to a *Priori* Diagnosis

It can be seen (Table 3) that the majority of high functioning children with PDD ($IQ > 79$ a priori diagnosis) were placed in PDD Group A (empirical diagnosis), and the majority of low functioning children with PDD ($IQ < 80$) were placed in the PDD B group. Empirical analysis suggested that 17 children with DLD should be classified as PDD; all of these were placed in PDD Group A. Analysis also suggested that 34 children with LoIQ should be classified as having PDD; these children were divided between PDD groups A and B. The fact that children with DLD and high-PDD were placed primarily in PDD Group A, whereas children with low-PDD and LoIQ had substantial assignment to both groups, suggests that the dividing line of 80 IQ may be too high. Of the children receiving psychiatric diagnoses of DSM-III-R PDD-NOS ($n = 18$), 78% were classified in the PDD group (compared to 95% of the 176 children with DMS-III-R Autistic Disorder), placing them into the PDD A and B groups in approximately equal numbers.

Longitudinal Stability of Autistic Subtypes

Preliminary analyses of group assignments at school age as compared to preschool assignments, are shown in Table 4. These data indicate that most children remained in their PDD versus nonPDD designation, with a highly significant relationship between preschool and school age classification ($\chi^2 = 220, p < .001$ for subtypes, $\chi^2 = 115, p < .001$ for PDD vs. nonPDD). Of those who shifted, the majority moved from PDD to nonPDD, and the majority of these were classified as high functioning. Relatively few low-functioning children with PDD shifted out

Table 3. Empirical Classification by Clinical Diagnosis.

Empirical Classification	Clinical Diagnosis				Total
	H-PDD	L-PDD	DLD	LoIQ	
Group A	49	30	17	19	115
Group B	4	98	0	15	117
NonPDD	8	4	182	68	262
Missing	0	1	2	8	11
Total	61	133	201	110	505

Note. H-PDD = normal nonverbal IQ PDD; L-PDD = nonverbal IQ below 80; DLD = developmental language disorder; LoIQ = nonautistic low IQ.

of the PDD classification by school age. Few children not assigned to the PDD group at pre-school were so assigned at school age.

Characteristics of Children in the PDD Groups A and B

Children in the PDD Groups A versus B were compared on multiple variables to determine the average functioning of the children in the groups, and to provide evidence of variables with the most discriminant validity. Many variables from the Wing Autism Diagnostic Checklist significantly discriminated children in the two groups (Table 5). A very high proportion of children in both groups showed abnormalities in the use of eye contact in interaction, as well as impairments in social play and sharing of interests, but the children in Group B showed a higher prevalence at the lower developmental

levels, such as impaired or absent lap play, and pointing. Abnormal greeting was also prevalent in both groups, with lack of spontaneous waving and ignoring visitors more prevalent in Group B. Seeking of comfort was not generally different between the groups, but the children in Group B were more likely to ignore others' distress whereas children in Group A were somewhat more likely to offer inappropriate help. Although both groups were impaired in imitative skills, the children in Group B showed much higher rates of complete failure to imitate, whereas the children in Group A showed somewhat higher rates of naive or bizarre imitation. Most children in both groups were impaired in peer relationships, with children in Group A showing more naive and inappropriate relationships and children in Group B showing a higher prevalence of a total lack of peer relation-

Table 4. Preschool by School Age Assignment.

Preschool	School age			Total
	Group A	Group B	NonPDD	
Group A	39	6	24	69
Group B	8	54	5	67
NonPDD	6	3	96	105
Total	53	63	125	241
	PDD		NonPDD	
PDD	107		29	
NonPDD	9		96	

Note. PDD = Pervasive Developmental Disorder.

Table 5. Wing Autistic Disorders Checklist Items.

