Exploring the borderlands of autistic disorder and specific language impairment: a study using standardised diagnostic instruments

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Background: Two studies were conducted to test claims that pragmatic language impairment (PLI previously referred to as semantic-pragmatic disorder) is simply another term for autistic disorder or pervasive developmental disorder not otherwise specified (PDDNOS). Method: In Study 1, 21 children aged from 6 to 9 years with language impairments were subdivided on the basis of the Children's Communication Checklist into 13 cases of pragmatic language impairment (PLI) and eight cases of typical specific language impairment (SLI-T). Parents completed the Autism Diagnostic Interview -Revised (ADI-R) and the Social Communication Questionnaire (SCQ), and the children were given the Autism Diagnostic Observation Schedule - Generic (ADOS-G). In Study 2, a further 11 children with SLI-T and 18 with PLI were assessed using the SCQ and ADOS-G. In addition, six children diagnosed with high-functioning autism and 18 normally developing children were assessed. Results: There was good agreement between ADI-R and SCQ diagnoses, but poor agreement between diagnoses based on these parental report measures and those based on ADOS-G. In many children, symptom profiles changed with age. Four PLI children from Study 1 and one from Study 2 met criteria for autistic disorder on both parental report (ADI-R or SCQ) and ADOS-G. Many of the others showed some autistic features, but there was a subset of children with pragmatic difficulties who were not diagnosed as having autism or PDDNOS by either instrument. These children tended to use stereotyped language with abnormal intonation/prosody, but they appeared sociable and communicative, had normal nonverbal communication, and showed few abnormalities outside the language/social communication domains. Conclusions: Presence of pragmatic difficulties in a child with communication problems should prompt the clinician to evaluate autistic symptomatology, but it is dangerous to assume that all children with pragmatic difficulties have autism or PDDNOS. Keywords: Autism, PDDNOS, pragmatics, language impairment, semantic-pragmatic disorder, diagnosis. Abbreviations: ADI-R: Autism Diagnostic Interview - Revised; ADOS-G: Autism Diagnostic Observation Schedule - Generic; CCC: Children's Communication Checklist; HFA: high functioning autism; MLU: mean length of utterance in morphemes; PDDNOS: pervasive developmental disorder not otherwise specified; PLI: pragmatic language impairment; SCQ: social communication questionnaire; SLI(-T): (typical) specific language impairment.

It has long been recognised that specific language impairment (SLI) is a heterogeneous category, but there is little consensus as to how best to subcategorise children. In the 1980s, two classification systems were independently published, both describing a subtype of language impairment in which expressive language was fluent, complex, and clearly articulated, but there were abnormalities in the way in which language was used (Rapin & Allen, 1983; Bishop & Rosenbloom, 1987). Children with this clinical profile tend to be verbose, to have problems in understanding and producing connected discourse, and to give conversational responses that are socially inappropriate, tangential and/or stereotyped. The term 'semantic-pragmatic disorder' was used to describe such a profile, though more recently Conti-Ramsden and Botting (1999) and Bishop (2000) have proposed the term 'pragmatic language impairment' (PLI). That term will be adopted in this paper, but should be regarded as subsuming earlier references to 'semantic-pragmatic disorder'.

The relationship between PLI and autistic disorder has been a matter of much debate. Rapin and Allen (1987) noted that the clinical profile of semantic-pragmatic disorder is often seen in children who have a diagnosis of high-functioning autism. However, they maintained that this profile is not confined to autistic disorder. Our focus in this paper is on children who have been given a primary diagnosis of language impairment, but where problems are not confined to structural aspects of language, but affect the appropriate use of language in context.

Soon after the category of 'semantic-pragmatic disorder' was described, several authors challenged the status of this category, arguing that the children to whom it was applied appeared to have many of the characteristics of autistic disorder (e.g., Lister Brook & Bowler, 1992; Gagnon, Mottron, & Joanette, 1997). More recently, Shields, Varley, Broks, and Simpson (1996a, b) concluded that PLI is a form of high-functioning autism. They reached this conclusion on the basis of a set of studies in which they demonstrated on both tests of social cognition and

neuropsychological measures that there was a much closer relationship between PLI and autistic disorder than between PLI and typical SLI. An information sheet published on a website belonging to the National Autistic Society (http://www.nas.org.uk/) notes that there is debate about whether semantic pragmatic disorder is merely a descriptive term for 'communicative difficulty found in verbal people with autism' and concludes that it 'has the same underlying triad of socio-cognitive deficits as high functioning autism'.

A different perspective on the relationship between autistic disorder and PLI was proposed by Bishop (1998, 2000), who suggested that some children had a profile of disorder that was intermediate between SLI and core autism. Bishop emphasised data from her own studies, and research on relatives of people with autism, that revealed the existence of cases who did not show the full triad of deficits that characterise autism, namely social interaction, communirestricted interests/behaviours, although some autistic features may be present. Using diagnostic data from school records of children attending language units, Bishop (1998) noted that there was a subgroup of children with 'semantic-pragmatic disorder' who were not noted as having any autistic features. These 'pure' cases did not differ from those with typical SLI on checklist scales assessing interests and social relationships, although they did differ on scales assessing pragmatic aspects of communication. However, other studies have illustrated how the pattern of impairments may change with age, so that children who appear to meet criteria for autism at one age no longer do so later on - and vice versa. This is particularly striking in the longitudinal data from Mawhood, Howlin, and Rutter (2000), who found that children who were clearly distinguished as either having a receptive language impairment or autistic disorder in the early school years were much harder to differentiate in adulthood. Such observations raise several possible scenarios about the relationship between PLI and autistic disorder:

Scenario 1. All children with PLI will meet diagnostic criteria for autistic disorder if past history as well as current behaviour is taken into account. The apparent existence of cases of PLI who do not appear autistic simply reflects the fact that autistic symptoms may improve over time. Reluctance to diagnose autism, or unfamiliarity with autistic symptomatology could be other reasons why autism fails to be diagnosed.

Scenario 2. Some children with PLI will not show the full triad of autistic impairments, even when past history is considered, but all will show some autistic features, making a diagnosis of PDDNOS more appropriate than one of specific developmental language disorder. (In the International Classification of Diseases (ICD-10; World Health Organisation, 1992), the term 'atypical autism' is used for such cases.)

