

ASPERGER SYNDROME

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What's So Special about Asperger Syndrome? *The Need for Further Exploration of the Borderlands of Autism*

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Readers may be surprised to find that this volume contains a contribution by an expert on childhood language disorders, as according to the fourth edition of the *Diagnostic and Statistical Manual of Mental Disorders* (DSM-IV; American Psychiatric Association, 1994) definition of Asperger syndrome (AS) the diagnosis requires that there is no clinically significant language delay. The aim of this chapter is to provide a counterbalance to the weight of research that is concerned with whether there is a continuum between AS and autistic disorder. I argue that, by concentrating on these two conditions, researchers have created the impression that there is a single continuum, with autistic disorder at one end and AS at the other. This impression has led many people to suppose that AS is the appropriate diagnosis for any child who falls within the autistic spectrum, is of normal intelligence, but who does not meet full criteria for autistic disorder. In this chapter, I present evidence against this view. My argument is that there are many children whose deficits resemble mild forms of autism but who do *not* have the constellation of features characterizing AS. Some of these children occupy a position intermediate between autistic disorder and developmental language disorder. The diagnostic boundaries between pervasive and specific developmental disorders are, according to this view, much less clear-cut than the textbooks seem to imply.

A second point is that the judgment of whether or not a child has nor-

mal language development depends critically on which aspects of language we consider. In particular, there are children whose ability to use language appropriately in communicative contexts is extremely poor, despite good or even superior knowledge of grammar and phonology (speech sounds). Questions about the relationship between language disorder and autistic spectrum disorders have been clouded by a failure to draw a distinction between formal knowledge of language structure and ability to use language to communicate effectively.

The structure of the chapter is as follows. First, I review the way in which autistic disorder and specific language impairment (SLI) have been conceptualized over the past 18 years. I then consider the evidence for children who fall between conventional diagnostic boundaries, moving on to focus specifically on children whose symptom profile overlaps with both autism and SLI. The category of "semantic-pragmatic disorder" has been proposed for such cases but has been difficult to validate because we lack suitable measures to quantify the relevant symptoms. I present some checklist data from a pilot study that was designed to tackle this issue. These checklist data confirm that there are significant numbers of language-impaired children who do have disproportionate difficulty with language content and use, and that they tend to have associated autistic features. However, there is substantial variation in symptom profile from child to child, and such children do not typically have distinctive semantic impairments. It is proposed that rather than subsuming such children under the umbrella category of pervasive developmental disorder not otherwise specified (PDD-NOS), they should be referred to as cases of pragmatic language impairment. It should be recognized that these children have some symptomatic overlap with *both* SLI and autistic disorder, rather than forcing them into one category or the other. Finally, I conclude by pointing out that the emphasis on differential diagnosis between AS and autistic disorder has diverted attention from other conditions which may be regarded as falling in the autistic spectrum, and which are clinically important but have attracted little research attention.

RELATIONSHIP BETWEEN LANGUAGE DISORDER AND AUTISTIC SPECTRUM: EVOLVING DIAGNOSTIC CONCEPTS IN DSM

DSM-III (American Psychiatric Association, 1980) drew a theoretical distinction between the *specific* developmental disorders and *pervasive* developmental disorders as follows:

Pervasive Developmental Disorders differ from the Specific Developmental Disorders in two basic ways. First, only a single specific function is af-

fected in each Specific Developmental Disorder, whereas in Pervasive Developmental Disorders multiple functions are always affected. Second, in Specific Developmental Disorder the children behave as if they are passing through an earlier normal developmental stage, because the disturbance is a *delay* in development, whereas children with Pervasive Developmental Disorders display severe qualitative abnormalities that are not normal for any stage of development, because the disturbance is a *distortion* in development. (p. 86).

At the time that DSM-III appeared, most children who were thought to merit a diagnosis of pervasive developmental disorder (PDD) were cases of infantile autism, plus a smattering of cases with rarer forms where there was onset in childhood or an atypical presentation. Developmental language disorder was regarded as a prototypical *specific* developmental disorder (see Figure 9.1). An early study by Bartak, Rutter, and Cox (1975), comparing children with autism and children with receptive developmental language disorder (then termed "developmental dysphasia"), offered some validation of this view: Children in the autistic group had a much broader range of impairments, affecting nonverbal as well as verbal communication, and they had a number of features that did not seem to correspond to any normal stage of development, and so could reasonably be described as distortions (see Figure 9.2).

A problem with the DSM-III scheme was that clinicians soon found that there were many children whose pattern of impairments did not correspond to clear-cut cases of either autism or developmental language disorder. In

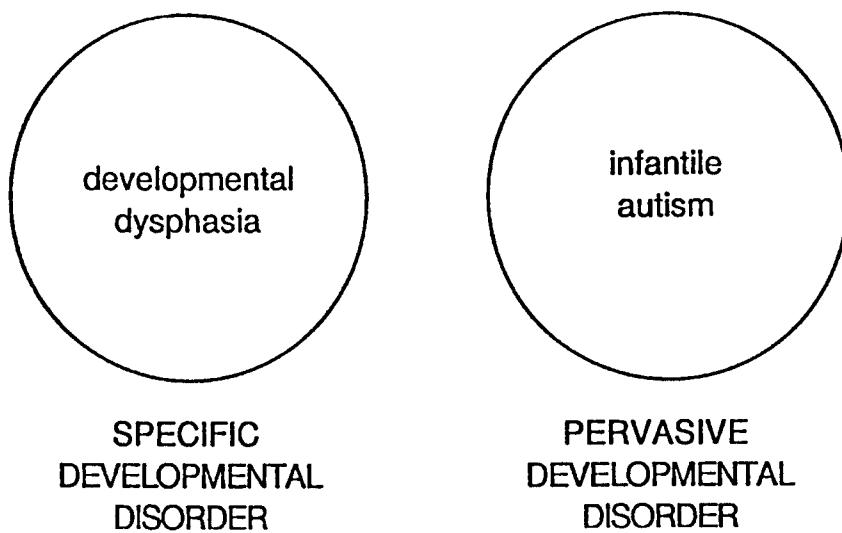


FIGURE 9.1. Diagnostic distinction between specific and pervasive developmental disorders as specified in DSM-III.

