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The CAST (Childhood Asperger Syndrome Test)

Preliminary development of a UK screen for mainstream primary-school-age children

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ABSTRACT The article describes a pilot and follow-up study of the preliminary development of a new tool to screen for Asperger syndrome (AS) and related social and communication conditions (the Childhood Asperger Syndrome Test, CAST) in children aged 4–11 years, in a non-clinical setting. In the pilot study, parents of 13 children with AS and of 37 typically developing children completed the CAST. There were significant differences between the AS and typical sample means. The pilot was used to establish preliminary cut-off scores for the CAST. In the main study, parents of 1150 primary-school-age children were sent the CAST, and 174 took part in the full data analysis. Results suggest that compared with other tools currently available, the CAST may be useful for identifying children at risk for AS and related conditions, in a mainstream non-clinical sample. Further research is ongoing.

KEYWORDS *Asperger syndrome; autism spectrum; epidemiology; screening*

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Background

Classic autism is now routinely identified by the age of 3 years (Howlin and Moore, 1997), and can be identified by as young as 18 months of age (Baird et al., 2000; Baron-Cohen et al., 1996). However, other conditions on the autism spectrum are not as easily identified, even though the prevalence of autism spectrum conditions may be around 60 per 10,000 (Baird

et al., 2000; Scott et al., in press). Part of this difficulty may be due to the broad range of presentation of features of social and communication difficulty in children across the broader autism spectrum. Indeed, our understanding and clinical definitions of Asperger syndrome and other broader pervasive developmental disorders remain somewhat vague and uncertain (e.g. Gagnon et al., 1997; Kugler, 1998; Volkmar, 1998). Recent research suggests the possibility of a 'broader phenotype' of the autism spectrum, with overlaps between autism, pervasive developmental disorders, language disorders, social anxiety problems, and other developmental difficulties in aspects of social communication (Bolton et al., 1994). However, to date this overlap has not been explored in any detail, and presentation of developmental ranges of difficulty in these areas in the general population has not been conducted.

Outside classic autism, identification of the broader spectrum remains relatively poor. The average age for diagnosis of Asperger syndrome (AS), a 'higher-functioning'[1] presentation of the autism spectrum, is currently 11 years of age (Howlin and Moore, 1997). Even worse, many individuals with AS are not identified until their teens or adulthood (Klin and Volkmar, 1997). Considering that autism spectrum conditions typically have an onset in infancy (DSM-IV: American Psychiatric Association, 1994), the delay in diagnosis for conditions like AS means that these individuals are not receiving the appropriate intervention and support at the earliest age. As a result, many of them struggle through their early years, being bullied or ostracized at school, and may develop depression or become suicidal (Howlin, 2000). Research has also suggested that there may be a high risk for associated psychopathology (such as antisocial, disruptive or anxious behaviour) in individuals with AS (Tonge et al., 1999), which could be addressed with the right educational and environmental modifications (e.g. Bregman and Gerdtz, 1997). There is thus a real need to be able to identify children who are experiencing difficulties educationally and socially, who may be failing to meet their full potential, and who may have AS, at a much younger age than is currently the norm (Howlin and Moore, 1997).

There are very few instruments available at present which screen specifically for AS. Howlin (2000) reviewed the existing literature on screeners for autism spectrum conditions including Asperger syndrome, and the reader is directed there for a fuller review. In brief, the only Asperger-specific screening tool developed and validated to date is the Asperger Syndrome Screening Questionnaire (ASSQ: Ehlers et al., 1999). However, the ASSQ has so far only been developed for use with clinical populations, and the authors suggest that generalizations of the tool should be limited to clinical settings. The ASSQ has established cut-off scores for both parent and teacher ratings of the child's presentations of behaviour,

giving varying rates of true and false positives (children who score as AS and who really do have AS, versus children who score as AS but who do not have AS). For parent ratings, the optimal ASSQ cut-off score derived was 19, giving a true positive rate of 62 percent (false positive 10 percent). Teacher ratings had an optimal cut-off of 22, leading to a slightly better true positive rate of 70 percent (false positive 9 percent).

Two other tools with some level of validation are as follows. The first is the Social Communication Questionnaire (SCQ, previously the Autism Spectrum Questionnaire or ASQ: Kazak-Berument et al., 1999), which has been developed to differentiate PDD from non-PDD children in a clinical sample but which, like the ASSQ, has not been validated on a non-clinical population. Indeed, the authors suggest that it would not be a good screening tool for use at a population level (Bailey, 2001). The SCQ does not differentiate AS from other autism spectrum conditions, or differentiate between different 'points' on the autism spectrum.

The second is the Pervasive Developmental Disorders Questionnaire (PDDQ: Baird et al., 2000), which has been developed and tested with a younger sample of children (age 5 years), and asks developmentally appropriate questions for that age. The PDDQ has advantages in that it addresses the broader autism spectrum, but its specificity does not appear to be well developed as yet. Whilst the PDDQ has been piloted with 40 children already diagnosed with AS, and 37 of those children (92.5 percent) failed five or more of the key items, its sampling in a broader population has limitations. Out of a sample of 7766 5-year-old children, 63 scored above cut-off on the PDDQ. Assessments were conducted on 29 of those children, with 11 (37.9 percent) meeting criteria for autism or pervasive developmental disorder (Baird et al., 2000). The PDDQ may suffer from being a very brief screen, with only 18 questions, of which nine are AS-relevant.