Scenario 3. PLI can be found in non-autistic children. If this account proves to be accurate, it raises the question of whether the nature of pragmatic difficulties is different for autistic vs. nonautistic children.

Although the debate about overlap between autistic disorder and PLI has continued over several years, we are not aware of any study that has addressed the question by applying standardised diagnostic procedures for autism to a group of children with the clinical picture of PLI. The current study aimed to remedy this situation, incorporating diagnostic methods that take into account the child's profile of behaviour in the first few years of life, when autistic symptoms are usually most apparent, as well as observations of current behaviour.

Until recently, studies of PLI were hampered by the lack of any clear criteria for identifying this pattern of disorder: investigators were forced to rely on subjective judgements as to how well children met clinical descriptions of this disorder. Recently, however, Bishop (1998) developed the Children's Communication Checklist, which showed promise as a method for selecting from the language-impaired population those children who have features of PLI.

The first study involved a contrast between two language-impaired groups of children. Group PLI were selected on the basis that they obtained low scores on the pragmatic composite of teacher ratings on the Children's Communication Checklist, whereas group SLI-T (typical SLI) did not. These children were compared on three diagnostic instruments, all of which targeted symptoms of autistic disorder.

Study 1

Methods

Participants. Children with language impairments were identified from one residential school and two language units, all of which specialised in the education of children with SLI. Teachers were asked to complete the Children's Communication Checklist (CCC; Bishop, 1998) for all pupils in their class aged from 6 to 9 years, where parental permission for participation had been obtained, and where the child did not have any physical handicap, sensorineural hearing loss, a definite diagnosis of autistic disorder or any other neurological or genetic syndrome.

Diagnoses of 'autistic spectrum disorder', 'atypical autism', or 'autistic features' were noted, but not regarded as grounds to exclude the child from the study. The term 'autistic spectrum disorder' posed particular problems, as it is sometimes used to refer to children who meet core diagnostic criteria for autism, but can

also be used to encompass milder cases. We decided to include children with this diagnosis because we were interested in the extent to which autistic symptomatology co-occurred with pragmatic impairments, and if we had excluded children in whom autistic features had been noted, this would underestimate the association. We also included two children who had a diagnosis of Asperger syndrome, as in both cases the child had early language delay, which meant that according to DSM-IV criteria the diagnosis was invalid. Rather it seemed that a diagnosis of Asperger syndrome, rather than autism, was used to reflect the fact that autistic symptoms were relatively mild. Overall, six of the 13 children in the PLI group, and two of the eight children in the SLI-T group had some autism-related diagnosis.

We selected all available children (N=13) with nonverbal IQs of 80 or above, who were able to talk intelligibly in sentences, and whose scores on the pragmatic composite of the CCC fell below 133, this being the cutoff identified by Bishop (1998) as most efficient at discriminating children with a diagnosis of semantic-pragmatic disorder from more typical cases of SLI. Criteria for inclusion in the SLI-T group were a pragmatic composite greater than 132, plus score on a standardised language test at least 1 SD below the normative mean. We aimed to identify an equivalent number of SLI-T and PLI children, but we fell short of this goal because of withdrawal from the study by one parent, and failure to meet IQ or language criteria in other cases, giving a final sample of eight children.

Assessments. All children were given Raven's Coloured Matrices (Raven, Court, & Raven, 1986) to confirm that their nonverbal ability was in the normal range (scaled score at least 80). Other experimental tests of language and executive functions were administered, but will not be described here. A narrative task was used to elicit a sample of speech, from which mean length of utterance in morphemes (MLU) was computed. A receptive language composite was computed on the basis of available standardised scores on at least two language tests from the following: British Picture Vocabulary Scale (Dunn, Dunn, Whetton, & Pintilie, 1982), receptive subtests from the Clinical Evaluation of Language Fundamentals - Revised (Semel, Wiig, & Secord, 1987), and the Test for Reception of Grammar (Bishop, 1989).

Teachers were asked to complete the Children's Communication Checklist (CCC: Bishop, 1998) for all children in the study. This checklist contains 70 items divided into nine scales: two scales assess aspects of language structure (speech and syntax), five scales assess pragmatic aspects of communication (inappropriate initiation, coherence, stereotyped conversation, use of context, and rapport), and two assess nonlinguistic aspects of autistic behaviour (social relationships and interests). Each item contains a statement describing a specific behaviour (e.g., 'talks repetitively about things that no-one is interested in') which is rated as 'definitely applies', 'applies somewhat', 'does not apply' or 'unable to judge'. Most items describe communicative difficulties, but a few describe strengths rather than weaknesses. The pragmatic scales can be combined to form a pragmatic composite. Inter-rater reliability for two professionals rating the same child on the pragmatic

composite was estimated at .80 by Bishop (1998), although inter-rater agreement between a parent and professional was only .47, for a heterogeneous clinical sample studied by Bishop and Baird (2001).

Diagnostic information relating to autistic disorder was obtained from three sources.