Item	Group A (<i>n</i> = 69)		Group B (<i>n</i> = 67)		<i>t</i>	<i>p</i>
	<i>M</i>	(<i>SD</i>)	<i>M</i>	(<i>SD</i>)		
SOCIAL DOMAIN						
Poor eye contact, posture, expression	.80	(.41)	.87	(.34)	-1.06	
Doesn't anticipate holding	.06	(.24)	.10	(.31)	-0.99	
Resists cuddling	.16	(.37)	.12	(.33)	0.67	
No look on social approach	.22	(.42)	.24	(.43)	-0.30	
No eye contact to get attention	.42	(.50)	.63	(.49)	-2.45	.02
Inappropriate eye contact	.09	(.28)	.15	(.36)	-1.12	
No variation in gaze to guide conversation	.39	(.49)	.46	(.50)	-0.84	
Impaired play or shared interests	.90	(.30)	.93	(.26)	-0.55	
No lap play	.12	(.32)	.49	(.50)	-5.18	.001
No pointing to share attention	.29	(.46)	.75	(.44)	-5.94	.001
No bring toy to show or share	.46	(.50)	.69	(.48)	-2.68	.01
Prefers solitary play	.68	(.47)	.79	(.41)	-1.46	
Involves other children mechanically	.07	(.26)	.15	(.36)	-1.42	
Directs other children as puppets	.12	(.32)	.06	(.24)	1.16	
Accepts passive role in play	.15	(.36)	.15	(.36)	-0.07	
Engages other around shared narrow interest	.20	(.41)	.12	(.33)	1.33	
Abnormal greeting	.77	(.43)	.91	(.29)	-2.29	.02
No rush to greet parent	.22	(.42)	.31	(.47)	-1.27	
No spontaneous wave	.38	(.49)	.73	(.45)	-4.42	.001
Ignores visitors	.29	(.46)	.57	(.50)	-3.38	.001
Greets with stereotyped phrases	.49	(.50)	.37	(.49)	1.41	
Approaches others indiscriminately	.19	(.39)	.31	(.47)	-1.68	
Abnormal seeking comfort	.59	(.50)	.64	(.48)	-0.57	
Never seek comfort	.07	(.26)	.10	(.31)	-0.65	
Seek comfort mechanically	.03	(.17)	.21	(.41)	-3.33	.001
Shows distress but no seeking comfort	.35	(.48)	.34	(.48)	0.06	
Asks for stereotyped comforting	.10	(.30)	.06	(.24)	0.89	
Intrudes, clings	.20	(.41)	.18	(.39)	0.35	
Abnormal giving comfort	.70	(.46)	.84	(.37)	-1.95	.05
Ignores others' existence, feelings	.30	(.46)	.64	(.48)	4.16	.001
Indifferent to or laughs at distress	.26	(.44)	.60	(.49)	4.18	.001
Minds other's pain because of change in routine	.23	(.43)	.10	(.31)	2.01	.05
Offers naive or inappropriate sympathy	.23	(.43)	.10	(.31)	2.01	.05
Impaired imitation	.75	(.43)	.94	(.24)	3.12	.002
No spontaneous imitation	.06	(.24)	.51	(.50)	6.64	.001
Automatic, mechanical imitation	.26	(.44)	.16	(.37)	1.38	
Imitates only simple movements	.39	(.49)	.51	(.50)	1.36	
Imitates actions but stereotyped	.17	(.38)	.09	(.29)	1.46	
Naive, bizarre imitation	.20	(.41)	.06	(.24)	2.52	.02
Impaired peer relationships	.81	(.39)	.84	(.37)	0.37	
No peer friendships	.52	(.50)	.73	(.45)	2.57	.01
Teased and bullied	.17	(.32)	.15	(.36)	0.57	
Any peer referred to as 'friend'	.26	(.44)	.02	(.12)	4.45	.001
Plays passive role with one friend	.16	(.37)	.09	(.29)	1.23	
Has friend with same narrow interest	.06	(.24)	.03	(.17)	0.80	
Impaired social play	.86	(.36)	.88	(.33)	0.44	
Fails to animate toys	.35	(.48)	.72	(.45)	4.60	.001
Animates repetitively	.45	(.50)	.22	(.42)	2.85	.005
Repetitive fantasy	.13	(.34)	.03	(.17)	2.19	.03

Table 5 continues.

Table 5. (Continued.)

Item	Group A (<i>n</i> = 69)		Group B (<i>n</i> = 67)		<i>t</i>	<i>p</i>
	<i>M</i>	(<i>SD</i>)	<i>M</i>	(<i>SD</i>)		
Impaired awareness of social rules	.61	(.49)	.82	(.39)	2.80	.01
Lacks modesty	.26	(.44)	.66	(.48)	5.01	.001
Lacks awareness of personal space	.29	(.46)	.55	(.50)	3.19	.002
Unaware of conversation taboos	.07	(.26)	.05	(.21)	0.68	
Public scenes	.30	(.46)	.45	(.50)	1.73	
COMMUNICATION DOMAIN						
Impaired use of language for communication	.83	(.38)	.91	(.29)	1.46	
Lacks meaningful vocalization or gesture	.06	(.24)	.25	(.44)	3.23	.002
No spoken language	.17	(.32)	.43	(.50)	4.38	.001
Speaks but can't initiate or sustain conversation	.46	(.50)	.40	(.49)	0.71	
Content one-sided, no turn-taking	.39	(.49)	.16	(.37)	3.04	.003
Impaired comprehension of language	.84	(.37)	.90	(.31)	0.94	
No response to language	.04	(.21)	.13	(.34)	1.87	
Responds to simple commands in context	.52	(.50)	.76	(.43)	2.99	.003
Responds to single words/phrases	.33	(.48)	.16	(.37)	2.31	.02
Overliteral interpretation	.22	(.42)	.05	(.21)	3.08	.003
Impaired use of speech	.74	(.44)	.45	(.50)	3.59	.001
Stereotyped, repetitive speech, echolalia	.57	(.50)	.45	(.50)	1.37	
Pronoun reversals, word errors out of context	.39	(.49)	.10	(.31)	4.09	.001
Idiosyncratic use of words/phrases	.16	(.37)	.03	(.17)	2.64	.01
Pedantic speech	.06	(.24)	.02	(.12)	1.34	
Abnormal pitch, rate, stress	.54	(.50)	.33	(.47)	2.49	.01
Impaired symbolic development	.84	(.37)	.69	(.47)	2.13	.05
No use of miniature objects	.19	(.39)	.37	(.49)	2.43	.02
Limited, unspontaneous use of miniatures	.19	(.39)	.18	(.39)	0.14	
Spontaneous but repetitive use	.59	(.50)	.25	(.44)	4.25	.001
Elaborate but repetitive representational play	.15	(.36)	.05	(.21)	2.02	.05
RESTRICTED BEHAVIOR REPERTOIRE						
Stereotyped posture or movement	.57	(.50)	.88	(.33)	4.37	.001
Stays in one position	.06	(.24)	.06	(.24)	0.04	
Moves around aimlessly	.42	(.50)	.64	(.48)	2.64	.01
Repetitive body movements	.10	(.30)	.37	(.49)	3.89	.001
Repetitive complex movements	.13	(.34)	.34	(.48)	2.99	.003
Stereotyped activity of body function/sensations	.26	(.44)	.58	(.50)	3.98	.001
Smearing saliva or excreta	.03	(.17)	.15	(.36)	2.49	.02
Swallows inedibles	.00	(.00)	.13	(.34)		
Self-injury	.03	(.17)	.13	(.34)	2.26	.03
Preoccupied with sensations	.23	(.43)	.46	(.50)	2.89	.005
Preoccupied with objects	.59	(.50)	.64	(.48)	0.57	
Unusual attachment to objects	.20	(.41)	.19	(.40)	0.13	
Collects objects	.07	(.26)	.08	(.27)	0.05	
Arranges in lines	.28	(.45)	.25	(.44)	0.28	
Preoccupied with parts of objects	.07	(.26)	.18	(.39)	1.88	
Preoccupied with repetitive acts involving objects	.26	(.44)	.40	(.49)	1.77	
Preoccupied with attributes of objects or people	.20	(.41)	.16	(.37)	0.58	
Preoccupied with maintenance of small details	.17	(.38)	.19	(.40)	0.30	
Preoccupied with maintenance of routines	.26	(.43)	.34	(.48)	1.24	
Restricted patterns of interests	.46	(.50)	.13	(.34)	4.48	.001
Asks same question	.26	(.44)	.08	(.27)	2.99	.003
Acts role repetitively	.01	(.12)	.00	(.00)		
Preoccupied with special interests	.25	(.43)	.09	(.29)	2.49	.02
Life style empty, routinized, little spontaneity	.28	(.45)	.28	(.45)	0.11	