The National Screening Committee (1998) recommends that screening for identification of as yet unidentified cases should only be conducted where it can be shown that earlier identification coupled with treatment or intervention has some beneficial outcome on that population. Additionally, the NSC recommends that screening tools should strive for as high a level of sensitivity, specificity and positive predictive value as possible. That is, a tool should identify as many of the true cases as possible in a population, without picking up too many non-cases (those who score positive on the screen but are later shown *not* to have the specified condition), and it should be the case that the likelihood of having the specified condition if one is positive on the screen is high. It is certainly felt that there is a need for development of a UK screener for AS and the broader autism spectrum, particularly as this is the area where there is the greatest current shortage of knowledge coupled with increasing demand (Howlin, 2000), and early

identification and intervention are thought likely to be beneficial (Howlin and Moore, 1997). Development of a tool that can identify possible cases of AS and broader autism spectrum with good levels of specificity, sensitivity and positive predictive value is therefore important. However, development of screening instruments is not without serious ethical considerations. Whilst there is general consensus that it is desirable to identify developmental disorders in childhood as early as possible (Baird et al., 2001; Robinson, 1998), and that early identification linked with appropriate intervention may improve outcome (e.g. Dawson and Osterling, 1997), this benefit needs to be balanced with concerns over the potential identification of disorder in children whose parents may be unaware there is any problem. Similarly, the implications of identification of less severe or broader difficulties in children, which are of valid interest for research purposes but may not warrant specialist educational support services, need to be carefully considered. The issues surrounding screening have been well reviewed in a recent paper by Baird et al. (in press).

These issues notwithstanding, the current ongoing study of social and communication development in primary-school-age children was set up to explore the differences between children who present with difficulties in these areas and those who do not, and to identify those few children who have severe enough difficulties to require diagnosis and support. Additionally, later exploration of psychological differences (cognitive, linguistic, theory of mind etc.) between children with and without social and communication development difficulties, and who fall within different ranges of presentation, is planned. Full ethics approval was obtained for the study, details of which are set out in the methods sections.

The ongoing study has several aims: (1) earlier identification of children who may have AS or related social communication difficulties; (2) an exploration of the educational and psychological needs of the identified children; (3) a better understanding of the differences and similarities between those children who meet criteria for AS or other autism spectrum conditions with associated educational and psychological difficulties, and those children who have 'borderline' problems in social interaction and communication but who do not have severe educational and psychological problems; (4) a thorough epidemiological exploration of the presentation, environmental, educational and familial factors pertaining to these children; and (5) development of a useful UK-based screening tool for AS and related conditions.

The present report describes early findings in relation to (1) and (5) above, outlining a pilot study and preliminary development of a brief parental questionnaire called the Childhood Asperger Syndrome Test (CAST). As the name suggests, this was designed to screen for cases of AS

and related social and communication difficulties in mainstream primary-school-age children (4–11 years) in the UK.

Pilot study

Participants

The participants were 13 children already diagnosed with Asperger syndrome (AS) or autism (age 3–9 years, mean 6:11, SD 1:11), and 37 normally developing control children (age 6–9 years, mean 6:7, SD 0:7).

Screen

The screening instrument being developed is the Childhood Asperger Syndrome Test (CAST). It is based on a variety of behavioural descriptions of the ICD-10 (World Health Organization, 1993) and DSM-IV core features of the autism spectrum (social impairments, communication impairments and repetitive or stereotyped behaviours). Some items in the CAST were based on items appearing in two other screening tools: the Pervasive Developmental Disorders Questionnaire (PDDQ: Baird et al., 2000) and the Asperger Syndrome Screening Questionnaire (ASSQ: Ehlers et al., 1999).[2] The PDDQ and the ASSQ were not considered appropriate tools for screening of AS in primary-school-age children for the reasons outlined in the background – namely that the ASSQ has only been validated on a clinical sample, and the PDDQ is itself in very early stages of development and has not been designed to focus on Asperger syndrome.

The AS-relevant questions in the CAST were designed to cover as wide a range of behaviours as possible, so as to facilitate detecting the high-functioning end of the autism spectrum. The CAST has 37 items in total, of which 31 are key items contributing to a child's total score. The remaining six items are control questions on general development and these are not scored. The six control items are items 3, 4, 12, 22, 26 and 33. The maximum a child can score is 31. The CAST is shown in Appendix 1.

Procedure

The CAST was completed by the parents of 13 children with an existing diagnosis of AS, and by the parents of 37 normally developing children aged 6–9 years attending a mainstream primary school outside the region. Parents were informed that we were developing a new screening tool to identify possible cases of AS and related social communication difficulty in primary-school-age children, and that their input would help us establish provisional cut-off scores and understanding of 'typical' scoring on the CAST. Additionally, for the pilot stage parents were invited to complete the CAST only if there were no special needs requirements

reported for their child, because at this stage we were interested in AS versus clinically typical children for simple establishment of cut-off and typical means.