- 1. The Autism Diagnostic Observation Schedule Generic (ADOS-G; Lord et al., 2000), Module 3. This is a semi-structured assessment of communication, social interaction and play/imagination. It uses standard activities to elicit and observe behaviours that are important in the diagnosis of pervasive developmental disorders. The examiner rates behaviours as they occur, and these ratings are incorporated in a diagnostic algorithm. Module 3 is designed for use with children who have fluent, multiclause speech and it takes around 45 minutes to administer. With the permission of Catherine Lord, we substituted the wordless picture book 'Frog, Where are you?' (Meyer, 1969) for the storybook usually used to elicit a narrative in ADOS-G, as this allowed for comparability with other studies by ourselves and others on children's narratives. All children were given this assessment by an examiner (CFN) who had been certified as having achieved acceptable levels of reliability.
- 2. The Autism Diagnostic Interview Revised, 3rd edition (ADI-R; Lord, Rutter, & Le Couteur, 1994). This is an interview with the child's principal caregiver that is designed to obtain detailed descriptions of behaviours that characterise pervasive developmental disorders. It typically takes around 3 hours. The interviewee is required to give detailed descriptions of the child's behaviour, rather than simply responding 'yes' or 'no'. Coded items are converted to numerical scores for the domains of reciprocal social interaction, communication, and repetitive behaviours/stereotyped patterns. Ratings of these domains (based predominantly on behaviour at age 4;0 to 5;0) can be used as the basis for a diagnosis of autistic disorder according to ICD-10 criteria. In this study we also considered how many children met autism diagnostic criteria when ratings of current behaviour were substituted for those at 4;0 to 5;0 years. The ADI-R was given by a trained interviewer (DB or CFN).
- 3. The Social Communication Questionnaire (SCQ; Berument, Rutter, Pickles, Lord, & Bailey, 1999). This 40-item screening questionnaire (previously known as the Autism Screening Questionnaire) is completed by parents to obtain information about core diagnostic features of autism. The items cover aspects of behaviour identical to those asked about in the ADI-R. We included the SCQ because our study provided a good opportunity to assess agreement between the SCQ and ADI-R in a high-functioning population. Berument et al. (1999) demonstrated excellent agreement between the SCQ and ADI-R, for parents who completed the SCQ after the ADI-R in a mixed sample of children with autism and non-autistic disorders. In the current study, the SCQ was mailed to parents before the ADI-R was administered, and so questionnaire responses could not have been influenced by experience of the interview.

Results

Background data. Table 1 shows data on age, sex, nonverbal ability, receptive language, and MLU on the narrative task. Both groups scored well within normal limits on the measure of nonverbal ability, but the PLI group had significantly higher scores, F(1,19)=5.06, p=.037, and were also significantly younger than the SLI-T group, F(1,19)=6.15, p=.02. The groups did not differ on Receptive Language, F(1,19)=3.7, p=.07, or MLU, F(1,18)=2.05, p=.169.

Autism diagnostic instruments. ADI-R Data from the ADI-R were scored according to the algorithm specified by Lord et al. (1994); children who scored above cutoff on all three domains (social, communication, and interests) were categorised as meeting ICD-10 criteria for autistic disorder. No cutoffs have been specified for PDDNOS. For this study, those scoring above the autism cutoff on two out of three domains were categorised as PDDNOS. The same criteria were then applied to scores based on current status: this procedure is not part of standard ADI-R scoring, but provides the opportunity to observe how children's autistic symptomatology changes over time.

As shown in Tables 2 and 3, using conventional criteria that base the diagnosis primarily on behaviour at age 4;0–5;0, eight children (six from the PLI group and two from the SLI-T group) scored above cutoff for autistic disorder on the ADI-R. A further six children (two from the PLI group and four from the SLI-T group) scored above cutoff on two of the three domains (five on social and communication, and one on communication and repetitive behaviours), meeting our criteria for PDDNOS.

The ADI-R gave clear evidence of change in symptomatology over time. Three of the eight children who met the conventional algorithm for autistic disorder at age 4;0-5;0 scored below cutoff when the algorithm was recomputed on the basis of current behaviour. Five out of six children who were above threshold on two domains using the conventional algorithm did not score in the autistic range on the basis of current behaviour.

SCQ. In the SCQ, each of the 40 items is scored zero or one, and the summed total used to categorise children. Following Berument et al. (1999), a cutoff of 15 or more on the SCQ was used to categorise a child

as having PDDNOS, and a cutoff of 22 or more for a diagnosis of autistic disorder. SCQ diagnoses are shown in Tables 2 and 3, and indicate good agreement between the SCQ and ADI-R: $\chi^2(4) = 19.29, p < .001$. Despite this good agreement on diagnostic categorisation, item-by-item agreement between SCQ and ADI-R was not impressive. ADI-R items can be coded as 0 (no abnormality), 1 (behaviour present in abnormal form but not sufficiently severe, frequent or marked to meet criteria for 2), or 2 (definite abnormality). ADI-R items that corresponded to SCQ items were recoded as 1 if the original score had been greater than zero, to allow for computation of percentage of items in agreement for each child (i.e., categories 1 and 2 were collapsed). Values ranged from 45% to 85%, with average agreement of 70.81%. Agreement rose only marginally (71.1%) if ADI-R codes of 0 and 1 were collapsed and contrasted with 2.

ADOS-G. We used the algorithm described by Lord et al. (2000) to identify children scoring above cutoffs for autism or PDDNOS on the social and communication domains. For both diagnoses, the child must score above cutoff on both individual domains, as well as on a composite summed score. Restricted and repetitive behaviours are not incorporated in the diagnostic algorithm, because it is judged that there is insufficient opportunity to observe these in such a short assessment. The authors stress that the ADOS-G is one source of information used in diagnosing autism and related disorders but is not sufficient on its own.

ADOS-G categorisation is shown in Tables 2 and 3. Seven children, six from the PLI group and one from the SLI-T group, had scores above threshold for autism, and a further three (two PLI and one SLI-T) scored above threshold for PDDNOS. The remaining five children from the PLI group and six from the SLI-T group did not meet criteria for an autism spectrum diagnosis.

Relationship between ADI-R, ADOS and previous diagnoses. In this sample of children, agreement between diagnoses based on ADI-R and those based on ADOS-G was poor: $\chi^2(4) = 1.86, p = .762$. The agreement was no better when agreement with ADOS-G was computed using the ADI-R scored on current behaviour: $\chi^2(4) = .93, p = .919$. This disagreement is not entirely unexpected: these diagnostic instruments provide different sources of information – one based on report by parents, who have oppor-

Table 1 Mean (SD) age, matrices, receptive language and MLU scores for children in PLI, SLI-T and control groups: Study 1

Group	N	Age (yr)	Matrices	Receptive	MLU ¹
Language-impaired children scoring > 132 on CCC = SLI-T group	7 boys, 1 girl	9.14 (0.64)	103.0 (10.34)	82.6 (9.18)	7.16 (1.12)
Language-impaired children scoring <= 132 on CCC = PLI group	12 boys, 1 girl	8.21 (0.92)	115.1 (12.87)	90.8 (9.59)	6.59 (0.71)

¹ data missing from one case because of equipment failure.