ships. Most children in both groups also showed impaired play, with children in showing more stereotyped play and children in Group B showing more total lack of pretend play.

The same pattern was seen in the Communication domain, where most children in both groups were impaired in the use of language for communication, with children in Group A higher on one-sided conversation, and the children in Group B higher on mutism. Children in Group B were more likely to comprehend only simple commands in context, whereas children in the Group A were more likely to respond to words and phrases or be overliteral in their comprehension.

The same developmental pattern was seen in the Restricted Repertoire domain, where stereotyped postures and movements and sensory preoccupations (“self-stimulation”) were found more frequently in Group B, preoccupations with objects were found with equal frequency in the two groups, and preoccupations with special interests or topics were found more often in Group A. Resistance to change was found with surprisingly low frequency in both groups.

Number of items checked in each domain at preschool and school age are shown in Table 6. In the social domain, patterns of means and medians were consistent, and showed that the children in Group A improve, whereas the children in Group B remain about the same. In the Communication domain, children in Group A showed

some improvement in the number of symptoms displayed. For both Communication and Restricted Repertoire, Group B children showed a small mean improvement, but unchanged median, suggesting only changes in the extreme scores. For Restricted Repertoire, Group A children show improved median score (3 to 1), but a mean that actually increased slightly, suggesting that most children showed fewer symptoms of restricted behaviors, but that a small number become substantially worse.

Items from the *Parent Report* history that highly discriminated children in Groups A and B are shown in Table 7. Twelve of the 30 items differentiated the groups at $p < .02$ or less. Behaviors seen more frequently in the histories of children in Group A were “respond to questions by repeating the question or failing to respond” and “has one line of pretend play that they go over and over with little or no variation...”, whereas all other items were more frequently seen in Group B, including remoteness, pica, mood changes, repetitive activities, and gestures. Items that were most highly endorsed for children in each group included ignoring or repeating questions for Group A, and low level play, no interactive games, and repetitive gestures for Group B. No item related to resistance to change or interruption, or perseverative activity discriminated the groups. Other items of interest that did not differentiate Groups A and B included echolalia, quiet as a baby, inappropri-

Table 6. Mean and Median Number of Items Checked on Each Domain of the Wing Autistic Disorders Checklist.

Domain	Preschool		School age	
	Group A (n = 69)	Group B (n = 67)	Group A (n = 53)	Group B (n = 63)
Social (0–9)				
Mean	6.78	7.66 ^a	4.44	7.25 ^b
Median	7	9	5	8
Communication (0–5)				
Mean	3.78	3.27 ^a	1.94	1.74 ^b
Median	4	3	2	3
Restricted repertoire (0–7)				
Mean	2.58	3.06	2.92	1.49 ^b
Median	3	3	1	3

^a Groups A vs. B means significantly different by t test at .02.

^b Groups A vs. B. means significantly different by t test at .001.

ate use of toys, ignoring affection, catastrophic reactions or tantrums, and anxiety/tension.

All items from the *Teacher Questionnaire* that were clearly developmental in nature highly discriminated Groups A and B, including comprehension of speech, of prepositions, development of grammar, asking questions, intelligibility, and educational achievements. Specific language abnormalities did *not* differentiate the groups, including immediate and delayed echolalia, pronoun reversal, idiosyncratic usage, repetitive speech, and muddled sequences. Specific aspects of social behavior that also did *not* differentiate the groups included amount and social use of eye contact, and spontaneous shows of affection. Other social and communicative behaviors that did highly discriminate children in Groups A and B are shown in Table 8; they include especially aspects of nonverbal communication, symbolic play, and peer relationships. As can be seen in Table 8, Group A mean score on Wing's social typology lies between 'responds to physical contact' and 'passive', and included many children who were 'active but odd' whereas the mean score of Group B children was between 'responds to physical contact' and 'aloof'.