Aims

To establish preliminary random sample scores; to check if apparently normally developing children score in a different range to a sample of children with AS or high-functioning autism (HFA).

Results

Table 1 shows the number of children in each group scoring at or above each point on the CAST. The mean score for the clinical sample was 21.08 (SD 5.51), range 15–31. The mean score for the 37 controls was 4.73 (SD 3.57), range 0–13. A one-way ANOVA shows these differences to be highly significant ($F(1, 48) = 150.13$, $p < 0.0001$).

Question validity

We also conducted analyses on the question responses by each group. Percentages of participants in each group who scored positive on each individual question were calculated, and chi-square analyses were conducted to explore the differences. Table 2 shows the performance by each group per question.

Taking a criterion of discriminating questions having a 20 percent or greater response in the AS/autism spectrum group, only four questions show no significant differences in response between the normal and AS/autism spectrum groups: question 6 ('Does s/he appear to notice unusual details that others miss?'), chi-square = 1.61, 1 d.f., $p = 0.20$; question 7 ('Does s/he tend to take things literally?'), chi-square = 3.47, 1 d.f., $p = 0.06$ (although this result almost reaches significance); question 9 ('Does s/he like to do things over and over again, in the same way all the time?'), chi-square = 1.52, 1 d.f., $p = 0.22$; and question 30 ('Does s/he sometimes say "you" or "s/he" when s/he means "I"?'), chi-square = 0.76, 1 d.f., $p = 0.38$. The differences in responses between the AS and autism spectrum children and the normally developing controls are substantial for the remainder of the questions. However, some questions had low positive response rates from both groups. This is particularly true of question 30 ('Does s/he sometimes say "you" or "s/he" when s/he means "I"?'), and suggests that the non-significant difference there, at least, may be due to floor effects.

Table 1 **Number of children scoring at or above each point on the CAST (pilot study)**

Total CAST score	*Number (and %) of children with AS*	*Number (and %) of control children*
0	13 (100)	37 (100)
1	13 (100)	34 (92)
2	13 (100)	31 (86)
3	13 (100)	29 (73)
4	13 (100)	22 (64)
5	13 (100)	15 (35)
6	13 (100)	10 (27)
7	13 (100)	6 (16)
8	13 (100)	6 (16)
9	13 (100)	6 (16)
10	13 (100)	5 (14)
11	13 (100)	4 (11)
12	13 (100)	4 (11)
13	13 (100)	3 (8)
14	13 (100)	0
15	13 (100)	0
16	10 (77)	0
17	10 (77)	0
18	9 (69)	0
19	7 (46)	0
20	6 (38)	0
21	6 (38)	0
22	6 (38)	0
23	5 (31)	0
24	5 (31)	0
25	5 (31)	0
26	5 (31)	0
27	3 (15)	0
28	3 (15)	0
29	3 (15)	0
30	2 (8)	0
31	1 (8)	0

Discussion of pilot study

As can be seen, all of the AS sample scored equal to or greater than 15, whilst none of the controls did so. These results suggested that choosing a provisional cut-off of 15 for our preliminary study would not generate any false positives, or lead to many (if any) cases needing an assessment for a possible social and communication condition, in a typical mainstream

Table 2 **Percentages of AS/autism versus normal controls scoring positive on relevant CAST questions (pilot study)**

	Question number																														
	1	*2*	*5*	*6*	*7*	*8*	*9*	*10*	*11*	*13*	*14*	*15*	*16*	*17*	*18*	*19*	*20*	*21*	*23*	*24*	*25*	*27*	*28*	*29*	*30*	*31*	*32*	*34*	*35*	*36*	*37*
AS % (*n* = 13)	86	38	67	71	81	90	43	90	62	62	43	71	33	29	71	81	43	38	90	76	76	76	52	90	19	76	90	38	76	86	62
Normal % (*n* = 37)	5	3	14	57	57	30	27	5	0	8	11	16	5	3	8	40	3	11	16	8	22	5	3	11	8	38	24	5	14	16	5

p-values for all questions were $p < 0.0001$, with the exceptions of the following:
question 6: chi-square = 1.61, 1 d.f., $p = 0.20$, n.s.
question 7: chi-square = 3.47, 1 d.f., $p = 0.06$, n.s.
question 9: chi-square = 1.52, 1 d.f., $p = 0.22$, n.s.
question 14: chi-square = 7.91, 1 d.f., $p = 0.005$.
question 17: chi-square = 8.45, 1 d.f., $p = 0.004$.
question 19: chi-square = 8.85, 1 d.f., $p = 0.003$.
question 21: chi-square = 6.08, 1 d.f., $p = 0.01$.
question 30: chi-square = 0.76, 1 d.f., $p = 0.38$, n.s.

primary-age population. Neither would it risk generating many (if any) false negatives (i.e. missing too many possible positive cases of AS). Although three of the CAST questions showed no significant differences between the two groups in the pilot study, and one just missed significance, we decided initially to retain all the questions for the main study. This decision was made in part because of the small sample sizes involved in the pilot study. The results show that the majority of the CAST questions differentiate very clearly between the AS/HFA group and the normally developing controls, and it may be that one could safely drop the non-significant questions. This issue would be addressed following the main study.