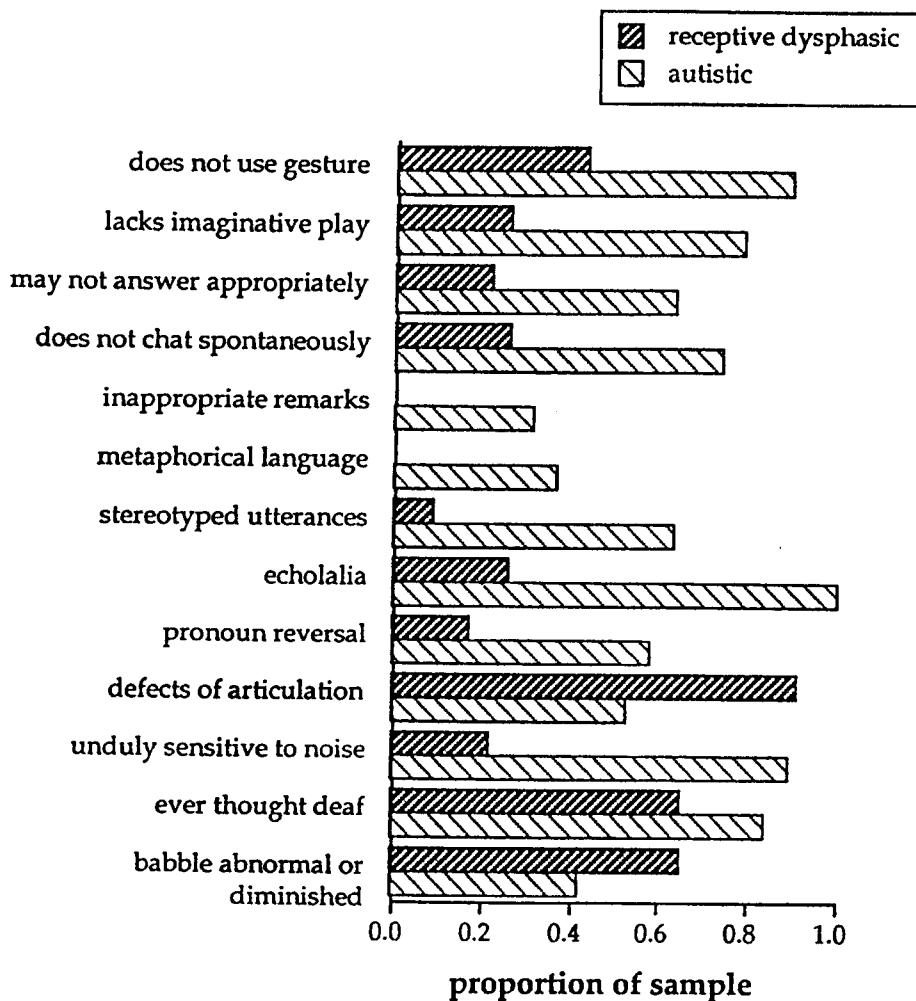


FIGURE 9.2. Summary of findings from Bartak et al. (1975), comparing 19 autistic children with 23 children with developmental receptive dysphasia. Most characteristics were scored positive if the child had ever shown that behavior, regardless of whether it was still present at the time of assessment.

1981, Wing published a paper drawing attention to Asperger's descriptions of children who appeared to have no difficulty in learning the intricacies of language but who had unusual interests and impairments of social behavior. Most clinicians welcomed the term "AS," which provided a diagnostic label for a group of children who had hitherto been difficult to categorize.

The limitations of the DSM-III scheme were addressed to some extent in later revisions, so that by DSM-IV, three modifications were in place:

1. The diagnostic criteria for what was now referred to as "autistic disorder" were refined. The language impairments of autistic children

were described in more detail, stressing that the abnormalities affected both verbal and nonverbal communication.

2. The category of AS was added (with diagnostic criteria as described in Chapter 2, this volume). Although the diagnostic criteria specify that "there are no clinically significant delays in language," this is defined in relation to mastery of language milestones and in terms of sentence length and complexity rather than to how language is used.
3. The category of PDD-NOS was added to provide a diagnosis for cases in which there is a "severe and pervasive impairment in the development of reciprocal social interaction, verbal and nonverbal communication skills or the development of stereotyped behaviour, interests and activities," including cases where there is late age at onset, or "atypical" or "subthreshold" symptomatology (pp. 77-78).

Figure 9.3 illustrates this framework.

A categorical diagnostic label implies that a cluster of symptoms tends to co-occur, and that individuals in the particular part of three-dimensional space corresponding to that cluster will be more numerous than those in surrounding regions. This is reflected in DSM-IV, where PDD-NOS merits eight lines of description, compared with just over two pages for AS, and five and a half pages for autistic disorder. The message is clear: PDD-NOS is thought of as a default diagnosis, to be applied only to rare children for whom other diagnoses are inappropriate. Accordingly, PDD-NOS is not a focus of clinical or research interest, and there is little published information about the nature, correlates, prognosis, and treatment of children with this diagnosis.

HOW COMMON ARE CHILDREN WHO FALL BETWEEN DIAGNOSTIC BOUNDARIES?

The existence of children who fall in the unlabeled areas of Figure 9.3 cannot be doubted, but it is difficult to establish is how rare they are. Quite simply, children who do not meet diagnostic criteria for autistic disorder, AS, or SLI tend not to be included in research studies. However, Klin (personal communication, 1996) reported that a diagnosis of PDD-NOS was *more* common than either AS or autism in children referred to the Yale Child Study Center.

Further evidence for "intermediate" or "subclinical" cases comes from a study of relatives of individuals affected with autistic disorder. Bolton et al. (1994) used a standardized interview to assess functioning in the domains of language, social interaction, and stereotyped behavior and reported that although few relatives met criteria for autistic disorder, many of them had impairments in just one or two of these domains.

Turning the question on its head, we may also consider how common

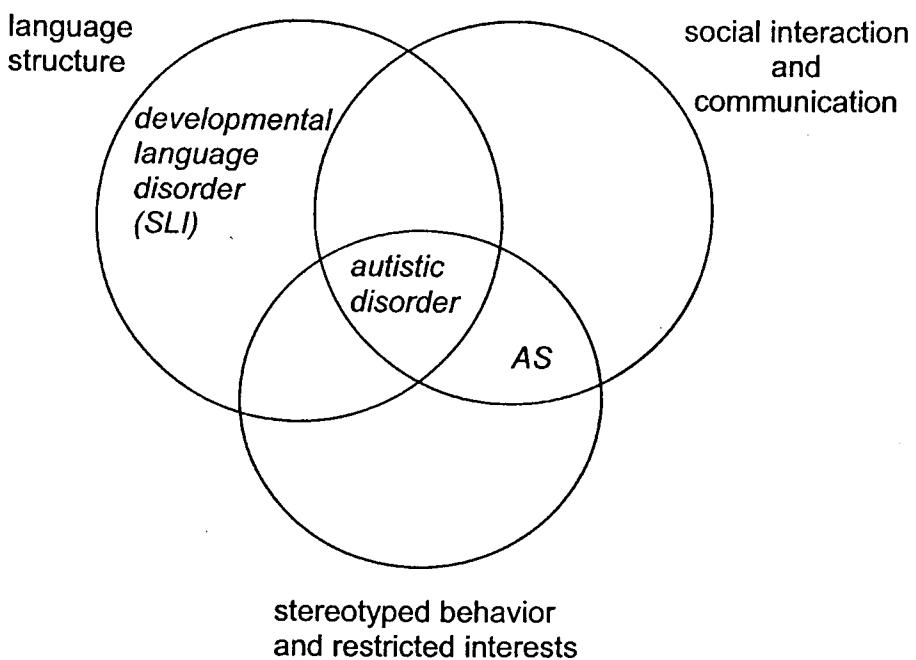


FIGURE 9.3. Set diagram representation of the diagnostic possibilities offered by DSM-IV. Each set shows impairment in one domain of functioning: "Language structure" refers to mastery of grammar and speech sounds, as reflected in age at passing language milestones, and complexity and clarity of spoken language. "Social interaction and communication" refers to use of language and nonverbal means to communicate with others. "Stereotyped behavior and restricted interests" refers to diversity of interests and creativity as well as repetitive behaviors. The default category PDD-NOS would be applied to children who had impairments in one or two of the three areas of functioning but who fell outside the boundaries of one of the other categories (i.e., those areas that are unlabeled in the figure).