Table 2 Coding of PLI cases as autistic (a), PDDNOS (p) or unaffected (n) according to ADOS, ADI, and SCQ

Child ID	ADI 4–5 yr	ADI now	SCQ	ADOS	ccc	Autism-related diagnosis from school records
A06	а	а	р	а	122	Autistic spectrum disorder (mild)
A08	а	а	a	а	116	Autistic spectrum disorder
A09	а	n	а	а	112	Autistic spectrum disorder
B06	а	p:CR	p	а	101	Semantic-pragmatic disorder
A11	а	a	a	p	119	Autistic spectrum disorder/Asperger's syndrome
A04	а	а	а	n	121	Autistic spectrum disorder
B03	p:SC	n	n	а	119	Seen by autistic advisory services but no diagnosis
A07	p:SC	p:SC	а	n	115	Asperger's syndrome
B02	n	n	n	а	119	Deficit in attention, motor control, and perception (DAMP)
B01	n	n	n	р	114	None
B04	n	n	n	n	105	Semantic-pragmatic disorder
B05	n	n	n	n	129	None
B09	n	n	n	n	126	None

Notes:

- 1. The letter part of the ID indicates the child's school.
- 2. For children with a diagnosis of PDDNOS on ADI, letters are used to indicate the domains that are impaired: S = social interaction, C = communication; R = repetitive behaviours.

Table 3 Coding of SLI-T cases as autistic (a), PDDNOS (p) or unaffected (n) according to ADOS, ADI, and SCQ

Child ID	ADI 4–5 yr	ADI now	SCQ	ADOS	CCC	Autism-related diagnosis from school records
A01	a	a	а	n	134	None, but 'a loner', 'liking sameness'
A05	a	n	а	n	136	Autistic spectrum disorder (mild)
C01	p:CR	n	р	n	147	None
B07	p:SC	n	р	р	141	None
A02	p:SC	n	n	n	140	Atypical autism
C02	p:SC	n	р	n	143	None
C03	n	n	n	а	144	None
A16	n	n	n	n	137	None

Notes:

- 1. The letter part of the ID indicates the child's school.
- 2. For children with a diagnosis of PDDNOS on ADI, letters are used to indicate the domains that are impaired: S = social interaction, C = communication; R = repetitive behaviours.

tunity to observe their child in a wide range of contexts, and one based on a 45-minute assessment. Furthermore, we might expect ADOS-G to overdiagnose autistic disorder, insofar as the algorithm does not require the child to show abnormal repetitive behaviours to be categorised as autistic. Because of these differences, it is usually recommended that a diagnosis of autism be based on combined information from both parental interview and current observation, as was the case in the study by Lord et al. (2000).

If we follow that recommendation here, and diagnose autistic disorder or PDDNOS only in those children who meet diagnostic criteria on both ADI-R and ADOS-G, then four children in this sample, all from the PLI group, merit that diagnosis, and a further three (two from the PLI group) meet criteria for PDDNOS. However, it should be pointed out that some authorities would regard it as too stringent to insist that both ADI-R and ADOS-G should converge on the same diagnosis before categorising the child as autistic: autism is generally viewed as a lifelong disorder. Difficulty in classifying children whose clinical picture improved with age was noted as one reason for poor agreement between experts in dis-

tinguishing between autistic disorder and atypical autism in a study by Mahoney et al. (1998).

It is noteworthy that categorisation on the ADI-R relates much more closely to diagnostic information from school records than does categorisation on ADOS-G. Six of the eight children who were coded as autistic on the basis of ADI-R had a diagnosis of autistic spectrum disorder. Two of the six children who were above cutoff on only two domains had an autism-related diagnosis (Asperger syndrome and atypical autism respectively). None of the seven children who were diagnosed as non-autistic on the ADI-R had an autism-related diagnosis.

The letter part of the child's identification code denotes the school attended. Scrutiny of Tables 2 and 3 shows a clear relationship to the child's diagnostic category. On the ADI-R, only one of 11 children from schools B and C met criteria for autism, whereas 7 of 10 from school A did so, including two who were in the SLI-T group.

Discussion

Overall, the results from Study 1 emphasise the heterogeneity of children with communication

problems: many language-impaired children have some pragmatic abnormalities, and some have other features of autism. Some children identified as having PLI on the CCC met diagnostic criteria for autism on both parental report and direct observation, but these were a minority.

It is difficult to come to any firm conclusions about the proportions of children with PLI who do and do not have autistic features, because this was so dependent on educational placement. Because the ADI-R is based on parental report, it is unlikely to be contaminated by bias from school staff, suggesting that these differences are real and reflect differing selection criteria between schools. It seems plausible that as a school gains a reputation for expertise in working with children with complex communication problems, it will increasingly attract pupils with high-functioning autism, especially if the entry criteria do not exclude such cases. Had we based our study solely on school A, it would have been tempting to come to the conclusion that PLI is usually synonymous with PDDNOS. Our results offer a note of caution and stress the importance of sampling children from a range of educational placements when conducting research studies in this field.

Many questions remain about the characteristics of those children with PLI who do not appear to have significant autistic sympomatology. Do these children have a similar profile to other cases of PLI, but at a milder level, or do they have a different constellation of features? We were not able adequately to address these questions in Study 1 because of the small sample size, and therefore decided to extend the sample by recruiting cases from three additional schools. For this second study, we did not have resources to administer the ADI-R to parents, but were able in most cases to gather information about the child's early history using the SCQ. As well as recruiting children with SLI-T and PLI, we studied a small subgroup of children who had a diagnosis of high-functioning autism, so that we could compare their profiles with those of the PLI group.

A group of typically-developing control children was also assessed to establish normal profiles on ADOS-G in this age range. Normally-developing children were included in the 'nonspectrum' sample reported by Lord et al. (2000), but their data were not distinguished from those of children with other developmental disorders, so it is not possible to establish whether the ADOS-G algorithm ever misclassifies typically-developing children.