Main study

Participants

The participants were 199 mainstream primary-school-age children (age 4–11 years, mean 8:1, SD 1:9).

Procedure

The CAST was sent to the parents of 1150 children age 4–11 attending mainstream primary schools in Cambridgeshire. The schools involved were informed of the purpose of the study – that we were developing a potential new tool to screen for possible cases of AS and related social communication difficulties in primary-school-age children – and the CAST was distributed via schools to parents with an accompanying explanatory letter (Appendix 2). Parents were informed that the questionnaire they had received was part of a study exploring social and communication development in primary-school-age children, looking at the differences seen and the difficulties some children have. It was explained that a small percentage of children have severe difficulties in social communication, and that these children might have a condition such as Asperger syndrome. Parents were asked to indicate if they would be willing to be approached by the research team at a later date for face-to-face assessments, and it was made clear that this was not necessarily an indication of a difficulty on their child's part. Ethical agreement for the study was established on the basis that we would indicate to a family if there was a problem and the family were concerned about their child's development, and that the family would be counselled about further action to take as necessary. Children clearly requiring further clinical assessment or intervention were thus linked into appropriate services. The research team has strong links with child clinical services in the area, and these services were readily available when required.

Additionally, the schools involved were visited by the research team,

and the opportunity for scheduling a talk to the staff about AS and related social communication difficulties was presented. As part of the larger ongoing study, teacher information packs about AS and related conditions and how to manage children with these difficulties within the classroom are being devised to be provided to all schools involved in the research who show an interest in the resources.

The CAST was distributed and returned to the team by the schools involved. Four schools took part, located in four distinct geographical and health authority areas. This was to test the CAST with a larger random sample in the general population, in order to assess how many children would score at or above the preliminary cut-off, and how many of these would meet criteria for AS or a related autism spectrum condition. We used 15 as our preliminary cut-off score, because 100 percent of the AS sample in the pilot study scored at or above this point, but none of the normally developing controls did so. This would enable us to test if this cut-off led to high levels of sensitivity and specificity.

In addition, the Social Communication Questionnaire (SCQ: Kazak-Berument et al., 1999), a comparison screening tool, was sent in a second mailing to responding families, with a Freepost system so that replies were mailed directly to the research team. The SCQ has been used to identify possible cases of autism spectrum amongst clinical samples, and is based on the Autism Diagnostic Interview–Revised (ADI–R: Lord et al., 1994), a standardized tool for diagnosing conditions on the autism spectrum. Whilst the SCQ has not been standardized for a non-clinical population we chose it as our comparison screen because it was developed from the ADI–R, and we were using the ADI–R and the Autism Diagnostic Observation Schedule–Generic (ADOS–G: Lord et al., 1999) as our assessment tools to make research diagnoses. Thus, we hypothesized that the SCQ would be based on a similar clinical conception of the autism spectrum as our own. Like the CAST, the cut-off score on the SCQ is 15 (out of a possible maximum of 40). The purpose of distributing the SCQ as well as the CAST was to compare the sensitivity and specificity of the two tools within a larger mainstream primary-school sample. It has been suggested recently (Law et al., 2000) that screening one population with two screening tools is of benefit in the development of a screening measure, as it allows not only comparison of sensitivity and specificity, but also positive predictive value and likelihood ratio (LR: the odds that a given cut-off level will correctly identify a child with the specified difficulty).

Validation

Following receipt of the two screening questionnaires, children who scored at or above cut-off on the CAST alone, at or above cut-off on the SCQ alone,

or at or above cut-off on both instruments, were assessed using the ADOS–G (Lord et al., 1999) or the ADI–R (Lord et al., 1994). Additionally, we assessed those children scoring near cut-off (up to 3 points below) on either screen.

Those children within this sample who had not already received a definitive clinical diagnosis of autism spectrum disorder were assessed by the first author using either the ADI–R or the ADOS–G. In practice, the majority of cases were assessed using the ADOS–G, as the ADI–R takes around 3 hours to complete. All assessments were videotaped with consent. These assessments were not to provide clinical diagnoses, as the ADI–R or ADOS–G used alone cannot provide diagnoses. Rather, the assessments were to establish whether children met research criteria on established and standardized tools for autism spectrum condition. However, as has been outlined earlier, where a child met criteria for an autism spectrum condition on the ADOS–G or ADI–R the family was given feedback about the possibility of requiring further clinical assessment if they wished, and were put in touch with clinical services as appropriate.

Whilst the first author is fully trained in the use of the ADI–R and ADOS–G, reliability was checked on a random sample of cases via ADOS–G consensus meetings with other researchers and clinicians qualified with the tool, but not involved in the present study.

Results

Results at screening stage The response rate from the 1150 families approached through the local primary schools was low. Of 1150 families, we had 199 replies (17.3 percent) for our screen. However, this return rate is not unexpected in postal survey research studies. This was probably due to the CAST being distributed close to the summer break. Of these 199, 25 indicated that they did not wish to take part further in the study. The remaining 174 were sent the SCQ, and replies were received from 139 (79.9 percent).[3]

Table 3 shows the number and percentage of children from the sample of 199 who scored at or above each point on the CAST. Table 4 shows the number and percentage of children scoring at or above each point on the SCQ. Table 5 shows the percentage of children scoring at or above cut-off on both the SCQ and the CAST, or on one but not the other, or scoring near cut-off on either, or scoring below cut-off on either.