are social impairments in children with language difficulties. It is commonly assumed that any social limitations in language-impaired children are just a secondary consequence of their communicative problems (see Bishop, 1997, for a review). However, it is difficult to reconcile this view with work by Paul, Spangle-Looney, and Dahm (1991), who reported that in a sample of children with expressive language delay at 2 years of age, 48% were impaired on the socialization scale of the Vineland Adaptive Behavior Scales at 3 years of age, even when items with a verbal component were excluded from consideration. Receptive language delay in 3-year-olds was virtually *always* accompanied by socialization problems.

Taken together, these pieces of evidence indicate that there is no necessary association between the three domains of impairment that characterize autism: They can be dissociated, especially in higher-functioning individuals. "Subthreshold symptomatology," or impairments affecting only two of the three domains shown in Figure 9.3, are not uncommon. Many

children whose predominant presenting problems are with spoken language are found to have broader limitations of socialization when properly assessed. In practice, such individuals may be labeled PDD-NOS, or, if their language problems are particularly striking, they may be included as cases of developmental language disorder. My own studies suggest that the latter situation commonly arises, and that the population of children diagnosed as having developmental language disorder contains a substantial minority who have abnormalities in the use of nonverbal as well as verbal communication, and some who have restricted or peculiar interests similar to those in AS.

HOW SHARP IS THE BOUNDARY BETWEEN AUTISM AND DEVELOPMENTAL LANGUAGE DISORDER?

The pioneering study by Bartak et al. (1975), comparing cases of infantile autism and developmental receptive dysphasia, is typically cited as evidence for a qualitative distinction between these two disorders. However, this study did include five cases which, at the outset, were recognized as having both a language disorder and some autistic features: These cases were termed the "mixed group." Furthermore, as the children have been followed up into adolescence and adulthood, clearer evidence has emerged of more pervasive impairments of social interaction and abnormal interests in some of the original dysphasic group (Mawhood, 1995), and the separation of the two groups by a discriminant function analysis has proved much less efficient. The following vignettes from Mawhood illustrate that individuals initially classified as cases of developmental receptive dysphasia had abnormalities in the areas of social behavior and interests in adulthood, although these typically were far less severe than those seen in adults with autism.

(participant DLD8, age 22 years)

In all aspects of self-care he was entirely independent and he could use a telephone and manage his finances himself. Most of his spare time was spent pursuing his preoccupation with trains and his less intense interest in CB radio. He spent a lot of time hanging around railway stations, going on train rides when he could afford it, and looking at train magazines. His social overtures were somewhat limited and he would speak if spoken to but would not make the first move. There were two friends that he visited regularly; one shared his interest in trains, the other was interested in CB radio. These relationships were clearly selective, did involve some apparent pleasure in each other's company, and some sharing of confidences, but there was still nonetheless a slightly odd quality to them because of their restricted range of interests. He did not appear to be lonely. (p. 384)

(participant DLD9, age 25 years)

[At 9 years of age] the quality of his peer relationships was said to be slightly abnormal. He showed some evidence of ritualistic behaviour and marked quasi-obsessive activities and attachments to odd objects. When followed-up . . . he was living independently in a flat belonging to an organisation that specialised in providing accommodation for disabled people. . . . He looked after himself entirely independently. . . . He was able to cope with most things provided he had come across them before but had difficulty with the unexpected, e.g., his cooker started to smoke and he didn't know what to do. . . . He did voluntary work cleaning a local monument twice a week, but other than this filled his time with doing his domestic chores, weight training at a local health club and looking after his extensive collection of model aeroplanes. He was very interested in this collection, but not really preoccupied. . . . His speech was now fairly well developed and his grammar was largely correct with occasional errors. . . . He could follow a fairly simple plot but would get lost if the story became too complex. If he gave an account of events he would pay attention to minute detail, and he could converse well although he would tend to talk about things he was interested in. . . . He could make acquaintances but sometimes started conversations in slightly inappropriate ways, e.g., by talking about the *Encyclopedia Britannica* and saying "Did you know that . . . ?" He did not appear to want any close friends although he met up with the curate at his church every couple of weeks for a chat and a meal, and seemed to enjoy his company. . . . He showed some understanding of how other people were feeling and his mother said he would show appropriate concern if someone was not feeling happy. Girls were of absolutely no interest to him. He could only see the difficulties involved rather than any of the positive emotional aspects and said "girls are just trouble." (pp. 385-386)

These cases suggest that far from being separate conditions, receptive language disorder and autism might lie on a single continuum. Yet this view is radically different from conventional wisdom as embodied in our textbooks and depicted in Figure 9.1. How are we to resolve this paradox?

THE CONCEPT OF A "SEMANTIC-PRAGMATIC" SUBTYPE OF DEVELOPMENTAL LANGUAGE DISORDER

Light was thrown on this question with the publication of a nosology of developmental dysphasia by Rapin and Allen (1983). Rather than adopting traditional distinctions between "expressive" and "receptive" forms of language disorder, Rapin and Allen drew attention to the different ways in which expressive and receptive language could be impaired. Of particular interest was their delineation of a category of "semantic-pragmatic deficit

syndrome," in which children had relatively good mastery of language form (i.e., grammar and phonology) but abnormal content and poor use of language. Rapin (1982) gave the following account of semantic-pragmatic syndrome¹:

Children with this syndrome have no difficulty decoding phonology or producing well formed sentences. Their deficit affects comprehension and use of language. They have trouble understanding discourse. . . . The children usually have an intact or superior auditory memory and are fluent. They may repeat whole sentences verbatim or recite TV commercials. While they often have no difficulty retrieving verbal labels for objects or pictures, they nonetheless have an anomia in spontaneous speech. As a result, some of their words miss the mark despite the fact that they belong to the appropriate semantic field. This gives their speech a loose, tangential, or somewhat inappropriate quality. Their train of thought appears illogical and difficult to follow. . . . The children's comprehension deficit is likely to be overlooked or underestimated because their spontaneous speech is so fluent and because they understand single words and simple phrases and are sociable. (p. 145)

In their nosology, Rapin and Allen were not concerned with differential diagnosis from autism or AS: They regarded the characterization of a child's language disorder as orthogonal to a psychiatric diagnosis, and they noted that semantic-pragmatic disorder is commonly seen in children with autism (although other language profiles are also found). However, importantly, they noted that the same type of language difficulties could be observed in children who did not have autism, a point reiterated in a recent review by Rapin (1996).