Standardized test scores for Groups A and B are shown in Table 9. Except for Stanford-Binet (S-B) Vocabulary (where only 10 Group B children were in scorable range), Group A children

scored higher than Group B children. As would be expected, Group A children showed a profile of relative strengths in abstract visual reasoning, and relative weaknesses in verbal comprehension and all domains of adaptive behavior.

Medians were also calculated for standardized test scores, using *all* children, by assigning the lowest score to children who were unable to achieve basal scores. For Group A children, medians were within 1 point of means, except for PPVT, where the median was 5 points higher. For Group B children, means were within a few points of medians except on the S-B. Because fewer than half of the children were able to score at basal levels on the S-B, medians were 0.

Standardized testing at school age was analyzed to determine longitudinal trends for children in the A versus B groups. Data were available on only a subset of the preschool children, and more detail on the longitudinal follow-up will be presented in separate publications. To ensure that the subset on whom standardized testing were available at follow-up were not different from the total preschool sample, standard and behavioral scores were compared for the total sample and the subset (Table 10). In all cases, mean behavioral scores and standardized scores of the two samples were very close. Scores for preschool and school age, for children with data at both time points, are found in Table 11. The scores and profiles were generally

Table 7. Highly Discriminating Items from Parent History.

Item	Group A (<i>n</i> = 69)		Group B (<i>n</i> = 67)		<i>t</i>	<i>p</i>
	<i>M</i>	(<i>SD</i>)	<i>M</i>	(<i>SD</i>)		
Ignores questions	1.46	(.80)	1.10	(.96)	2.38	.02
Sensorimotor play only	.97	(.95)	1.39	(.83)	2.71	.01
Dislikes interactive games	.99	(.95)	1.39	(.87)	2.58	.01
Repetitive pretend play	.99	(.90)	.45	(.80)	3.68	.001
Repetitive gestures	.91	(.85)	1.48	(.78)	4.02	.001
Repetitive activities	.80	(.92)	1.21	(.90)	2.65	.01
Remote w. familiar people	.57	(.83)	.97	(.90)	2.72	.01
Unaware of painful falls	.27	(.66)	.70	(.82)	3.34	.001
Underactive	.13	(.42)	.36	(.71)	2.29	.02
Laugh or cry unexpectedly	.58	(.76)	1.02	(.75)	3.37	.001
Unaware of mom's absence	.32	(.68)	.60	(.82)	2.16	.03
Mouths inedibles	.44	(.74)	1.03	(.89)	4.25	.001

Note. 0 = absent, 1 = sometimes, 2 = marked/frequent.

Table 8. Discriminating Items from Teacher Questionnaire.

Item	Group A		Group B		<i>t</i>	<i>p</i>
	<i>M</i>	(<i>SD</i>)	<i>M</i>	(<i>SD</i>)		
Comprehend gesture (0–4)	2.99	0.78	2.13	0.99	5.47	.001
Comprehend face expression (0–2)	1.05	0.62	.67	0.68	3.22	.002
Copy gesture(0–5)	3.00	1.02	2.13	1.07	4.73	.001
Use gesture symbolically (0–2)	1.06	0.75	.56	0.71	3.93	.001
Use gesture as sub for speech (0–5)	2.66	1.50	1.71	1.11	3.97	.001
How obtain needs (0–8)	5.18	2.20	3.69	2.09	3.93	.001
Willing to communicate (0–7)	3.72	1.67	2.27	1.62	5.04	.001
Shares interests (0–3)	1.57	0.74	1.08	0.69	3.93	.001
Level of play (0–8)	3.09	1.98	.84	1.26	7.80	.001
Ability to make friends (0–5)	2.15	1.14	1.15	1.10	5.08	.001
Level of social play (0–7)	2.96	1.41	1.70	1.23	5.46	.001
Joins peers in leisure activity (0–4)	1.47	0.89	.82	0.86	4.15	.001
Att'n span for enjoyed activity (0–2)	1.51	0.53	1.08	0.58	4.38	.001
*Quality social interaction (Wing type 0–6)	2.52	1.21	1.89	1.32	2.84	.005

*0 = does not interact – aloof and indifferent; 1 = interacts to obtain needs, otherwise indifferent; 2 = responds to (and many initiate) physical contact only; 3 = generally does not initiate, but responds to social (not just physical) contact, if others, including age peers, make approaches. Joins in passively. Tries to copy but with little understanding. Shows some pleasure in passive role; 4 = Makes social approaches actively, but these are usually inappropriate, naive, peculiar, or bizarre – one-sided. Behavior is not modified according to needs, interests, and responses of person approaches; 5 = Shy, but social contacts appropriate for mental age with well known people, including age peers; 6 = Social contacts appropriate for mental age with children and adults.

stable, with school-age scores moderately predictable from preschool scores (for Group A, all preschool-school age scores were correlated between .42 and .66, for Group B, scores at the two time points were correlated between .47 and .71). For the children in Group A all test scores increased slightly, with significant increases in

sentence memory and expressive vocabulary and trends for socialization, comprehension, and verbal reasoning. The children in Group B, in contrast, showed declines on all test scores, with significant declines on all tests except for Abstract Visual Reasoning (nonverbal IQ), the only nonverbal test examined. (See Table 11). These

Table 9. Standard Score Means.