As can be seen from Table 3, 6.5 percent of children in this random sample scored at or above the cut-off of 15. Since the percentage of children with a possible autism spectrum condition would not be expected to exceed around 0.6 percent (e.g. Baird et al., 2000), this suggests either that

Table 3 **Number of children (*N* = 199) scoring at or above each point on the CAST**

Total CAST score	*Number (and %) of children*
0	199 (100%)
1	190 (95.5%)
2	175 (87.9%)
3	156 (78.4%)
4	123 (61.8%)
5	103 (51.8%)
6	72 (36.2%)
7	56 (28.1%)
8	50 (25.1%)
9	41 (20.6%)
10	35 (17.6%)
11	30 (15.1%)
12	24 (12.1%)
13	19 (9.5%)
14	15 (7.5%)
15	13 (6.5%)
16	10 (5.0%)
17	7 (3.5%)
18	7 (3.5%)
19	4 (2.0%)
20	2 (1.0%)
21	1 (0.5%)
22	1 (0.5%)
23	1 (0.5%)
24	1 (0.5%)
25	1 (0.5%)
26	0
27	0
28	0
29	0
30	0
31	0

a cut-off of 15 or more may be too low, or that the responders were not a representative sample (i.e. concerned parents were more likely to reply). Table 4 reveals exactly the same pattern, namely 6.5 percent of children scoring at or above 15, and the same arguments apply. Table 5 shows that whilst some of the same children are identified as high scorers on both instruments, there are some who score high on one but not the other.

Some of the children due to be assessed dropped out of the study, or

Table 4 **Number of children (*N* = 139) scoring at or above each point on the SCQ**

Total SCQ score	*Number (and %) of children*
0	139 (100%)
1	123 (88.5%)
2	110 (79.1%)
3	91 (65.5%)
4	78 (56.1%)
5	62 (44.6%)
6	53 (38.1%)
7	49 (35.3%)
8	44 (31.7%)
9	37 (26.6%)
10	26 (18.7%)
11	21 (15.1%)
12	17 (12.2%)
13	15 (10.8%)
14	12 (8.6%)
15	9 (6.5%)
16	7 (5.0%)
17	6 (4.3%)
18	6 (4.3%)
19	5 (3.6%)
20	4 (2.9%)
21	3 (2.2%)
22	2 (1.4%)
23	2 (1.4%)
24	1 (0.7%)
25	0
26	0
27	0
28	0
29	0
30	0
31	0
32	0
33	0
34	0
35	0
36	0
37	0
38	0
39	0
40	0

Table 5 **Percentage of children scoring above different cut-offs on CAST and SCQ separately or together (*N* = 139)**

		CAST		
		15+	*12–14*	*<11*
SCQ	*15+*	2.9%	1.1%	0.6%
	12–14	2.3%	1.1%	0.0%
	<11	1.1%	0.0%	90.8%

were uncontactable owing to having moved from the area, for example. This meant that one child who scored above cut-off on both screens, one child who scored above cut-off on our screen only, and five children who were borderline scorers (i.e. 12 to 14) remained unassessed.

Results at validation stage

Table 6 shows the diagnoses and/or ADOS–G/ADI–R results for the remaining participants. As can be seen, in the total sample of 139 (on whom there were both CAST and SCQ scores), four had pre-existing diagnoses of AS or autism spectrum. All four of these were picked up by both the CAST (scoring above 15) and the SCQ. In addition, 10 more children were assessed who had scored 15 or above on one or the other of the two screening instruments. Four of these met criteria for AS or autism spectrum on the ADOS–G or ADI–R. Three of these four children were identified by the CAST alone, and one was identified by the SCQ alone. Thus, eight children from our sample met criteria for AS or autism spectrum.

Question validity

We rechecked question responses from the main study sample against performance shown by the normally developing children in the pilot sample. Results here confirmed that the majority of the questions in the CAST led to highly significant differences in response between AS or autism spectrum children and non-AS children. In fact, the only statistical difference between the pilot and the preliminary study was that question 7 in the preliminary study demonstrated a significant difference (chi-square = 5.63, 1 d.f., $p = 0.02$), and that question 9 moved nearer to demonstrating a significant difference between groups (chi-square = 3.24, 1 d.f., $p = 0.07$). Both questions 6 and 30 remained non-significant (chi-square = 1.61, 1 d.f., $p = 0.20$, and chi-square = 1.18, 1 d.f., $p = 0.30$, respectively).

Prevalence implications

If we consider these results in relation to the sample size originally contacted of 1150 (being conservative owing to the likelihood of a bias in the 199 responders to the screen), this equates to a prevalence of 70 in 10,000. Whilst this number sounds high, it is in line with recent findings suggesting prevalence rates of around 60 in 10,000 (Baird et al., 2000; Scott et al., in press).