Study 2

Methods

Participants. Children with language impairments were recruited from three special schools, all of which catered for pupils with a wide range of communica-

tion problems, including SLI and autism. As in Study 1, children with nonverbal ability in the normal range were allocated to SLI-T and PLI groups on the basis of whether or not their CCC pragmatic composite (from teacher or therapist ratings) was above 132. Children were excluded from these groups if they had a preexisting diagnosis of autistic disorder, but diagnostic terms such as 'on the autistic spectrum' were not used to exclude children. Some children who were screened had only mild language impairments, and so an additional criterion was imposed for inclusion in the SLI-T group only, which was that the child had to score at least 1 SD below the normative mean on at least two standardised language assessments. Using these criteria, 11 children with SLI-T and 18 with PLI were recruited. In addition, six children with an existing clinical diagnosis of high-functioning autism (HFA) were assessed.

Eighteen typically-developing control children aged from 6 to 9 years were recruited from state primary schools that were selected as covering a wide range of social backgrounds among their pupils. These children were selected at random from those for whom parental permission for participation was given, except that an excess of boys was selected for comparability with the clinical groups. CCC ratings were available for all but one of these children (14 from teachers and 3 from parents), and were all well above the cutoff of 132 (mean = 154.3, range = 142 to 160).

Assessments. The protocol was identical to that used in Study 1, except that the ADI-R was not included, and a standardised measure of expressive language was obtained from existing data on the Clinical Evaluation of Language Fundamentals – Revised (Semel et al., 1987). The ADOS-G was administered by one of the authors, and cases of uncertainty in coding were resolved by discussion between both authors. Where the full CELF-R had not been administered, we estimated expressive language level from the Recalling Sentences subtest, which is very sensitive to SLI (Conti-Ramsden, Botting, & Faragher, 2001). Parental data from the SCQ were available for 9 of the 11 children with SLI-T and 15 of the 18 with PLI.

Results

Background data. There were no significant age differences among the four groups (see Table 4). There was a main effect of group on Raven's nonverbal score, F(3,49)=4.14, and specific comparisons using Scheffé tests at .05 level indicated that only group SLIT scored significantly below the control group on this measure. On both receptive and expressive language composites, the three clinical groups scored significantly lower than the control group, but did not differ from one another. The overall effect of group on MLU was significant, F(3,47)=5.19, p=.004, and comparisons using Scheffé test indicated that the differences between the control group and the SLI-T and PLI groups reached significance.

ADOS-G diagnoses. As noted above, little is known about the extent to which the ADOS-G algorithm

gives false positive diagnoses in normally-developing children. In the current control sample of 18 children, there were two who scored above the lower threshold on both social and communication scales and on the composite, corresponding to a diagnosis of PDDNOS. These appeared to be cases of misdiagnosis: the parental ratings on the SCQ gave no hint of any autistic features (scores of 1 and 4), and the CCC pragmatic composite was well within normal limits (scores 145 and 156). The examiner's impression was that the first child was anxious and reticent during the ADOS-G; the second never looked at the examiner, and had unusual conversation dominated by line-by-line recall of a film he had seen.

Diagnostic categorisation of the clinical cases according to SCQ and ADOS-G is shown in Table 5. The findings are not dissimilar to those from Study 1. Agreement between SCQ and ADOS-G was poor, $\chi^2(4) = 4.48, p = .344$. Although several children, especially those in the PLI group, were categorised as autistic or PDDNOS on either ADOS-G or SCQ, only one PLI child met criteria for autism on both assessments. In this sample, we found a higher proportion of children than in Study 1 who had no evidence of autistic features on either assessment: this was the case for six of the nine children with SCQ data in the SLI-T group and three of the fifteen in the PLI group. Surprisingly, among children with a pre-existing clinical diagnosis of high-functioning autism, there was a variable picture: only two of the six scored above threshold for autism on both ADOS-G and SCQ.

Analyses of data from combined samples: Study 1 and Study 2

Data from both studies were combined to address two further questions. First, how valid is the CCC as a measure of pragmatic impairment? Second, are there qualitative differences in symptom profiles between children in the PLI group who do and do not have autistic features?

Validity of CCC ratings

Our categorisation of children as pragmatically impaired was based on the CCC. To date, the only evidence for validity of this instrument comes from two studies: one showing pragmatic composite scores relate to clinical subtype in a language-impaired sample (Bishop, 1998), and another showing that the CCC relates coherently to diagnostic status in children attending a neurodevelopmental clinic (Bishop & Baird, 2001). The current study provided a further opportunity to consider validity, by assessing the relationship between CCC and ADOS-G impairment scores. Many of the items assessed in the ADOS-G social and communication scales are conceptually close to pragmatic areas assessed by the CCC, and so we might expect the two assessments to give similar results. The combined sample from both studies (excluding control children) was used to compute the correlation between the total algorithm score (social + communication) on the ADOS-G and the pragmatic composite of the CCC. This was statistically significant (Pearson

Table 4 Mean (SD) age, matrices and MLU scores for children in PLI, SLI-T and control groups: Study 2

Group	N	Age (yr)	Matrices	Receptive language ¹	Expressive language ¹	MLU
SLI-T	10 boys, 1 girl	9.45	97.09	78.35	67.36	7.12
		(1.06)	(11.58)	(7.06)	(12.26)	(1.24)
PLI	15 boys, 3 girls	9.20	102.00	85.13	72.94	7.32
	• •	(1.00)	(13.42)	(13.80)	(14.21)	(1.47)
autistic (HFA)	6 boys	9.44	95.67	77.69	69.50	6.91
, ,	·	(1.41)	(13.88)	(12.26)	(10.21)	(1.01)
control	15 boys, 3 girls	8.56	110.8	105.59	101.47	8.60
	• •	(1.00)	(10.38)	(9.06)	(10.00)	(1.01)

¹Constraints on testing time led to missing data for one control child on receptive language and three control children on expressive language.