	Group A		Group B		<i>t</i>	<i>p</i>
	<i>M</i>	(<i>SD</i>)	<i>M</i>	(<i>SD</i>)		
Stanford-Binet Abstract Visual	94.82	20.63	66.57	14.55	6.89	.001
Stanford-Binet Verbal	80.76	14.34	65.89	17.38	3.28	.003
S-B Vocabulary (ave. = 50)	43.41	6.24	41.10	11.84	0.60	ns
S-B Comprehension (ave. = 50)	40.25	6.75	35.46	7.45	2.08	.05
S-B Short-Term Memory	82.08	13.61	68.47	17.87	2.87	.01
Peabody Picture Vocabulary Test	71.29	19.74	49.03	16.98	5.25	.001
Expressive One-Word Picture Vocabulary Test	84.63	21.60	66.15	21.27	3.70	.001
Vineland Communication	75.20	16.79	47.88	9.73	11.65	.001
Vineland Socialization	68.33	10.96	53.00	6.12	10.11	.001
Vineland Daily Living	68.28	13.53	46.25	12.22	9.97	.001

declines may be due in part to the greater number of children able to take the tests at school age; examination of medians, however, shows that the decline was not totally due to mean effects. Stanford-Binet scores remained at a median of 0, but the median scores decreased for the following: PPVT from 45–40; Vineland Communication from 48–37; Vineland Socialization from 52–49; and Vineland Daily Living from 50–29. Thus, the decline in functioning relative to age peers seemed genuine.

Validation of Subgroups

A cluster analysis was performed with SAS using nonverbal ratio IQ and Vineland subdomain scores. The cluster method chosen was the *k*-means method applied to coordinate data. In this method, a transformation of scale is performed on each variable so that Euclidean distances among cases have the same scale (standardization). The number of cluster, *k*, is predetermined, and each case is classified in only one of the *k* clusters. In this case, because the results were to validate a two-group solution, *k* = 2 was chosen. The purpose of the method is to minimize the sum of the absolute distances by moving cases from one cluster to another. Unlike many other clustering methods, this method is appropriate for large datasets as well as those datasets in which variables are not independent. This analysis found a solution with a high functioning group (*n* = 65) and a lower functioning group (*n* = 75). The correspondence with the A versus B grouping was: 75 children in the higher group by both analyses, 71 children in the lower

group by both analyses, 31 children in the higher cluster group but members of Group B, and 4 children in the lower cluster group but in Group A. Thus, there was agreement of 81% between the two methods.

Characteristics of Children Diagnosed as PDD by a Priori Clinical, but Not Empirical, Criteria

Twelve children with clinically diagnosed high or low functioning PDD were not assigned to either Group A or B (see Table 3). Characteristics of these children are shown in Table 12. By comparing these characteristics to those of children in Groups A and B (Tables 6 and 9), it can be seen that the 12 children are quite high functioning: they show less autistic symptomatology in all three domains than children in either Group A or B, and have generally higher language (although not nonverbal) scores than children in Groups A and B.

Algorithm for Assigning Children to the A and B Groups

Because the multiple regression methods used here employed logit forms of derived scales, assembled from items across diverse measures, it was considered important to find a small set of clinically usable variables that would allow other researchers and clinicians to determine the probability of a child belonging to the A or B group. For these analyses, we considered the 194 children with PDD, excluding the 13 children for whom the analysis had determined a nonPDD placement (*n* = 181). Classification of

Table 10. Characteristics of Total Preschool Sample and Preschool Sample with School-Age Data.

Test/Measure	Group A		Group B	
	Total Presch.	Pre. + School	Total Presch.	Pre. + School
Autistic Disorder # of social symptoms	6.48	6.78	7.32	7.66
Autistic Disorder # of communication symptoms	3.65	3.78	3.14	3.27
Autistic Disorder # of restricted repertoire symptoms	2.42	2.58	2.90	3.06
Expressive One-Word Picture Vocabulary Test	83.77	84.63	64.08	66.15
Peabody Picture Vocabulary Test	70.11	71.29	48.30	49.03
Stanford-Binet Comprehension (ave.=50)	40.05	40.25	34.26	35.46
Stanford-Binet Vocabulary (ave.=50)	43.04	43.41	38.20	41.10

Table 11. Preschool and School-Age Scores.

Measure	Group A				Group B			
	Preschool		School age		Preschool		School age	
	<i>M</i>	(<i>SD</i>)	<i>M</i>	(<i>SD</i>)	<i>M</i>	(<i>SD</i>)	<i>M</i>	(<i>SD</i>)
Vineland Commun. ^f	74.46	16.08	76.10	23.75	47.66	9.78	40.92 ^c	13.40
Vineland Daily Living	68.19	11.98	69.91 ^a	21.56	45.92	12.57	33.15 ^c	15.56
Vineland Socialization	68.33	9.99	71.55	16.72	53.95	6.17	50.54 ^b	10.59
S-B ^g Vocab. (<i>M</i> = 50)	42.94	6.45	44.12	8.87	40.18	11.63	32.00 ^d	8.25
S-B Comprehension (<i>M</i> = 50)	39.70	6.86	41.70 ^a	9.23	35.58	7.69	28.75 ^d	7.28
S-B Verbal Reasoning (<i>M</i> = 100)	80.73	13.67	84.43 ^a	19.75	66.19	18.28	50.94 ^c	12.88
S-B Sentence Memory (<i>M</i> = 50)	40.87	6.83	43.16 ^b	9.16	34.06	8.99	30.82 ^a	7.72
S-B Abstract Visual Reasoning (<i>M</i> = 100)	93.60	21.64	95.06	20.47	66.33	14.30	65.54	17.48
Peabody Picture Vocabulary Test	74.10	16.55	76.21	23.89	66.22	14.23	45.97 ^c	14.43
Expressive One-Word Picture Vocabulary Test	87.77	18.30	97.15 ^c	23.58	76.94	18.20	67.08 ^c	22.60

