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Diagnosing Autism Spectrum Disorders in Pre-school Children Using Two Standardised Assessment Instruments: The ADI-R and the ADOS

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Abstract The reliable diagnosis of Autism/Autism Spectrum Disorder in pre-school children is important for access to early intervention and for accurate ascertainment for research. This paper explores the combined use of two standardised assessment instruments—the Autism Diagnostic Interview Revised (ADI-R) and the Autism Diagnostic Observation Schedule (ADOS)—in a large sample of pre-school children. The children were recruited to research studies, and a 'best estimate' clinical diagnosis reached. The findings show good agreement between the instruments especially for children with core Autism. The instruments appear to have a complementary effect in aiding diagnosis and confirm the importance of a multidisciplinary assessment process with access to information from different sources and settings. The presence of repetitive behaviours during the ADOS appeared of diagnostic significance.

Keywords Clinical diagnosis · Repetitive behaviours · Parent report

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Introduction

Autism is a neurobiological condition, where children experience life-long pervasive difficulties with social interaction and communication, and demonstrate restricted and repetitive behaviours. The broader spectrum around Autism includes individuals across a range of severities, language and intellectual abilities. The prevalence of Autism Spectrum Disorders (ASD) has been reported to be at least 60 per 10,000 children under 6 years of age (e.g. Chakrabarti & Fombonne, 2001, 2005).

To make a diagnosis of Autism/ASD according to ICD-10/DSM-IV-TR (American Psychiatric Association, 2004) requires a multidisciplinary assessment procedure that includes a detailed developmental history and description of current behaviours, assessment of cognitive and language abilities, and observations of functioning in a variety of settings (Le Couteur, 2003; Ozonoff, Goodlin-Jones, & Solomon, 2005). The information then needs to be combined into a consensus opinion. This process can involve the use of standardised instruments in combination with clinical judgment, usually of at least two or more professionals with ASD expertise (Baird et al., 2006; de Bildt et al., 2004). Two of the most widely used diagnostic instruments used for this purpose are the Autism Diagnostic Interview-Revised (ADI-R) (Le Couteur, Lord, & Rutter, 2003; Lord, Rutter, & Le Couteur, 1994) and Autism Diagnostic Observation Schedule (ADOS) (Lord et al., 2000; Lord, Rutter, Di Lavore, & Risi, 2001). The ADI-R is an investigator-based interview designed to provide a framework for the developmental history for a lifetime differential diagnosis of Pervasive Developmental Disorders and information about current functioning for individuals (with a mental age of 2 years or above) from early childhood to adult life (American Psychiatric Association, 1994; World Health Organisation,

1992). It was originally developed to form part of a multidisciplinary research based diagnostic assessment in combination with the ADOS (see methods section for more comprehensive description of both these measures). Currently the ADI-R provides a summary diagnostic algorithm that distinguishes between Autism and not Autism. The ADOS is a play and activity based assessment that provides, through the specification of 'social presses', standard contexts for observation of aspects of social behaviour, communication, play and restricted and repetitive behaviours in individuals (across the ability range) suspected of having a possible ASD. There are four different modules for use with children or adults of different developmental and language levels from no expressive or receptive language through to verbally fluent individuals. Each module takes about 30–45 min to administer. The ADOS summary diagnostic algorithm distinguishes between Autism, ASD and not ASD.

For a diagnosis of core Autism according to ICD-10/DSM-IV-TR, there needs to be evidence of delay or deviation within the first 36 months of life (WHO, 1992). However, parents often suspect difficulties earlier, the average age of first concern being reported as ~19 months (De Giacomo & Fombonne, 1998; Gray & Tonge, 2001). The reported early symptoms of Autism include a lack of joint attention (Charman, 2003), a failure to develop spoken language (McConachie, Le Couteur, & Honey, 2005) and a failure to respond to name (Baranek, 1999).