Table 5 Numbers from each group with given diagnostic categorisation according to SCO and ADOS-G: children in Study 2

ADOS-G	Unaffected	PDDNOS	Autism		
Unaffected PDDNOS	6 SLI-T, 3 PLI 2 PLI	2 SLI-T, 3 PLI, 1 HFA	3 PLI, 1 HFA 1 SLI-T, 1 PLI, 1 HFA		
Autism	1 PLI, 1 HFA	1 PLI	1 PLI, 2 HFA		

Note: Data on SCQ were not available for two children with SLI-T and three children with PLI (all five unaffected on ADOS-G).

r=-.55, p<.01, N=54). Nevertheless, as can be seen in Figure 1, whereas most children with high ADOS-G impairment scores have a low pragmatic composite on the CCC, the converse is not true: there are many children who have no evidence of autism on ADOS-G but who have pragmatic impairments as assessed by CCC. Furthermore, as can be seen from the figure, the relationship between these current measures of functioning and the SCQ categorisation is weak, consistent with Study 1.

In other work, the CCC has been shown to provide useful information when completed by parents as well as teachers (Bishop & Baird, 2001). We were able to obtain CCC data from 62 parents of children in this study, and their pragmatic composite ratings correlated .47 (p < .01) with those obtained from teachers. Where a child obtains a low teacher rating on CCC but a normal parental rating on the SCQ, one possibility is that the parent is reluctant to endorse ratings of abnormal behaviour. To test this possibility, we looked at parental CCCs for nine of the PLI cases who were non-autistic on the SCQ. In all but one case, the parental rating yielded a CCC pragmatic composite below 133. (For SLI-T cases with normal scores on the SCQ, the comparable figure was 2 out of 8 cases with parental CCC pragmatic composite of 132 or less.) This is reassuring in

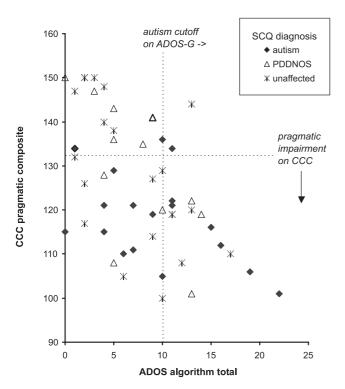


Figure 1 Scatterplot showing the relationship between CCC pragmatic composite and ADOS-G total on Social and Communication algorithm items, children from Study 1 and 2 combined. On ADOS-G, a high score indicates impairment, and the cutoff for PDDNOS is 7 and for autism is 10. On the CCC, a low score indicates impairment, and a pragmatic composite of 132 or less is used to categorise children as having PLI

suggesting that parents, like the professionals, detect pragmatic difficulties in their children, and that low SCQ scores do not mean they are simply underreporting their difficulties.

Specific symptoms in relation to diagnostic subgroup

Our analyses so far have concentrated on the total scores on the ADOS-G that are used in the diagnostic algorithm. Several different aspects of behaviour contribute to these global scores, and many other aspects of behaviour are evaluated in ADOS-G but are not used in the algorithm. This raises the question of whether specific autistic features relate to language subgroup.

To address this question, children were recategorised according to their scores on the diagnostic instruments, according to the criteria shown in Figure 2. The five children from the PLI group who met criteria for autism on both parental report (ADI-R or SCQ) and ADOS-G were moved to the HFA group. The remaining children from the PLI group were subdivided into a PLI-high group, who scored above cutoff for PDDNOS on the ADOS-G, and a PLI-low group, who scored lower than this. The discrepancies that we found between formal assessments and clinical diagnoses created problems in deciding how to treat children who had been categorised as cases of HFA on the basis of clinical diagnosis, but who did not subsequently score above threshold on both ADOS-G and ADI-R. If we had grouped these children with the PLI cases, we would run the risk of overestimating autistic symptomatology in PLI. If we excluded them, we would fail to capture the full range of clinical variation, and might create artificially sharp boundaries between disorders. We therefore retained these children in the HFA group.

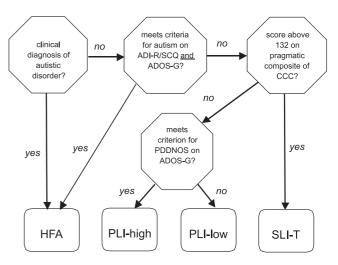


Figure 2 Flowchart showing how children from studies 1 and 2 were recategorised for analysis of combined groups

Because of how they were defined, we expect the PLI-high and PLI-low groups to differ on ADOS-G ratings. However, it is nevertheless interesting to ask how both groups fare on items that do not count in the algorithm, and which items contribute to an above-threshold score in the PLI-high group. In addition, we may ask how the PLI-high compares to the HFA group – do they have milder symptoms overall, or is it rather the case that they are impaired on a different subset of items?

Table 6 shows individual items from the ADOS-G in terms of the percentage of children who obtain an abnormal rating (i.e., score of 1, possible abnormality, or 2, definite abnormality). Meaningful statistical analysis is not possible with such small sample sizes and large numbers of items, so we shall restrict ourselves to describing patterns in the data.

Focusing first on the PLI children who score below the PDDNOS threshold on ADOS-G (PLI-low), over half of them are coded as abnormal on items a2, 'speech abnormalities associated with autism' and a4, 'stereotyped/idiosyncratic words/phrases'. However, these children appeared unimpaired in conversational behaviours and nonverbal communication: proportions rated abnormal on items a5, a7, a8, b1, b2 and b9 were low and comparable to those seen in controls. (Although over half the children were coded as impaired on a6, 'asks for

information', this was also true for the control group, and so this cannot be regarded as abnormal at this age.) Most of these children were rated as unimpaired on imagination/creativity. Although d4, 'excessive interest in objects/topics' and d5, 'compulsions/rituals' were seen in some of these children, the majority had no ratings of abnormality outside the language/social communication domain.

Turning now to the PLI children who scored above the PDDNOS cutoff on ADOS-G (PLI-high), in many respects their profile is similar to that of the children with autism. One item that does distinguish them is d4, 'excessive interest in objects/topics' which was rated abnormal in all but one of those with autism, but only 31% of those in the PLI-high subgroup. Items from scales c and d are not used in the diagnostic algorithm, but there was a trend for the PLIlow subgroup to obtain consistently lower rating than the PLI-high subgroup. It is also noteworthy that more than half the children in the PLI-high subgroup and the autism group were rated as impaired on e1, 'overactivity/agitation' (nearly all were rated as 1, indicating a child who fidgets and moves around), whereas this code was rare in the PLI-low subgroup.