Note. ^a paired *T* test between preschool and school age $p < .1$, ^b $p < .05$, ^c $p < .02$, ^d $p < .01$, ^e $p < .001$.

^f Vineland scores are standard scores. ^g S-B = Stanford-Binet (4th ed.).

A versus B was treated as the criterion dependent measure. Test variables were selected for clinical interpretability and suspected relationship with subgroup membership. These variables were as follows: S-B Abstract Visual Reasoning standard score; nonverbal ratio IQ; Vineland Socialization Domain standard score; Wing's typology; age; Social Abnormalities I score; Social Abnormalities II score; sum of items from section I of the Wing Autism Diagnostic Checklist, and sum of items across all

sections of the Wing Autism Diagnostic Checklist.

Each variable was examined alone by linear regression to determine whether it was significantly related to the classification measure (A vs. B). Tests of linearity were performed to ensure that there were no nonlinear relationships between the classification variables and the test variables. Each variable was then run in a logistic regression to confirm its relationship with the classification variable, by examining its predic-

Table 12. Characteristics of Children Diagnosed With PDD by Clinical But Not Empirical Criteria.

Variable	<i>M</i>	(<i>SD</i>)
No. of items checked on WADIC Social Domain	4.25	(1.81)
No. of items checked on WADIC Communication Domain	2.33	(1.67)
No. of items checked on WADIC restricted repertoire domain	1.25	(1.21)
Stanford-Binet Abstract Visual	86.92	(17.62)
S-B Vocabulary (ave. = 50)	43.89	(7.32)
Peabody Picture Vocabulary Test	79.73	(21.06)
Expressive One-Word Picture Vocabulary Test	95.04	(22.46)
Vineland Communication	80.33	(17.45)
Vineland Socialization	79.42	(12.89)

Note. $n = 12$.

tive ability. (All regressions for these analyses were done with SPSS.)

Linear regression analyses indicated that all of the variables selected were significantly related to the classification measure ($p < .002$ or less). Individual logistic regression analyses indicated that all variables except the sum across all sections of the Wing Autism Diagnostic Checklist significantly predicted classification. The two variables that functioned best as single indicators were the Vineland Socialization standard score and nonverbal ratio IQ. The Vineland Socialization standard score alone classified the children with 86% accuracy, with a score of 60.3 optimally dividing the A and B groups. The nonverbal ratio IQ alone classified the children with 81% accuracy, with a score of 65.3 optimally dividing the A and B groups.

A stepwise logistic regression was then performed on a model containing all of the test variables that significantly predicted classification. Both Wald and likelihood-ratio chi squares were used to determine the significance of additional variance explained by each added variable.

A four-variable solution yielded an overall correct classification rate of 96%, with a sensitivity of 97% (probability of correctly classifying a child who belonged in Group B) and a specificity of 94.5% (probability of correctly classifying a child who belonged in Group A). Conversely, it had a positive predictive value of 96% (probability that a child classified in Group B was correctly classified) and a negative predictive value of 96% (probability that a child classified in Group A was correctly classified). The variables in this solution were Vineland Socialization standard score, nonverbal ratio IQ, sum of Social Abnormalities I, and age. In this model, the probability of a child with a diagnosis of PDD belonging to Group B (the lower functioning group) is shown by the following two formulas:

$$p = 1/(1 + e^{-x}),$$

where $x = 17.08 - (.34 \times \text{Vineland Socialization standard score}) - (.14 \times \text{nonverbal ratio IQ}) +$

$(.52 \times \text{Social Abnormalities I}) + .11$ (age in months).

In order to arrive at an algorithm that would have maximum clinical utility, models were derived for each pair of variables. The model using the Vineland Socialization standard score together with the Social Abnormalities I score yielded an adequate classification accuracy of 92%. For this model, sensitivity = 92%, specificity = 93%, positive predictive value = 91%, and negative predictive value = 94%.

The formula for this model is: $x = 15.64 - (.35 \times \text{Vineland Socialization standard score}) + (.45 \times \text{Social Abnormalities I})$.

Because only two variables are involved, we were able to derive a table for joint values of the two variables, with all possible values of Social Abnormalities I and Vineland scores by increments of 5, ranging from 35 to 85 (see Table 13). Above and below these Vineland scores, variation in Social Abnormalities I makes no measurable difference, as can be seen from Table 13. It can be seen that probabilities of assignment to Group B vary with Vineland score, with the cut point depending on the degree of autistic symptomatology. For a low degree of autistic symptomatology, the Vineland cut point score between Groups A and B is around 45–50; for intermediate degrees of autistic symptomatology, the cut point is a Vineland score of about 55–60, and for very high degrees of autistic symptomatology, the Vineland cut point is around 70. Thus, the values in this table can be used, with 92% accuracy, to calculate the probability of a child with clinically diagnosed PDD belonging to Group B (with 1 minus this value representing the probability that the child belongs to Group A). If a more rigorous classification is needed, the four-variable model described above can be used.