In a study of *nonautistic* children receiving special educational facilities for those with specific developmental language disorders, Bishop and Adams (1989) described a number of distinctive communicative characteristics of children who met the clinical picture of semantic-pragmatic disorder (see Table 9.1). Similarities with observations of communicative behavior in children with AS are evident, but the children studied by Bishop and Adams had severe delays in passing early language milestones as well.

Bishop and Rosenbloom (1987) noted that children who are diagnosed as having semantic-pragmatic disorder seemed to differ qualitatively from other language-impaired children: They tended to have associated social and behavioral abnormalities similar to those seen in autism. These similarities have led some authors to argue that semantic-pragmatic disorder

¹Rapin and Allen (1987) refer to "semantic-pragmatic deficit syndrome," and Bishop and Rosenbloom (1987) to "semantic-pragmatic disorder." The latter term is used henceforth in this chapter.

TABLE 9.1. Conversational Characteristics of Language-Impaired Children with Semantic-Pragmatic Disorder

The following examples of conversational extracts are taken from children ages 8 to 12 years who fit the clinical picture of semantic-pragmatic disorder (Bishop & Adams, 1989). "C" denotes child, "A" denotes adult. Categories marked "*" are those in which children with semantic-pragmatic disorder differed not just from age-matched controls but also from younger control children aged 4 to 5 years.

Expressive problems in semantics/syntax*

Example 1

- C: We went on a bus because Lee was sick out of the window. [Use of term that is overspecific in meaning, e.g., "because" or "but" when "and" is required]

Example 2

- A: Why did you have to go to the doctor?
C: I used to have a headache. [Wrong verb tense/aspect]

Failure of literal comprehension

Example 3

- A: Where did you go on holiday?
C: In September.

Failure to use context in comprehension (overliteral interpretation)

Example 4

- A: (after long session of work) Can you stand to do some more?
C: (Stands up.)

Example 5

- A: Are there any other times when you have parties?
C: No.
A: What about at Christmas?
C: It snows.

Too little information*

Example 6

- C: (talking about a jeweler's shop) when you take the ring off it ones both of them are crowns. [Unestablished referent: unclear what "them" refers to]

Example 7

- C: My brother was feeling sick on Monday.
A: Right.
C: And I took my trouser off.
A: Uhuh. Why did you take your trousers off?
C: He was sick on my trouser. [Logical step omitted, critical information provided only when A asked for clarification]

Too much information*

Example 8

- A: Where have you been on a boat?
C: Where have I been? Haven't sailed a cruiser, you know. [Unnecessary denial of something that A had not assumed]

Example 9

- A: Is that a good place to break down? (referring to photograph of stranded motorist)
C: The answer whether it's a good place to break down is no, because if see if anybody broke down, cos there's no telephone to telephone, there's no telephone for the breakdown. [Excessive elaboration]

(continued)

TABLE 9.1. (cont.)

<u>Unusual content*</u>
<i>Example 10</i>
A: What's going on there? (<i>referring to photo of birthday party</i>)
C: It's someone's birthday. Something could be dangerous, you know, like a fire from the candles. [Topic drift: child connects to previous utterances but steers conversation to a favored topic]
<i>Example 11</i>
A: Have you ever been to the doctor?
C: I had an apple a day. [Stereotyped language: no evidence that child was attempting to be humorous]
<i>Example 12</i>
C: Do you like candyfloss?
A: No.
C: Do you hate it?
A: I think it's all horrible and sticky.
C: Why? [Remorseless questioning by child]

is just another term for autism (Lister Brook & Bowler, 1992). There are two ways of interpreting this. It could mean that semantic-pragmatic disorder is simply a more acceptable label for children who actually would meet criteria for autistic disorder according to DSM-IV or *International Classification of Diseases* (World Health Organization, 1992). To accept this conclusion, not only would one have to assume a high rate of misdiagnosis among children attending special schools for those with language impairment, but one would also be left with the problem of how to classify the "intermediate" cases described in research studies that applied diagnostic criteria very carefully, such as that by Bartak et al. (1975) and Bolton et al. (1994).

A more reasonable hypothesis is that there are continuities between autistic disorder and semantic-pragmatic disorder, either in terms of underlying causes or in terms of symptomatology (Bishop, 1989). If this is the case, then semantic-pragmatic disorder would belong more properly in the autistic spectrum, corresponding to a subtype of PDD-NOS, rather than be seen as a subtype of specific developmental disorder. Tantalizing evidence suggesting etiological similarities comes from Woodhouse et al. (1996), who found that children with semantic-pragmatic disorder, like those with autism, had unusually large heads, whereas those with other kinds of developmental language disorder did not. In my own studies, I have been interested in trying to devise better methods for documenting the communicative and related impairments that characterize semantic-pragmatic disorder in order to assess the validity of the concept and to provide a means for comparing and contrasting children with this diagnosis and those with unambiguous diagnoses of autism or AS.

THE CHECKLIST FOR LANGUAGE-IMPAIRED CHILDREN: A PILOT STUDY²

A major difficulty confronting anyone wishing to study subtypes of language disorder is that we lack suitable instruments for the objective assessment of the behaviors that characterize semantic-pragmatic disorder. Most language tests measure complexity of expressive language form, verbal memory, or comprehension of vocabulary or sentences of increasing length and complexity. The impairments that are seen as typifying semantic-pragmatic disorder, such as verbosity, overliteral responding to questions, or problems in understanding connected discourse, are not identified on such measures. These behaviors are not only difficult to assess using contemporary measures but probably also more variable than other aspects of language behavior. Thus a child who appears "verbose" in some situations might be silent in others. For these reasons, it seemed worthwhile developing a checklist to obtain ratings from people who know the child well and have therefore had an opportunity to observe the child's behavior over time and in a range of situations. The disadvantages of ratings are well-known: Raters may differ in how they interpret questions, and they may use different criteria for deciding that a behavior applies. For this reason, a rating scale is only useful if one can demonstrate reasonable agreement between two independent raters. The study described here represents a preliminary attempt to devise a rating scale that would provide data that were both reliable and valid in distinguishing children with semantic-pragmatic disorder from other language-impaired children.

Design of the Checklist

After obtaining feedback from teachers and therapists on pilot versions of questions, a Checklist for Language-Impaired Children (CLIC) was devised.