It is interesting also to contrast the ADOS-G profiles of the SLI-T subgroup with the control

Table 6 Percentage of sample rated as abnormal (1 or 2) on ADOS-G items¹

		SLI-T	PLI-low	PLI-high	HFA	Control
item N:		19	13	13	11	18
a1	overall level of language	52	46	46	54	5
a2	speech abnormalities assoc with autism	21	69	92	90	11
аЗ	immediate echolalia	5	15	23	9	0
* a4	stereotyped/idiosyncratic words/phrases	21	53	53	100	11
a5	offers information	5	0	46	36	11
аб	asks for information	52	61	53	45	55
* a7	reporting of events	15	15	61	81	5
* a8	conversation	15	7	61	63	22
* a9	gestures	26	23	46	36	22
* b1	unusual eye contact	36	30	84	90	33
* b2	facial expression	42	38	100	90	44
b4	shared enjoyment in interaction	31	7	38	72	22
b5	empathy	68	38	84	90	27
* b6	insight into social relations	78	61	100	100	38
* b7	quality of social overtures	21	30	100	100	16
* b8	quality of social response	36	23	76	90	16
* b9	amount reciprocal social interaction	10	7	38	54	22
* b10	overall quality of rapport	47	53	92	90	27
c1	imagination/creativity	42	15	46	54	16
d1	unusual sensory interest	10	7	38	63	0
d2	hand/finger mannerisms	0	0	15	9	0
d3	self-injurious behaviour	0	0	7	9	0
d4	excessive interest in objects/topics	10	23	30	90	0
d5	compulsions/rituals	0	15	15	9	0
e1	overactivity/agitation	31	7	53	54	0
e2	tantrums/aggressive	0	0	7	0	0
e3	anxiety	0	7	15	18	11

¹ Item b3, linked language production and nonverbal communication, is excluded, because this cannot be coded if the child is rated abnormal on b1 or b2. Algorithm items are denoted. *PLI-high children met criteria for PDDNOS on ADOS-G; PLI-low children scored below this level.

children. Except for 'overall level of language', these children closely resemble the control group on items from the communication scale, a. Despite their linguistic limitations, they could converse, report events, and use nonverbal means of expression. Although some children were rated as having poor eye contact and limited facial expression, this was true for a similar proportion of control children. Many of these children were coded as impaired on items assessing empathy (b5) and insight into social relations (b7). Coding of these items depends in part on how the child responds to questions about emotions, friendships and marriage, which are coded in relation to the child's nonverbal mental age. Our impression was that many children had difficulties in understanding the questions, even when they were phrased simply. In ADOS-G one does have the opportunity of using a code to indicate the item cannot be evaluated because it presupposes abilities that are not present, and it could be argued that this code should have been adopted for items b5 and b6 for children with poor comprehension. However, this would have meant that many children with PLI and autism would have merited such a code, making it less likely they would meet diagnostic criteria on the algorithm. Given that our goal was to see whether there were children with PLI who were not classifiable as autistic, we decided to err on the side of over- rather than under-diagnosis of autism on the ADOS-G, and to assume that fairly high levels of empathy and insight should be shown by normally intelligent children of this age. This kind of diagnostic problem is not unusual in the field of autism: Mahoney et al. (1998) noted that difficulty of deciding whether a specific behaviour was consistent with the child's developmental level was a major cause of diagnostic disagreement among experienced clinicians. We may also note the relatively high rates of impairment that were observed on some ADOS-G items among our control group: social interactional skills are still developing in middle childhood, and it is clear that some difficulties in relating smoothly to an unfamiliar adult can be expected in children of this age.

A final analysis was conducted using the CCC data to see whether the two PLI subgroups and the autism group had different profiles of scores. No significant effect of group was found when the 10 CCC scales for these three groups were entered in a Manova (Wilks' lambda = 0.63, p = .748), and follow-up univariate Anovas on individual subscales were all nonsignificant.

General discussion

Relationship between PLI and autistic disorder

This study emphasises the complex and varied range of difficulties that can be found among children with

communication impairments. In the introduction, we outlined three possible scenarios. The data reported here allow us to reject the first of these. It is not the case that all children with PLI are misdiagnosed cases of autistic disorder. If we adopt the conventional criterion of basing a diagnosis on a combination of historical and observational information, then only four of the 13 children in the PLI group of Study 1 merited a diagnosis of autistic disorder. Three of these already had a diagnosis of autism spectrum disorder. Thus we did not uncover a high rate of undiagnosed autism among children with PLI. Even if less stringent criteria are used (e.g., meeting criteria on ADI-R only), we find children with PLI who are not autistic (see Tables 2 and 5). In Study 2 we did not have information from the ADI-R, and used instead the SCQ, which agrees well with ADI-R. We found only one of the PLI children who was above threshold for autism on both SCQ and ADOS-G.

The second scenario, which regards PLI as synonymous with PDDNOS, is also contradicted by these data. It is clear that many children in our sample had some autistic features. This was particularly true when one looked at parental report of behaviour at age 4;0 to 5;0 years. However, this was seen in cases of SLI-T as well as cases of PLI. Furthermore, in both studies there were children in the PLI group who had no indications of autistic disorder, either on parental interview or on direct observation. This pattern corresponds to the third scenario outlined in the introduction, namely that one can find pragmatic difficulties in children who do not have significant additional autistic symptomatology.

Before accepting that as a valid account of the relationship between PLI and autistic disorder, however, we need to take into account potential sources of error. Despite every effort to enhance reliability by training those administering the procedures, there is bound to be some error of measurement associated with the instruments used here. This can arise both because of unreliability of the person making the ratings, and also because the child's behaviour may not be representative in an observational setting, or a parent's account of their child may be distorted. We have to ask ourselves whether those children who are rated as cases of PLI yet who do not meet criteria for PDDNOS have been misdiagnosed. This seems unlikely for two reasons. First, the CCC data from parents indicated that the pragmatic difficulties identified by the teacher were also picked up by the parents, and that the parents were not simply failing to recognise or describe developmental problems in their child. Second, none of the Study 1 children with PLI who were rated as non-autistic on the ADI-R had any record of an autism-related diagnosis - in sharp contrast to the other children in the PLI group.