DISCUSSION

Summary of Results

Statistical analysis confirmed the existence of two distinct groups within the PDD spectrum. Group A contains the higher functioning chil-

Table 13. Percent Probability of Membership in Group B.

Social Abnormal. I	Vineland Socialization Standard Score										
	35	40	45	50	55	60	65	70	75	80	85
0	97.68	88.49	58.42	20.42	4.48	0.85	0.16	0.03	0.01	0.00	0.00
1	98.51	92.34	68.78	28.70	6.85	1.33	0.24	0.04	0.01	0.00	0.00
2	99.04	94.98	77.56	38.70	10.34	2.06	0.38	0.07	0.01	0.00	0.00
3	99.39	96.74	84.42	49.75	15.32	3.20	0.60	0.11	0.02	0.00	0.00
4	99.61	97.90	89.47	60.83	22.10	4.93	0.94	0.17	0.03	0.01	0.00
5	99.75	98.65	93.02	70.89	30.79	7.52	1.46	0.27	0.05	0.01	0.00
6	99.84	99.13	95.43	79.25	41.10	11.30	2.28	0.42	0.08	0.01	0.00
7	99.90	99.45	97.04	85.69	52.25	16.66	3.52	0.66	0.12	0.02	0.00
8	99.94	99.65	98.09	90.38	63.18	23.87	5.42	1.04	0.19	0.03	0.01
9	99.96	99.77	98.78	93.64	72.91	32.96	8.24	1.61	0.30	0.05	0.01
10	99.97	99.86	99.22	95.85	80.85	43.54	12.35	2.51	0.47	0.09	0.02
11	99.98	99.91	99.50	97.31	86.88	54.74	18.09	3.88	0.73	0.13	0.02
12	99.99	99.94	99.68	98.27	91.21	65.48	25.73	5.95	1.14	0.21	0.04
13	99.99	99.96	99.80	98.89	94.21	74.84	35.21	9.03	1.78	0.33	0.06
14	100.00	99.98	99.87	99.29	96.23	82.35	46.01	13.47	2.77	0.52	0.09
15	100.00	99.98	99.92	99.55	97.56	87.97	57.20	19.62	4.27	0.81	0.15
16	100.00	99.99	99.95	99.71	98.43	91.98	67.70	27.69	6.54	1.26	0.23
17	100.00	99.99	99.97	99.81	98.99	94.73	76.67	37.52	9.89	1.96	0.36
18	100.00	100.00	99.98	99.88	99.36	96.58	83.75	48.50	14.68	3.05	0.57
19	100.00	100.00	99.99	99.92	99.59	97.79	88.99	59.63	21.25	4.70	0.89
20	100.00	100.00	99.99	99.95	99.74	98.58	92.69	69.85	29.73	7.18	1.39
21	100.00	100.00	99.99	99.97	99.83	99.09	95.21	78.41	39.89	10.81	2.17
22	100.00	100.00	100.00	99.98	99.89	99.42	96.89	85.07	51.00	15.98	3.36

Note. PDD=Pervasive Developmental Disorder; DLD=Developmental Language Disorder; LoIQ=Nonautistic Low IQ.

dren with PDD, the misdiagnosed children with DLD, and half of the misdiagnosed children with LoIQ. Group B contains the lower functioning children with PDD, and half of the misdiagnosed children with LoIQ. The best (although not perfect) single score for distinguishing children in Group A versus B is a nonverbal IQ of 65.3 or a Vineland Socialization standard score of 60.3.

It should be emphasized that all measures on which the children in Groups A and B differ (such as verbal IQ, nonverbal IQ, severity of symptoms, etc.) are continua, with scores ranging from low to high in children across both groups. The Golden statistical method, however, suggests that the relationships among these continuous variables are different within these two groups, and that therefore, the groups are dis-

continuous in the sense that they are drawn from different underlying populations.

Most children in both groups showed all of the major symptoms on the Wing (1985) checklist (e.g., abnormal greeting, impaired peer relationships), especially in the social and communication domains. Symptoms in the restricted repertoire domain were somewhat less frequent; within that domain, preoccupations were common, but resistance to change was relatively infrequent. Within each major symptom group, however, the groups differed on specific items; children in Group B showed more developmentally lower or more severe manifestations of the symptoms (e.g., no peer relationships, no spontaneous waving, mutism), whereas children in Group A showed developmentally higher or less severe manifestations (e.g., naive attempts at

peer relationships, greets with stereotyped phrases, one-sided conversations).

Parent report indicates early differences in behavior, with children in Group B showing more severe remoteness, impaired play, repetitive gestures and activities, and abnormalities such as pica. Again, no item related to resistance to change differentiated the groups. Teacher reports confirm that all developmental acquisitions are higher in Group A. Also discriminating are aspects of nonverbal communication, symbolic play, and peer relationships, as well as Wing's social typology. Items that seem more directly affiliative, such as eye contact and spontaneous affection did not differentiate the groups, nor did specific language abnormalities such as echolalia. Standardized test scores also show a superiority for children in Group A.