As shown in Table 9.2, the checklist represented a range of communicative behaviors. Most of these items were selected with the aim of including behaviors that were highlighted in clinical classifications of language impairment but were not readily assessed on standardized tests. Additional items were included to assess nonverbal communication and behaviors that have been described as associated with PDD (i.e., social interaction, interests, attention, and gross motor skills).

Each CLIC item had five possible responses. In general, these were de-

²This section was written prior to the publication of an update on the empirical validation of Dr. Bishop's instrument. For this update, the reader is referred to Bishop (1998).

TABLE 9.2. Areas Covered by Checklist

Intelligibility (F)	Conversational responsiveness (C)
Expressive phonology (F)	Conversational assertiveness (C)
Grammar (F)	Conversational coherence (C)
Morphosyntax (F)	Use of context in comprehension (C)
Vocabulary (C)	Echolalia (past or present) (A)
Literal comprehension	Jargon (past or present) (A)
Narrative skills (C)	Eye contact (A)
Speech rate	Peer relations (A)
Fluency	Relations with adults (A)
Speech volume (C)	Interests (A)
Intonational melody (C)	Attention
	Gross motor skills

Note. Items coded F, C, or A contributed to composite scales (see text).

signed to fall on an ordinal scale, with one pole of the scale corresponding to the most normal response and the other to the most abnormal. However, for some behaviors it was difficult to devise items of this kind, because departures from normal behavior could take different forms, and for some items both poles represented abnormality, with more typical behavior corresponding to the mid-point of the scale. Table 9.3 shows a sample item. Raters were asked to check the item that was *most like* the child. If they felt that the child's behavior fell between two adjacent options, they were asked to check both options. Such dual responses were assigned a score intermediate between the two responses.

Sample

Head teachers from three residential schools were approached, and all agreed to take part, although the amount of data provided by each school depended on the willingness of individual staff members to participate. Each of the schools specialized in the education of children with specific language disorders and had stringent entry requirements, excluding children with mental handicap or serious behavioral problems. The aim was to obtain independent ratings for each pupil from two members of staff, one a teacher and one a speech-language therapist, each of whom had known the child for at least 3 months. A total of 17 teachers and 27 speech-language therapists participated in data collection. School head teachers were also sent a "diagnostic checklist" for each child, which included questions about whether the child had ever had a diagnosis of definite or possible semantic-pragmatic disorder, autistic disorder, autistic features, AS, or "clumsiness"/coordination disorder. It was explained that these categories were not mutually exclusive, and the respondent was encouraged to check as many diagnoses as seemed to apply. Information was also gathered about any other

TABLE 9.3. Sample Item from CLIC**CLIC Item 10**

How responsive is the child to conversational overtures from a familiar person?

- Frequently ignores conversational overtures from others (e.g., if asked "What are you making?" the child just continues working as if nothing had happened).
- Sometimes ignores conversational overtures from others.
- May not always give a verbal response to a conversational overture but will almost always respond nonverbally; by demonstration, gesture, nodding or shaking the head, smiling, etc. (e.g., if asked "What are you making?" the child will hold up or point to the thing that he is making and respond by looking or smiling).
- Usually gives an appropriate verbal response to a conversational overture but does not elaborate beyond the minimum response that is required (e.g., if asked "What are you making?" will reply "A boat").
- Typically gives a full and appropriate response that goes beyond the minimum required (e.g., if asked "What are you making?" will reply "A boat; we're going to sail it on the lake when it's finished").

Note. The full checklist is not shown here, because a new version is under evaluation. Readers interested in using CLIC should contact the author for details of the most recent version.

physical or sensory deficits. Only those who were diagnosed as having a developmental language disorder were included, and any children with a diagnosis of possible or definite autism were excluded. Table 9.4 shows the sample composition. Two checklists were completed by independent raters for the majority of pupils, but there was a subset of children for whom only one rating was available. Mean age for the whole sample was 11.07 years, with a range from 5 to 16 years.

Combination of Items into Language Dimensions

Each checklist item was recoded on a 5-point scale, such that maximum deficit corresponded to a score of -3, average performance to a score of 0, and

TABLE 9.4. Constitution of Sample for Checklist Study

Diagnostic information obtained from school	Checklist completed by therapist and teacher	Checklist completed by therapist only	Checklist completed by teacher only
Has developmental language disorder: not diagnosed as semantic-pragmatic disorder	71	28	3
Has developmental language disorder: diagnosed as possible or definite semantic-pragmatic disorder	26	5	0

superior performance to a score of 1. (For bipolar items, where the middle of the scale represented normality, both poles might be given the same negative score.) After excluding individual items with low interrater reliability (those assessing literal comprehension, speech rate, and fluency), scores were combined into three scales (see Table 9.2): The first (F-scale) was composed of items assessing language *form* and the second (C-scale) measured language *content and use*. A third scale, (A-scale) assessed presence of *autistic features*, including several nonverbal behaviors.

Composite scale scores are reported only for those checklists in which all items constituting the scales had been completed by both raters ($n = 83$). For this sample size, a correlation of .22 is significant at the 5% level, and a correlation of .28 at the 1% level. The F-scale (language form) had a possible range of -12 to +4, and gave interrater agreement (r_i) of .848. The C-scale (language content and use) had a possible range of scores from -22 to +6. Interrater agreement on this scale was $r_i = .468$. The A-scale (autistic features) had a possible range from -12 to 0, with $r_i = .777$. Ratings on the C-scale and A-scale were unrelated to age ($r < |.22|$). Scores on the F-scale showed a statistically significant correlation with age (for teachers' ratings, $r = .256$; for therapists' ratings, $r = .447$, $p < .05$).

The correlation between the F-scale and A-scale was nonsignificant for teachers ($r = .016$), but a weak *negative* association was seen in ratings by therapists ($r = -.237$), with better scores on language form being associated with more autistic features. There was a positive association between the C-scale and A-scale for both teachers ($r = .469$) and therapists ($r = .406$).

Distribution of Scores on Two Dimensions

Because more data were available from therapists than from teachers, the therapists' data were used to explore relationships between the scales. Analyses were based just on the subset of children who had no missing data.

Figure 9.4 shows the scatterplot of data on the two language scales, with children subdivided according to whether or not they had a school diagnosis of semantic-pragmatic disorder. It is evident from inspection that children with such a diagnosis tend to cluster in the lower-right-hand side of the figure, as would be expected if the diagnosis were being used for children with a relative strength in language form and weakness in content/use.

One noteworthy aspect of Figure 9.4 is that it illustrates that there are several children with marked impairments on both the C-scale and the F-scale. Indeed, overall, there was a modest but significant correlation between scores on F- and C-scales ($r = .289$). Thus, there are children who had problems in both domains, as well as others who have problems predominantly on just one scale.