Despite the early features of this disorder becoming better recognized, delays in diagnosis still occur, for several possible reasons. The particular criteria needed for diagnosis may not be apparent at very young ages, the range of normal variation is great, and symptoms may change or only appear infrequently. There are only a limited range of assessment tools targeted at the pre-school age range (as opposed to screening instruments), potentially impeding assessment, and a lack of local community based specialised professionals and/or resources may delay the progression of diagnosis (Gray & Tonge, 2001). Despite these potential difficulties, diagnoses in the Autism spectrum made in the pre-school period have been shown to be reliable (Charman & Baird, 2002; Cox et al., 1999; Gillberg, Ehlers, Schaumann, & Jakobsson, 1990; Lord, 1995; Moore & Goodson 2003; Stone et al., 1999; Volkmar, Charwarska, & Klin, 2005), and there is emerging evidence that there is much to be gained from an early diagnosis, in order to maximize the benefits of early intervention strategies (Bryson, Rogers, & Fombonne, 2003).

Aims of the Paper

The ADI-R and ADOS were developed to be used in combination to contribute to a valid diagnosis, and are the tools generally recommended for autism research (Tanguay, 2000). Despite this, the levels of agreement between these two assessment tools have only recently been considered. A study of 6–11-year olds (Bishop & Norbury, 2002) found that diagnostic categorisation reached by the ADI-R and the Social Communication Questionnaire (SCQ) (Berument, Rutter, Lord, Pickles, & Bailey, 1999) showed good agreement; however agreement between these tools and the ADOS was less satisfactory. Noterdaeme, Mildenberger, Sitter, and Amorosa (2002) found largely accurate classification of school-age children with a diagnosis of autism or receptive language disorder, using the ADI-R and ADOS. Discrepancies noted were attributed to factors such as parents' under-reporting of early difficulties. Recently, Mazefsky and Oswald (2006) have examined the diagnostic utility of these two measures in a heterogeneous clinical sample of children (aged from 22 months to 8 years) referred to a developmental disorders assessment clinic. The ADI-R and ADOS algorithm classifications had ~75% agreement with the team diagnoses. Discrepancies were usually false positives on the two measures. The paper did not report any direct comparisons between the two instruments. For children and adolescents with intellectual disability, de Bildt et al. (2004) report that the two instruments provide valuable information on social, communicative and stereotyped behaviours especially in individuals with mild or moderate intellectual impairment. The authors conclude that, although overall the agreement between ADI-R and ADOS is 'fair' (kappa of 0.67 for autism in 5–8-year olds but 0.16 for study subjects older than 8 years), the combination of both instruments is the best way to measure PDD. However, they caution both that the diagnosis of PDD remains difficult in very low-functioning children, and that overall the algorithm cut-off scores for both instruments should not be used as absolute criteria. This observation is in keeping with Robertson, Tanguay, L'Ecuyer, Sims, and Waltrip's (1999) observation that the ADI-R and ADOS measure slightly different aspects of manifestations of disorder and that the combination provides a 'conservative approach to diagnosis'. Finally, few studies have considered the use of the two instruments in young children. Ventola et al. (2006) considered the efficiency of ADOS and ADI-R, and found that children screened and assessed at mean age 22 months were unlikely to score above cut-off on the ADI-R repetitive behaviours domain, despite receiving a clinical diagnosis of autism. Given the increasing numbers of toddlers and young pre-schoolers presenting for diagnostic evaluation, it is important to explore the utility of the ADI-R and ADOS in clinical diagnosis.

There were two aims for this study. The first was to investigate the agreement between the ADI-R and ADOS domain scores and the overall diagnostic categorisation

for autism or ASD in a well defined cohort of pre-school children. The second was to consider how a BECD could be reached when the diagnostic algorithm cut off scores for these two standardised instruments do not apparently agree.

Methods

Study Sample

One hundred and one pre-school children aged 24–49 months were assessed with both the ADI-R and ADOS. The cohort was recruited from two previous studies, one an evaluation of a group parent training intervention, and the other a study of the relationship between executive function and autistic symptomatology in very young children (McConachie et al., 2005; Shearer, 2001). The Northern and Yorkshire Multi-Centre Regional Ethical Committee and all relevant Local Ethics Committees approved both studies. Informed consent was obtained from parents for each study. All children were initially identified from within the North East of England by local speech and language therapists and paediatricians as having speech or communication difficulties, or suspected ASD. At the time of recruitment, not all had a firm clinical diagnosis (Honey, McConachie, Randle, Shearer, & Le Couteur, 2006). All of those for whom the question of autism had been raised had either recently concluded or were still undergoing diagnostic assessment.