Subgroups were validated by good concordance with a cluster analysis solution and by differing developmental trajectories into school age. Group membership was generally stable into school age, but the children in Group A showed a much greater probability of appearing nonPDD by school age. School-age behavioral scores generally suggest that children in Group A improve in social interaction and communication, and that most improve in restricted repertoire and a few get substantially worse. Children in Group B tend to remain relatively stable in number of symptoms in each domain, or show a small improvement. Similarly, standardized testing at school age suggests that children in Group A remain stable or show small improvements in scores, whereas children in Group B show small but consistent and significant declines in cognitive functioning relative to age peers.

Several variables related to cognitive and social developmental level and social abnormalities successfully predicted membership in Groups A or B. Vineland Socialization standard score, nonverbal ratio IQ, score on Social Abnormalities I, and age together predicted subgroup membership with 96% accuracy. Vineland Socialization and Social Abnormalities together predicted group membership with 92% accuracy. It is noteworthy that although the Wing typology alone did not predict sub-

group membership with good accuracy, the items on Social Abnormalities I largely reflect the classic autistic symptoms characteristic of children described by Wing's aloof type. Thus, both development of social skills and presence of social abnormalities, in combination, are important for subgrouping the children, and (as suggested by Barth, Fein, & Waterhouse, 1995) make partly independent contributions to the clinical picture.

It is interesting to speculate on how the groups would have been different if the final set of measures had included more language measures. Perhaps children with PDD with relatively intact language, such as those with Asperger's Disorder, would have been placed in the NonPDD group. However, many verbal items were included in the Parent, Teacher, and Neuro scales that were originally used to form the groups, and the groups differed significantly on many behavioral and test measures of language. The composite scales were not used in the final classificatory algorithm, because they were from derived scales that would be difficult for others to use, and did not prove to be as powerful discriminators as the final set of variables (Vineland, Nonverbal IQ, Social Abnormalities). Furthermore, if one examines the verbal standard scores for Group A, most are at or near the normal range; therefore, individuals with Asperger's Disorder, with social deficiencies and perseverations but good language might well fit into Group A.

Relationship to Previous Findings

The present study strongly confirms the suggestion by Cohen, Paul, and Volkmar (1987), and by Tsai (1992) that high- and low-functioning autism be considered as potentially separate diagnostic entities. Not only did the larger study find that children divided a priori by cognitive level (IQ of 80) differ in many other ways (summarized in Rapin, 1996), but the current results indicate that empirical analysis of the PDD sample found strong evidence of an underlying dichotomy between high- and low-functioning children (termed Groups A and B).

Also consistent with previous findings (summarized by Lord & Venter, 1992), we found that the high-functioning children tend to improve over time in both cognition and behavior, although there is a suggestion that a small subgroup may become more perseverative and preoccupied at school age. The lower functioning children, in contrast, tend to remain relatively stable behaviorally, but decline in relative cognitive function.

Children in Groups A and B shared many impairments and differ primarily in ways accountable for by their cognitive levels. In addition to scores on standardized tests, they differ in the specific manifestations of behavioral abnormalities. For example, whereas almost all children in both groups have impaired peer relationships, the children in Group B tend to ignore peers, and the children in Group A have inappropriate and unsuccessful relationships. This pattern of difference by developmental level or severity could be seen in every symptom area.

Present data also allow preliminary evaluation of Tsai's (1992) suggested criteria for high-functioning autism. Our findings are in agreement with the outlined criteria, but suggest that the proposed nonverbal IQ score of 70 should be reduced to 65, and that the proposed criteria for language and social functioning should be broadened to include children with somewhat greater impairments.

CONCLUSIONS

In addition to these major findings, results also suggest that stereotypies and preoccupations, although frequent in children diagnosed with PDD, may be less central or universal than are impairments in social interaction, communication, and play. Resistance to change, in particular, occurs with such low frequency as to challenge its appropriateness as a diagnostic criterion. Sparing of written language and number concepts in the children with PDD were notable.

Results also suggest that most children diagnosed with PDD-NOS (although not as high as

children with AD) do belong on the PDD spectrum and are statistically placed within Groups A or B. The division into Groups A and B shows some concordance with current psychiatric diagnostic systems, especially with DSM-IV (see companion paper by Waterhouse et al., 1996).

This empirical subclassification of PDD into two types, of course, needs to be replicated and validated. Use of the same statistical methodology with other PDD samples will allow replication to be attempted. In addition, following samples of children into adolescence or adulthood would provide further validation of different developmental trajectories of the groups.

If the findings are confirmed by replication and follow-up, the implication would be that the two PDD groups found may represent distinct disorders. If future research confirms this discontinuity, then collapsing across groups for the purpose of analyzing data concerning biological markers or the effectiveness of treatment might well obscure significant findings. This would suggest that future research into etiology, pathophysiology, course, symptomatology, treatment, and prognosis should divide sample of children with PDD, perhaps according to one of the algorithms presented, and analyze findings for the groups separately. Contrary to what some might suggest, there is no implication that children in either group deserve anything less than the best special education, but the best may not be the same for everyone; groups of children need to be clearly defined if we are able to determine what the best approach is likely to be for any given child, and if outcome research is to be enlightening.

The data do not allow conclusions on whether the subgroups represent an additive or interactive set of deficits (in which case the Group B children would have Group A deficits plus others), or whether the groups represent separate and distinct disorders, each affecting brain systems contributing crucially to social development. Future research on brain system abnormalities in the the two types of children will clarify this issue.

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