By combining information from ADOS-G in studies 1 and 2 we were able to get a better impression of the characteristics of non-autistic children with PLI.

These tended to be sociable, talkative children who used nonverbal as well as verbal communication, but who produced stereotyped language with abnormal (often exaggerated) intonation and prosody. In some cases, their conversation focused on topics of particular interest, but nevertheless they were coded as having good reciprocal social interaction, rather than talking without regard for the interlocutor. In general, these children did not show nonverbal repetitive behaviours characteristic of autism. We might have expected these children to have a different profile on the CCC subscales when compared with other cases of pragmatic impairment, but this was not so, perhaps because of the low reliability of the individual CCC subscales (see Bishop, 1998).

In both Study 1 and Study 2, ADOS-G sometimes categorised children with language impairment as autistic, even though they did not have significant autistic symptomatology on parental report. This result contrasts with that obtained in a pilot study by Noterdaeme, Kurz, Mildenberger, Sitter, and Amorosa (1999), who used an earlier version of ADOS to compare eight children with autism and eight with receptive language disorder. In their study, only one of the eight non-autistic children scored above threshold on the language/communication scale and one on the social interaction scale. (It is unclear if this was the same child.) To some extent this could reflect the different criteria used in the old and new versions of ADOS. It is also possible that children with pragmatic difficulties were not included in the languageimpaired sample in the Noterdaeme et al. study.

It should be stressed that although we have subdivided children into groups for analytic purposes, the findings of these studies have reinforced our view that there are no sharp dividing lines between SLI and PLI on the one hand, and PLI and autistic disorder on the other. Minor changes in the criteria used to categorise cases would lead to several children changing category because the boundaries between these disorders are so imprecise. The idea of a continuum, with no clear boundaries, between autism and PDDNOS has been put forward by Lord et al. (2000) and Mahoney et al. (1998); our data suggest that boundaries between SLI and PDDNOS are also fuzzy. The model proposed by Bishop (2000), which adopts a dimensional rather than categorical approach to communicative disorders, seems best to capture the variability seen here.

Clinical implications: 1. Assessment

In this study we used the CCC to identify cases of PLI. It should be borne in mind that the agreement between parent and professional pragmatic composite ratings on CCC is far from perfect (.47 in both the study by Bishop & Baird, 2001, and the current data), and the inter-rater reliability for individual subscales is not high. On the other hand, as seen in Figure 1, the CCC scores do detect pragmatic im-

pairments that relate to the social and communicative abnormalities assessed by ADOS-G. Overall, this research supports the utility of the CCC as a screening device for pragmatic problems, but it should be borne in mind that scores obtained with this checklist lack precision, particularly when the subscales are used. Where the CCC could be useful is in identifying when a language-impaired child should be referred for more detailed assessment of social and communicative functioning.

The SCQ is a new screening instrument that is quick and easy to administer to parents, and which enables one to identify children scoring above cutoffs for autistic disorder and PDDNOS. In a validation study by Berument et al. (1999), the SCQ had been administered after the ADI-R. This raised the question of whether good agreement between these two diagnostic procedures reflected the fact that parents had been exposed to a long interview about autism before filling in the questionnaire, which may have familiarised them with the content of the SCQ and helped them to interpret questions appropriately. In our study, agreement remained high, even though parents were given the SCQ in advance of the interview. Our data support the validity of the SCQ when given to parents who are unfamiliar with autism diagnostic procedures.

The lack of agreement between ADOS-G and ADI-R diagnoses was surprising, but may in part reflect the unusual nature of the sample studied here, which focused specifically on children who provide diagnostic challenges. Lord et al. (2000) noted that the ADOS-G is highly efficient at categorising children whose clinical diagnoses place them as definitely autistic or nonautistic, but it is less good at accurately identifying cases of PDDNOS with milder symptoms - the algorithm frequently classifies these children as either autistic or non-autistic. Furthermore, the authors of the ADOS-G and ADI-R have stressed that these procedures provide complementary information, and should not be regarded as alternative approaches to diagnosing autism. We are not aware of other published studies examining the relationship between the two assessments, but the data reported here emphasise that direct observation and parental interview can give a very different picture for some children.

Clinical implications: 2. Diagnostic issues

The results obtained here emphasise the continuities between autistic disorder and language impairment, but they do not support the notion that all children with pragmatic problems have autism or PDDNOS. If a child is found to have pragmatic difficulties, this should prompt the clinician to consider the possibility of autistic disorder. Undoubtedly, there are children in whom autistic disorder goes unrecognised, and where oddities in the use of language may be the first clue to the existence of broader

impairments affecting social interaction and interests. However, it is not helpful to equate pragmatic impairment with autism or autistic spectrum disorder. Those diagnoses should be made on the basis of the whole clinical picture, rather than on the basis of communicative impairment alone. We predict that, as research on this topic advances, we will find that pragmatic impairment is a symptom that can have a range of different causes.

Perhaps because it is widely believed that autism is a life-long disorder, relatively little attention has been paid to the diagnostic difficulties that arise when a child's clinical picture changes substantially with age. Such cases were not unusual in this study, with the child scoring well above the cutoff for autism on parental report of behaviour at 4–5 years, but appearing much less impaired at the time of study. To minimise diagnostic disagreements, classification systems need to give clear guidelines on how to categorise such cases.

Acknowledgements

The research reported here was funded by a grant from the Wellcome Trust awarded to Dorothy Bishop. We would like to thank the staff and pupils at Rose Hill Language Resource Base, Oxford; The Link Primary School, Croydon; Gap House School, Broadstairs; Blossom House School, Wimbledon; Thomas Fairchild School, London; West Kidlington County Primary School, Oxford; Stockham Primary School, Wantage; Sir Thomas Abney School, London; Jenny Hammond School, Leytonstone; and John Horniman School, Worthing. John Horniman is an I CAN school. I CAN is the national educational charity for children with speech and language impairments. We also extend our appreciation to all of the children and parents who participated in this study. We thank Jodie Simon for her help with data collection, Kate Nation for discussion and comments on the manuscript, and Professor Michael Rutter for making a pre-publication version of the Autism Screening Questionnaire available to us.

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Manuscript accepted 23 April 2002