A further analysis was done to see whether the degree of *mismatch* be-

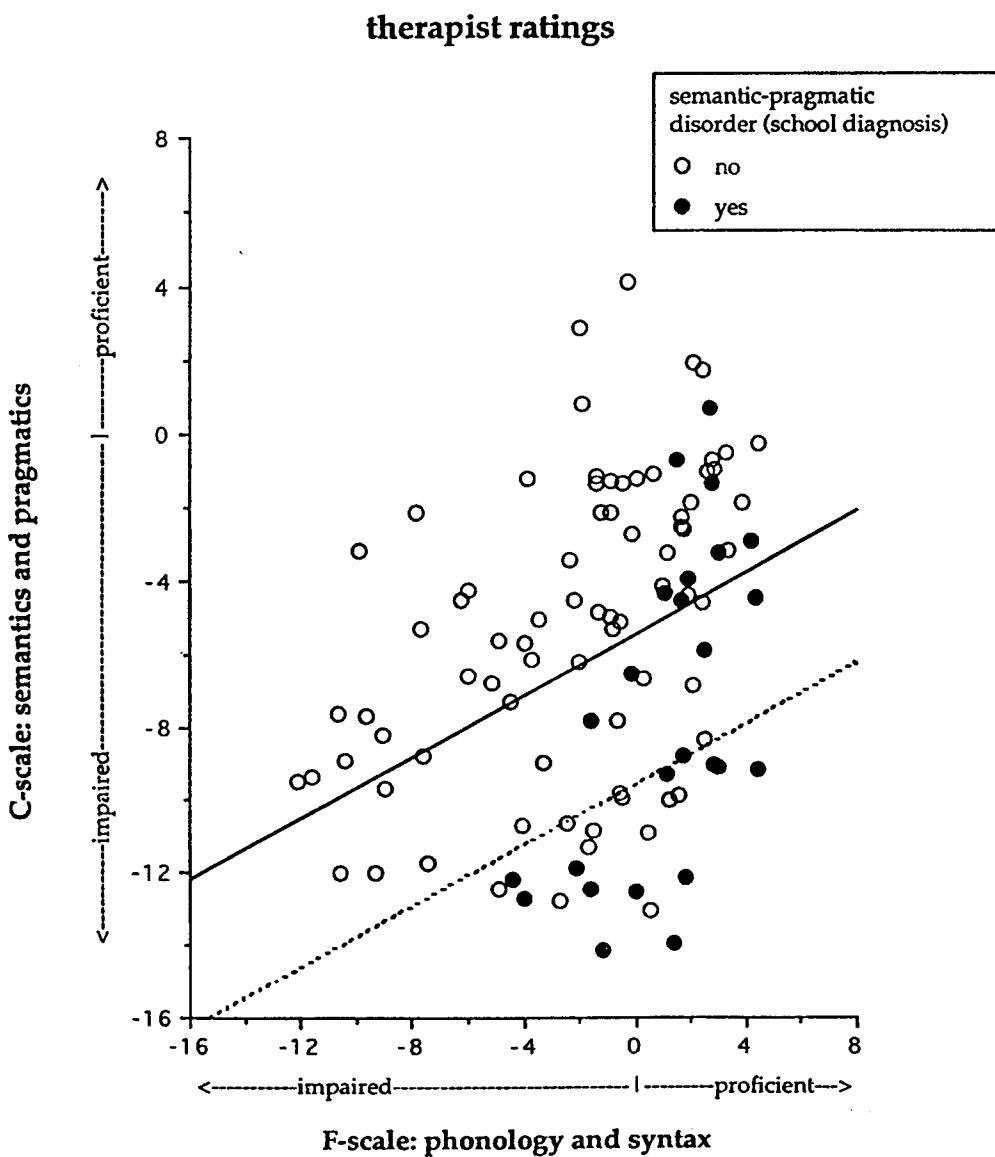


FIGURE 9.4. Scatterplot showing distribution of F-scale and C-scale scores in relation to school diagnostic categorization as definite or possible semantic-pragmatic disorder ("yes" or "no"). The two subgroups differed significantly on the F-scale (mean for "yes" = 1.01; $SD = 2.34$; mean for "no" = -2.3; $SD = 4.19$; $t(114) = 4.05$; $p < .001$), on the C-scale (mean for "yes" = -7.52, $SD = 4.47$; mean for "no" = -5.54, $SD = 4.32$; $t(114) = 2.11$; $p < .05$). (Data were missing for two children in the "yes" group and three in the "no" group.) The bold line is the regression line for predicting C-scale scores from F-scale scores. The SP deficit score corresponds to the shortest distance between a point and the bold line, with those below the line being negative, and above the line positive. Points below the dotted line are cases where C-scale scores are more than 1 SD below the level predicted from F-scale scores.

tween C- and F-scales may be better at differentiating groups than the absolute level of impairment on either scale. This can be quantified using the regression equation for predicting C-scores from F-scores to compute residuals which reflect the extent to which C-scale scores were discrepant with F-scale scores. In effect, these scores, which will be referred to as *SP deficit scores*, measure how far away from the bold line a child's score falls.

These SP deficit scores clearly differentiated children who did and did not have a school diagnosis of semantic-pragmatic disorder ("no" group mean = 0.24; $SD = .936$; $n = 84$; "yes" group mean = -0.59; $SD = .99$; $n = 28$; $t(110) = 4.00$, $p < .001$), and also had acceptable interrater reliability ($r_i = .548$; $n = 65$).

Item Analysis and Individual Variation

Although the analyses presented so far offer support to the notion of a subgroup of children with semantic-pragmatic disorder within the language-impaired population, there are aspects of the data that caution us against regarding this as a "syndrome." An item analysis comparing the pattern of responses seen in children who had a school diagnosis of semantic-pragmatic disorder and those who did not was used to find those response options (i.e., from all five possibilities offered for each item) that most clearly discriminated groups. Table 9.5 shows these response profiles. It is noteworthy that four of these items (lack of eye contact, poor peer relations, poor rapport with adults, obsessional interests) came from the Autistic Features scale. However, as can be seen from that table, different patterns of deficit characterize different children in the semantic-pragmatic group. Table 9.5 also shows which children had school diagnoses of "autistic features," AS, or developmental clumsiness (the latter category being of interest because of a possible association with Asperger's disorder). The only child thought to have a definite diagnosis of AS (shown as + in the table) had significant language delay and would not have met DSM-IV criteria for this disorder. Although several of the children with semantic-pragmatic disorder were regarded as clumsy, this was not a discriminating item on the checklist: Clumsiness was fairly common throughout the sample.

Pragmatic Language Impairments and Pervasive Developmental Disorder

As we have seen, the original definition of PDD requires two conditions be met: The child's disorder involves several areas of development, and the impairments correspond to distortions rather than delays in development.