A 'best-estimate' clinical diagnosis (BECD) was made by the senior authors (ALC, HM) based on all available clinical information across settings, along with the ADI-R, ADOS and all other research assessment information (as reported in McConachie et al., 2005). This procedure is in line with accepted best practice for research assessments (Dunn, 2000). The senior authors are clinicians in the Regional Children's PDD service, and thus had access to additional clinical information and reports about many of the children. Where there was uncertainty, the ADI-R interview schedule was consulted and the ADOS assessment videotape watched to reach a consensus judgement. Forty-nine children were grouped as showing 'Autism' where they met all ICD-10 criteria for a corresponding diagnosis, and 28 were classified as having an 'ASD' where the diagnosis was more likely to be under another heading of pervasive developmental disorder (PDD-NOS, APA, 1994). All children had significant social, communication or behaviour abnormalities identified in their development before 36 months of age. Children defined as 'Other' had all been clinically referred for specific speech or language difficulties, and did not have social communication difficulties consistent with an ASD diagnosis (24).

Diagnostic Assessment Tools

The ADI-R is an investigator-based semi-structured diagnostic interview which was designed to provide a developmental history framework for a lifetime differential diagnosis of Pervasive Developmental Disorders and information about current functioning (over previous 3 months), for individuals with a mental age of 2 years or above) from early childhood to adult life. It is usually undertaken with the parents/main carer of the child, has 111 questions and usually takes about 2–3 hours to complete. Interviewers are intensively trained in its administration, with inter-rater reliability on individual algorithm items ranging from $r = 0.63$ to 0.89. Most items are scored from zero (no impairment with respect to the behavioural definition for each item) to three (severe impairment for the individual and their family), relying on the interviewer to make judgements on the child's behaviour based on the recall of information from parents/carers. The scoring algorithms generated draw on items relating to social interaction, communication and repetitive behaviours, with a scoring cut-off for Autism. Scores are transformed following the protocol in the manual (e.g. 3's become 2's). Lord et al. (1994) reported internal consistency (alpha coefficients) for the domains from 0.69 to 0.95. The ADI-R algorithm diagnosis is not a clinical diagnosis but the result of combining the coded information from the interview. To date there is no algorithm cut-off within the ADI-R for ASD, and the published interview (Le Couteur et al., 2003) has not been modified for use with young pre-school children. For this study of pre-school children the current behaviour scores were used for the algorithm.

The ADOS is a standardized semi-structured observational play and activity assessment of the child, and usually lasts about 40 min. As with the ADI-R, intensive training in the administration of the ADOS is required. Inter-rater reliability of items is good ($\kappa \geq 0.6$). The exception is the coding of some items such as some repetitive behaviours and sensory abnormalities (Lord et al., 2000). The module for administration is chosen according to the developmental and language level of the child. Modules 1 and 2 are appropriate for young pre-school children. There are ten sets of materials and play activities in Module one appropriate for children with no speech or single words, and 14 activities in Module two suitable for more fluent young children with phrase speech, from which around 30 behaviours are coded on a 3 or 4 point scale (as for the ADI-R). Selected algorithm items relating to social interaction and communication are then entered into an algorithm. These scores are transformed following the protocol in the manual (e.g. 3's become 2's) for the diagnostic algorithms. To obtain an ADOS classification of Autism or ASD, an individual's scores must meet the separate

cut-offs for both the communication and social domains and the cut-off for the summation of the two. Repetitive behaviours are recorded and coded as part of the clinical observations but do not contribute to the ADOS summary algorithm. Internal consistency is high: Alpha coefficients are 0.86–0.91 for the social domain (across modules), 0.74–0.84 for communication, and 0.63–0.65 for repetitive behaviours (modules 1 and 2) (Lord et al., 2000).