Sensitivity and specificity

This preliminary study did not allow for assessment of all children whose parents responded to the CAST; thus it is not possible to establish precise sensitivity and specificity data. To do so would require knowing the number of children who have AS or related social communication difficulty who did not score above cut-off on the CAST (i.e. the false negatives). However, we can report initial positive predictive value and specificity data on the basis of the results to date, but with the addendum that this assumes that the children reported with AS or related conditions versus those without are correctly classified. Additionally, because the screen includes a section asking for details of existing diagnoses, we know that there were no children with existing AS or autism spectrum that were missed by either the CAST or the SCQ. Thus of the *known* cases of AS or ASD, *none* were misclassified by the screening tools as non-cases.

With a cut-off on our screen of 15, it can be seen that 82 percent of children scoring at or above this point either met criteria for an autism spectrum condition or had a deficit relating to social communication (e.g. language delay or social anxiety disorder). We did not conduct further diagnostic assessments of these children at this point, so we do not know what form the language problems took, for example. The positive predictive value (PPV) of the CAST for AS and related social communication conditions was 0.82, with a specificity of 0.99. This compares with a PPV for the SCQ of 0.75, and a specificity of 0.99, for these conditions.

Looking at only AS and autism spectrum criteria, the CAST correctly identified 87.5 percent of cases. However, 36.4 percent of those scoring above cut-off did not meet criteria for AS or autism spectrum (even though 50 percent of those did meet other social communication difficulty criteria). The PPV for AS and autism spectrum for the CAST was 0.64, with a specificity of 0.98. In comparison, the SCQ correctly identified 62.5 percent of AS or autism spectrum cases, with 37.5 percent of those scoring above cut-off failing to meet criteria (of which 67 percent had other social communication difficulties). The PPV for AS and autism spectrum for the SCQ was 0.63, with a specificity of 0.98.

If the cut-off for the CAST were to be raised to 17 for identification of

Table 6 **Scores and diagnoses for participants**

Subject no.	*CAST score (cut-off = 15)*	*SCQ score (cut-off = 15)*	*ADOS–G/ADI–R result*	*Diagnosis*
2	18	15	Meets ASD criteria	Asperger syndrome (AS)
32	25	18	N/A[a]	Existing autism diagnosis
58	20	19	N/A[a]	Existing AS diagnosis
130	18	24	N/A[a]	Existing AS diagnosis
131	18	20	S[b]	Not ASD
66	15	5	Meets ASD criteria	Asperger syndrome (AS)
79	19	12	Meets ASD criteria	Possible PDD, language delay existing diagnosis
116	15	14	N/A[a]	Existing MR + social anxiety disorder diagnosis
119	19	12+	C and RB[c]	Suspected ASD
150	16	14	Not ASD	Early language delay
191	16	11	Not ASD	None
14	10	16	Not ASD	None
51	13	23	Meets ASD criteria	Possible PDD, STM loss, infant brain damage
185	14	15	Not ASD	LD + dyspraxia
53	14	13	Not ASD	Existing ADHD diagnosis
177	12	14	Not ASD	Early language delay

[a] ADOS–G or ADI–R assessments were not given to those children who already had a clinical diagnosis of autism, Asperger syndrome, or other social communication difficulty.
[b] S = meeting autism criteria for social difficulty on ADI–R algorithm, but not for communication and repetitive/stereotyped behaviours.
[c] C and RB = meeting autism criteria for communication disorder and repetitive/stereotyped behaviours on ADI–R algorithm, but not for social difficulty.

possible AS or autism spectrum cases, the specificity increases to 0.99, with a PPV of 0.86. Sensitivity is likely to be worsened, as this cut-off catches only 75 percent of true cases (as opposed to 87.5 percent with a cut-off of 15), but there are fewer false positives, with only 14.3 percent of those scoring above 17 failing to meet criteria.

Discussion

The aim of this study was the preliminary development of a UK screening tool for Asperger syndrome (AS) and related social communication difficulties in primary-school-age children. Other screens that have been developed in the past either have only been assessed with known clinical populations, and thus may not be relevant to screening in a non-clinical sample, or have had limited success at identifying children at the higher-functioning end of the spectrum. In fact, there have been few tools developed specifically to identify Asperger syndrome (Howlin, 2000).

Results of this study suggest that the CAST (Childhood Asperger Syndrome Test) may be effective at screening for AS and related social communication conditions in primary-school-age (4–11 years) children in the general population. Compared with the SCQ, the CAST was better able to detect in this sample those children at risk for AS and related disorders who had not already received clinical diagnoses. Seven of the eight children (87.5 percent) who met criteria for autism spectrum conditions were identified by the CAST, whilst the SCQ identified five (62.5 percent). This suggests that the SCQ may perhaps not be as suitable either for use with a non-clinical population, or for identifying less clear-cut cases of AS or related conditions (i.e. those children who are being missed at this age by services).[4]