The CLIC study offers some support to the view that there is a subset of children in the language-impaired population who meet both these requirements. First, there was a clear link between impairments on the Content and Use language scale and the Autistic Features scale but no relation between

TABLE 9.5. Response Profiles of Individual Children with Diagnosis of "Semantic-Pragmatic Disorder"

Age (yr)	6	6	8	8	9	9	9	10	10	11	11	12	13	13	13	14	14	14	14	15	15	15	16
Sex	F	F	M	F	M	M	M	M	M	M	M	F	M	M	M	M	M	M	M	M	M	M	M
Autistic features ^a	?	—	—	—	—	—	?	—	—	—	—	?	—	—	—	—	—	—	?	—	—	—	—
Asperger syndrome ^a	—	—	—	—	—	—	?	—	—	—	—	+	—	?	—	—	—	—	—	?	—	—	—
Clumsiness	+	—	+	—	—	—	+	—	—	—	—	+	?	?	?	?	—	+	?	—	+	?	—
Intelligibility (excellent)	—	—	—	—	—	—	+	—	—	—	—	+	?	?	?	?	—	—	—	—	—	—	—
Expressive phonology (all sounds correct)	—	—	—	—	—	+	—	—	—	—	—	+	+	+	+	+	+	+	+	+	+	+	+
Intonation (expressive but stereotyped)	+	+	+	+	—	+	+	—	—	—	—	+	—	—	—	+	—	—	—	—	—	—	—
Grammar (complex, adult-like)	—	—	—	—	—	—	+	—	—	—	—	+	+	+	+	+	—	—	+	—	—	+	+
Morphosyntax (occasional errors only)	+	—	+	—	+	+	+	—	—	—	—	+	+	+	+	+	+	—	—	+	—	—	+
Conversational responsiveness (sometimes ignores overtures)	—	—	—	+	—	+	+	—	—	—	—	—	—	—	—	—	—	—	—	—	—	—	—
Conversational assertiveness (tends not to initiate conversation)	—	—	—	+	—	—	+	—	—	—	—	—	—	—	—	—	—	—	—	—	—	—	—
Conversational coherence (controls or abruptly changes topic)	—	+	+	—	—	—	+	+	—	—	—	+	+	+	+	—	—	+	+	+	+	+	+
Use of context (overliteral comprehension)	—	—	+	+	+	—	—	+	+	—	—	—	—	—	—	—	—	—	—	—	—	—	—
Eye contact (seldom/never)	—	—	—	—	+	—	—	—	—	—	—	+	—	—	—	—	—	—	—	—	—	—	—
Peer relations (avoided/neglected by others)	—	—	—	—	+	—	—	+	—	—	—	—	+	—	+	—	—	—	+	—	—	—	—
Adult relations (hard to establish rapport)	—	—	—	—	+	—	—	+	—	—	—	—	—	—	—	—	+	—	—	—	—	—	—
Interests (obsessional)	—	—	+	+	—	—	—	+	—	—	—	+	+	—	—	+	—	—	+	—	—	+	—

Note. + indicates presence; — indicates absence; ? indicates possible or previous; . indicates missing data.

^aInformation from school diagnostic checklist.

autistic features and impairments on the Language Form scale. Second, most children with a clinical diagnosis of semantic-pragmatic disorder did have ratings indicating impairment on one or more of the nonverbal behaviors from the A-scale (see Table 9.5). Third, whereas items assessing language form showed clear developmental trends, those assessing content and use did not. This is tentative evidence for the view that these items correspond to distortions rather than delays in development. However, this finding could reflect the relatively low reliability of the Content and Use scale and the selected nature of the cross-sectional sample used here; a proper test would involve longitudinal assessment using more reliable measures.

One limitation of the CLIC study was that we did not use a standard diagnostic assessment but relied on diagnostic information in children's records to exclude those with possible or definite diagnoses of autistic disorder. The checklist provides a useful first step toward reliably identifying pragmatic communicative deficits in children: This approach now needs to be combined with more systematic diagnostic studies, using observational and interview methods such as the Autism Diagnostic Observation Schedule (Lord et al., 1989) or the Autism Diagnostic Interview—Revised (Lord, Rutter, & Le Couteur, 1994), which will enable us to quantify autistic behaviors in the same children. This would make it possible to confirm that the diagnostic information provided by the school was accurate in stating that these children did not meet criteria for autistic disorder.

If we accept that significant impairments of language content and use are seen in children who do not meet criteria for autistic disorder, how should we interpret such findings? Should we adopt the concept of semantic-pragmatic disorder, or would it be more proper to abandon the term and simply diagnose such children as cases of PDD-NOS? There are problems with both solutions. The term "semantic-pragmatic disorder" implies a clear-cut diagnostic entity, yet the CLIC study showed fairly wide variation in the particular constellation of reported abnormalities. Although the low reliability of items could contribute to this variable picture, this is unlikely to be the whole story. In other studies I am conducting, which involve fine-grained analysis of conversational behavior of children with semantic-pragmatic disorder, individual differences in behavior are the rule rather than the exception. Thus there are some children with marked limitations in use of eye contact and others who make excellent use of gaze and facial expression; some children have clear evidence of returning to favored topics in conversation and others have no special subjects that they prefer. Some have marked peculiarities of prosody; others do not. Such observations suggest that a categorical label such as semantic-pragmatic disorder may be misleading in implying sharper diagnostic boundaries and greater uniformity of problems than is the case. Another problem with the term "semantic-pragmatic disorder" is that it implies semantic and pragmatic difficulties go hand in hand, whereas the CLIC study gave little support to this view. Table 9.5 shows those items that best discriminated children with a diagnosis of

semantic-pragmatic disorder from the remainder: None of these included items assessing semantic abilities, although such items had been included in the checklist. Semantic difficulties tended to characterize all language-impaired children but did not discriminate between subtypes.

What of the alternative, which would be to abandon the term "semantic-pragmatic disorder" altogether for PDD-NOS or "autistic spectrum"? The difficulty with this solution is that both PDD-NOS and autistic spectrum are vaguely defined, catch-all terms which encompass a huge variety of children (see, e.g., Towbin, 1997, for an account of PDD-NOS). Klin, Mayes, Volkmar, and Cohen (1995) argue that the use of PDD-NOS "adds little more than a demarcation of uncharted territory of clinical complexity" (p. S7). Clinically, these labels are unhelpful in specifying either the kind of educational provision the child requires or the severity of the problems. Also, by emphasizing the continuity with autism and related disorders, they draw attention to the social and behavioral impairments and give less prominence to children's structural language difficulties, which are also important, especially in younger children. In common with Conti-Ramsden, Crutchley, and Botting (1997), I recommend that the term "pragmatic language impairment" (PLI) be used to refer to children who occupy an intermediate position between core autistic disorder and specific language impairment (see Figure 9.5). As the diagram illustrates, however, there are no

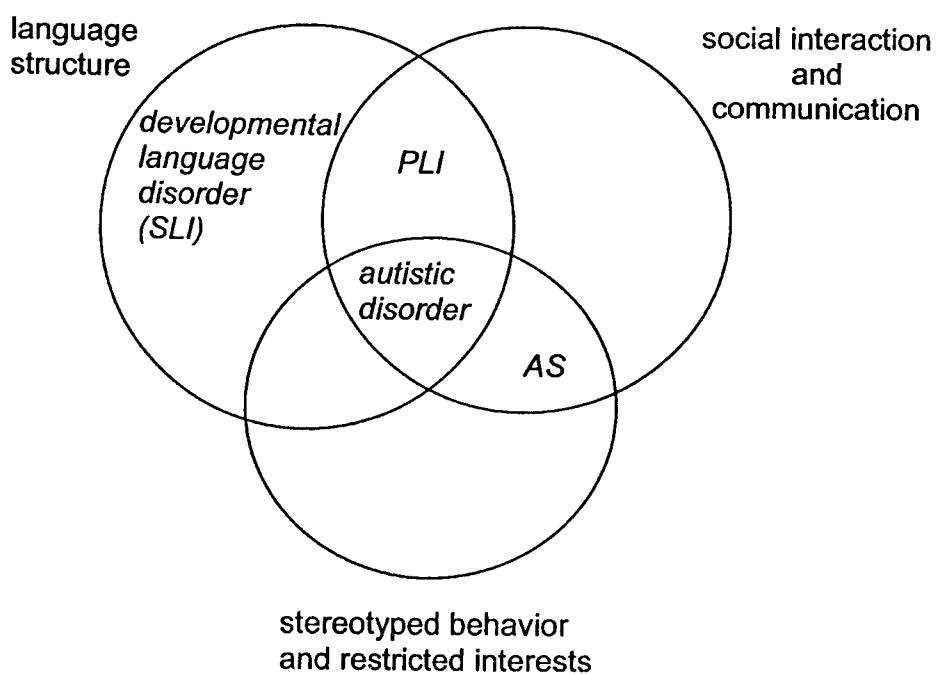


FIGURE 9.5. Set diagram showing how pragmatic language impairment relates to typical SLI and autistic disorder.

sharp boundaries between disorders, and this categorical label should be seen as a shorthand to describe children who occupy a particular region of a multidimensional space, rather than implying that there is a discrete syndrome.

If these children do fall between classic autistic disorder and SLI, why do so few of them attract an autism-related diagnosis? Further research is needed on this point, but I can offer some speculations, based on personal observations. First, the pragmatic difficulties themselves are relatively subtle. In the early study by Bishop and Adams (1989), children with a diagnosis of semantic-pragmatic disorder had higher levels of conversational inappropriacy than did other language-impaired children, but it was nevertheless the case that the majority of their conversational contributions were deemed appropriate. Second, the *social* limitations of children with a diagnosis of semantic-pragmatic disorder are typically milder than those seen in core autistic disorder. These children are usually outgoing and sociable, although the quality of their interaction may be odd or deficient. Finally, the language profile of these children can change radically over time. I have seen children whose early language milestones were seriously delayed (e.g., still using single words at 4 years of age) yet who were talking fluently in long, complex sentences by middle childhood. The combination of serious delays in acquiring formal language skills, coupled with an outgoing social manner, may lead professionals to assume that the language difficulties are the primary problem, with any social limitations being secondary to poor language comprehension or expression. It is only when the child grows older and starts to speak intelligibly that it becomes apparent that there are oddities of social interaction that cannot be attributed to linguistic limitations.

BEYOND ASPERGER SYNDROME

The CLIC study has been used to illustrate the fact that children who fall between diagnostic boundaries for AS, autistic disorder, and developmental language disorder are not uncommon: The region marked as PLI in Figure 9.5 is not as sparsely populated as the textbooks suggest. Furthermore, children inhabiting that region are fairly diverse. My emphasis has been on children who are receiving special educational provision for language difficulties; I suspect that even greater variability might have been seen had checklist data been gathered from a broader range of settings. Indeed, when relatively high-functioning children present with subtle deficits affecting a range of different behaviors, one has the impression that the particular diagnosis, and consequently the type of intervention received, may be more a function of the discipline of the specialist who is the point of first referral than of the particular symptom profile. The same child might receive a diag-

nosis of PDD-NOS or atypical autism from a psychiatrist, of developmental language disorder (semantic-pragmatic type) from a speech-language therapist, or right-hemisphere learning disability from a neuropsychologist (see Shields, 1991; Klin, Volkmar, et al., 1995).

What does this diversity represent and how should we respond to it? The question boils down to whether the different patterns of impairment represent different manifestations of a common underlying disorder or a group of distinct disorders. The answer depends partly on the level of description we choose to adopt. There is mounting evidence that if we are interested in uncovering biological causal factors, it can be misleading to focus on a stringently defined disorder; a broader phenotype gives a more coherent picture of heritability (see, e.g., Bailey et al., 1995). However, even if the ultimate genetic or other neurodevelopmental cause is similar for the whole range of PDD, there might be variability in the brain regions affected, and this could be one reason why such a wide range of patterns of neuropsychological deficit is observed. For the neuroscientist interested in brain-behavior relationships it might make sense to focus on much more tightly defined groups of children with similar patterns of symptoms, as this would provide the best opportunity for uncovering a common site of brain dysfunction. For instance, the specific behavioral profile characterizing AS seems to reflect right-hemisphere involvement, whereas high-functioning autism does not (Klin, Volkmar, et al., 1995). For the psychologist interested in exploring the cognitive and social processes that underpin a child's deficits, a focus on a single domain of functioning might be appropriate, and the emphasis would be on children who show selective impairments in just one area, because such children allow us to study a deficit in a relatively pure form, in the absence of other deficits that might cloud interpretation. Thus, for instance, in considering whether a "theory of mind" deficit can explain abnormalities of social behavior, it could make sense to focus on those rare children in the PDD spectrum with a relatively pure impairment of social behavior and relatively good formal language skills. Finally, for those interested in planning appropriate intervention, it is necessary to concentrate on assessing the individual child's pattern of strengths and weaknesses, so categorical labels become much less important, especially if they fail to capture the full range of variation seen clinically.

The category of AS has its uses. There are certain research questions that are best approached by focusing on one tightly defined subgroup of children, and the potential domain of PDD is so broad that it can make sense to start with a tightly defined condition and then expand outward, rather than trying to uncover order in PDD as a whole. However, I would recommend that such highly focused research should be complemented by studies that start with a more broadly defined group of PDD, attempt to measure the salient dimensions of variation, and look for correlates of these dimensions and natural clusterings of behaviors. An exclusive focus on narrow categories such as AS has led us to behave as if there are sharp boundaries

when these are probably artificial. It has also produced a disproportionate focus of research effort on a small subset of children who seem seriously underrepresentative of the range of cases seen in clinical practice. Not only will a complementary approach adopting a broader perspective redress this imbalance, but it may also help throw light on the puzzling deficits seen in AS by revealing similar difficulties in children with other profiles of impairment.

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