However, it was clear that with a cut-off of 15 the CAST picks up 6.5 percent of the overall sample (as did the SCQ). This cut-off may thus be deemed to be too low if one wishes to concentrate only on AS and autism spectrum conditions, although identification of children at risk for a broader range of social and communication difficulties may be appropriate at this point. Finding an appropriate balance between specificity and sensitivity is of utmost importance, and an issue which this preliminary research cannot fully address. Establishing accurate sensitivity and specificity data will require longer-term research. With the average age of diagnosis for AS and the higher-functioning end of the autism spectrum currently being about 11 years of age (Howlin and Moore, 1997), one would need to re-examine the sample over a minimum of 7 years, in order to establish whether those children who were age 4 when first assessed had been diagnosed with AS or a related condition by around 11 years of age, and how many of those were picked up or were missed by the CAST. The

ongoing study is in the process of gathering data on a further 500+ primary-school-age children screened with the CAST, and it is planned to assess not only the screen positives and those scoring near cut-off, but also a matched sample of low to mid scorers, to establish a more accurate picture regarding sensitivity and specificity. It is also hoped to follow the responders over time so that we can ascertain which children, if any, go on to receive clinical diagnoses relating to social and/or communication difficulties.

It is apparent from the response rates in the present study that there was a likely bias in responder parents – that is, many of the parents who responded to the CAST may well have been those who were concerned about aspects of their child's social and/or communication development. It is possible, therefore, that the distribution of scores on the CAST reported here, and the percentages of children scoring above cut-off, is not representative of a 'normal' distribution. With the current distribution of the CAST in our ongoing study we have attempted to address this by providing more detail in the parental cover letter and specifying the importance of hearing from parents who do not feel there are any difficulties with their child's social and communication development. Preliminary results suggest that this has helped to redress the issue of bias, though further analysis will be needed to confirm whether this is the case. However, possible responder bias does not render the CAST development invalid, depending on the purpose of the screen development. If one assumes that the CAST were to be developed as a universal screen, to be given to the parent of every primary-school-age child in the UK, then a protection against bias would be of great importance. The main purpose of the CAST is, however, to be developed as an early indicator for those children likely to be at risk for AS or related conditions, who are not achieving their educational potential and who have clinical or educational needs. It needs to be developed using a non-clinical sample as these children may not yet be in touch with clinical services, but it is likely that such children will be those for whom there is already parental and/or teacher concerns, or who may be struggling to have their needs recognized, or who are being misclassified as lazy, naughty, disruptive etc. This therefore *assumes* a bias in those for whom the CAST will be most relevant. These preliminary results suggest that the CAST is a useful tool for this purpose.

Results also suggested that the CAST as it stands may benefit from some slight modification. Two of the 31 AS-relevant questions (question 6 and question 30) demonstrated no significant differences in positive response rates between AS/autism spectrum children and non-AS children. It is not clear whether this was due to misinterpretation of the wording of the questions by parents, or to other factors such as floor effects. It is possible that

floor effects explain the non-significance of question 30. Future research is needed to establish the effect of removing or rewording such questions on the overall sensitivity and specificity of the CAST, and the ongoing study is looking at this initially using latent trait analysis of each CAST question in relation to identification of AS and related conditions.

In summary, these preliminary results indicate that the CAST may be an effective tool for the early screening of primary-school-age (4–11 years) children at risk for AS and related conditions, in a non-clinical sample. With ongoing development it could be established as a UK screener for the broader autism spectrum to be used within that population of children who are currently mislabelled as 'naughty' or 'disruptive', and whose educational and personal development is being compromised owing to lack of or delay in established diagnosis.

Appendix 1: the Childhood Asperger Syndrome Test (CAST)

Child's name:..... Age:..... Sex: male/female

Birth order:..... Twin or single birth:.....

Parent/guardian:.....

Parent(s) occupation:.....

Age parent(s) left full-time education:.....

Address:.....

.....

.....

Tel. no.:..... School:.....

Please read the following questions carefully, and circle the appropriate answer. All responses are confidential.

1	Does s/he join in playing games with other children easily?	Yes	No
2	Does s/he come up to you spontaneously for a chat?	Yes	No
3	Was s/he speaking by 2 years old?	Yes	No
4	Does s/he enjoy sports?	Yes	No
5	Is it important to him/her to fit in with the peer group?	Yes	No
6	Does s/he appear to notice unusual details that others miss?	Yes	No
7	Does s/he tend to take things literally?	Yes	No
8	When s/he was 3 years old, did s/he spend a lot of time pretending (e.g. play-acting being a superhero, or holding teddy's tea parties)?	Yes	No
9	Does s/he like to do things over and over again, in the same way all the time?	Yes	No
10	Does s/he find it easy to interact with other children?	Yes	No
11	Can s/he keep a two-way conversation going?	Yes	No
12	Can s/he read appropriately for his/her age?	Yes	No
13	Does s/he mostly have the same interests as his/her peers?	Yes	No

14	Does s/he have an interest which takes up so much time that s/he does little else?	Yes	No
15	Does s/he have friends, rather than just acquaintances?	Yes	No
16	Does s/he often bring you things s/he is interested in to show you?	Yes	No
17	Does s/he enjoy joking around?	Yes	No
18	Does s/he have difficulty understanding the rules for polite behaviour?	Yes	No
19	Does s/he appear to have an unusual memory for details?	Yes	No
20	Is his/her voice unusual (e.g. overly adult, flat, or very monotonous)?	Yes	No
21	Are people important to him/her?	Yes	No
22	Can s/he dress him/herself?	Yes	No
23	Is s/he good at turn-taking in conversation?	Yes	No
24	Does s/he play imaginatively with other children, and engage in role-play?	Yes	No
25	Does s/he often do or say things that are tactless or socially inappropriate?	Yes	No
26	Can s/he count to 50 without leaving out any numbers?	Yes	No
27	Does s/he make normal eye-contact?	Yes	No
28	Does s/he have any unusual and repetitive movements?	Yes	No
29	Is his/her social behaviour very one-sided and always on his/her own terms?	Yes	No
30	Does s/he sometimes say 'you' or 's/he' when s/he means 'I'?	Yes	No
31	Does s/he prefer imaginative activities such as play-acting or story-telling, rather than numbers or lists of facts?	Yes	No
32	Does s/he sometimes lose the listener because of not explaining what s/he is talking about?	Yes	No
33	Can s/he ride a bicycle (even if with stabilizers)?	Yes	No
34	Does s/he try to impose routines on him/herself, or on others, in such a way that it causes problems?	Yes	No
35	Does s/he care how s/he is perceived by the rest of the group?	Yes	No
36	Does s/he often turn conversations to his/her favourite subject rather than following what the other person wants to talk about?	Yes	No
37	Does s/he have odd or unusual phrases?	Yes	No

Special needs section

Please complete as appropriate.

38	Have teachers/health visitors ever expressed any concerns about his/her development?	Yes	No
	If yes, please specify:.....		
39	Has s/he ever been diagnosed with any of the following?:		
	Language delay	Yes	No
	Hyperactivity/attention deficit disorder (ADHD)	Yes	No
	Hearing or visual difficulties	Yes	No
	Autism spectrum condition, inc. Asperger syndrome	Yes	No
	A physical disability	Yes	No
	Other (please specify)	Yes	No

Appendix 2: parental cover letter

QUESTIONNAIRE INFORMATION SHEET

Dear Parent

We would like to invite you to take part in a research study being conducted by the University of Cambridge exploring how social and communication skills develop in primary-school-age children.

Children develop such skills in very different ways. Some children are very outgoing and sociable, others more quiet and reserved. A few children may be very shy. A very small number of children may have difficulties in their social development. This can be for a variety of reasons. For example social anxiety problems may underlie the difficulty in mixing. Very occasionally the difficulties may be due to Asperger syndrome or an autism spectrum condition (conditions where children have significant problems understanding social and emotional situations).

We are interested in exploring the full range of development of social and communication skills in children from the whole population. This research will then help us to better understand when children do have difficulties.

We are inviting parents of children aged 4 to 11 from Cambridge, Huntingdon and Fenland areas to help us. This involves filling in the questionnaire provided, which takes about 10 minutes. A Freepost envelope is provided so you can post this directly to us. In order for us to get a truly representative picture of the range of social communication styles, it is important we receive replies from everyone willing to participate.

After we have received all the questionnaires, we would like to invite around 10 percent of people to take part in more detailed face-to-face assessments to see how accurately our questionnaire works in characterizing social communicative style. Participation with the questionnaire survey does not commit you to helping with these more detailed assessments. We will write to families again asking if they would be willing to help with the second part of the survey, and if you wish you could decline to participate further at that or any other stage. If you have any concerns about your child or if we identify a possible developmental problem, we will be happy to discuss these with you and if you are in agreement arrange for a clinical specialist to see you and advise further.

If you are happy to take part in this study, we would be grateful if you would complete and return the questionnaire(s) in the Freepost envelope provided. All information you supply will be confidential to the research team. We would be interested to receive your questionnaire even if you do not wish to take part in later stages of the project.

You are of course free to withdraw from the study at any stage without providing an explanation, should you wish to do so. Neither participation nor non-participation in this research will affect any treatments or services your child may be receiving or be entitled to receive.

Should you wish to discuss this research further, or have any questions, the Project Coordinator, Dr Fiona Scott, can be contacted on 01223 746113 (fax: 01223 746122; e-mail: fjs25@cam.ac.uk).

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Notes

1 Whilst children with AS are 'higher-functioning' in terms of cognitive and language development, there remain severe difficulties in social interaction,

communication and obsessional, repetitive or routine behaviours which can cause substantial educational and psychological difficulties (e.g. Tonge et al., 1999).

2 Question 2 in the CAST closely matches item 2 in the PDDQ; question 22 in the CAST closely matches item 11 in the PDDQ; question 26 in the CAST is similar to item 1 in the PDDQ; question 29 in the CAST is similar to item 16 in the ASSQ; and finally question 33 in the CAST is similar to item 15 in the PDDQ.

3 For this initial sampling, we did not attempt a second 'reminder' mailshot. However, the ongoing study is utilizing a revised information sheet, and second mailings, to address the issue of responder bias and low response rates. Early indications are that this has worked effectively.

4 Since this preliminary study was completed, AS diagnoses have been confirmed clinically for participants S2 and S66, and they plus participants S51, S79, S116, S131 and S185 are all currently in touch with clinical child services. Parents of the other participants identified in Table 6 have so far chosen not to be involved further with services.

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