Assessment of Ability

The Mullen Scales of Early Learning (Mullen, 1995) are a standardised assessment of young children's ability, used in many studies of children with ASD (Lord et al., 1994). Age equivalents are reported for Visual Reception (a non-verbal scale), Receptive Language and Expressive Language (McConachie, Randle, Hammal, & Le Couteur, 2005).

Procedure

Children were usually seen in their own homes. The informant(s) for the ADI-R was one or both of the child's biological parent(s), usually the mother. All the assessments (apart from 3 of the ADOS assessments) used in this study were undertaken by one of two research associates trained to the accepted standards (Le Couteur et al., 2003; Lord et al, 2000) and blind to other clinical information about the children. The ADOS was video-recorded for later rating. Throughout both previous studies regular inter-rater reliability checks were made in joint group coding sessions. Both research associates maintained a consistent minimum level of 75% agreement across all ADOS items and scale points.

Statistical Analysis

In order to examine the degree of agreement, correlations were calculated between the ADI-R and ADOS social interaction and communication algorithm domain scores using Pearson's correlation coefficients. The total score for the four ADOS items relating to repetitive behaviours was correlated with the repetitive behaviours domain score from the ADI-R algorithm.

The levels of agreement between the ADI-R and ADOS were also compared (by calculating Kappa statistics; Cohen, 1988) with respect to overall diagnostic categorisation of Autism, and when looking at attainment of cut-off for each domain of the algorithm.

In order to incorporate the ADI-R into contributing to the diagnostic category of ASD, a revised use of the ADI-R algorithm was employed, i.e. if the age of onset was recorded as before 36 months and if two of the three domain cut-offs (social interaction, communication or repetitive behaviours) were reached, an ASD study descriptor was applied, as has been used in a previous study (Bishop & Norbury, 2002). Agreement with the two diagnostic tools was examined separately in relation to the three categories of BECD.

Results

Table 1 shows data on age, gender, age equivalents for three Mullen scales (Visual Reception, Receptive and Expressive Language) together with the profile of scores from the ADI-R and ADOS for the 101 study subjects. All subjects have been grouped according to the best-estimate clinical diagnosis (BECD) of Autism, ASD or Other (i.e. a non-ASD diagnosis). All but two subjects received Module 1 of the ADOS.

The social interaction total scores in the ADI-R and ADOS had a correlation coefficient of $r = 0.71$, indicating that as the total social domain score in the ADI-R increased the total social interaction score in the ADOS was also likely to increase. The correlation between the communication domains was also relatively strong, with a coefficient of $r = 0.64$. There was a weaker association between the ADI-R and ADOS repetitive behaviour scores with a correlation coefficient of $r = 0.51$.

The levels of agreement between the ADI-R and ADOS (Table 2) for above both the social and the communication algorithm cut-offs scores for Autism are moderate. The kappa level for a best estimate diagnosis of Autism is good, but moderate for a spectrum diagnosis.

In order to explore the relationship between BECD and the instrument classifications, the distribution of subjects according to their diagnosis on the different instruments is presented in Table 3, and illustrative individuals presented in Tables 4 and 5.

Best Estimate Clinical Diagnosis of Autism

The joint agreement of the ADI-R and ADOS with a BECD of **Autism** was 67% (33/49). Six children met criteria on the ADI-R but not the ADOS. However, of these, five did meet criteria for ASD on the ADOS. The sixth child met criteria for ASD separately for communication (2) and social interaction (4); however, the joint algorithm cut-off score is 7, which is why the child appears below spectrum cut-off on ADOS on Table 3 (marked a).

Eight children met the criteria for Autism on the ADOS but not on all domains of the ADI-R. The one child (Table 3, marked b) who appears below spectrum cut-off

Table 1 Sample characteristics by BECD group

		BECD[b]		
		Autism (n = 49)	ASD (n = 28)	Other (n = 24)
Gender		41 M/8 F	22 M/6 F	18 M/6 F
AGE (mths)		35.6 (7.0)	37.8 (6.0)	37.6 (5.7)
MULLEN	Visual reception	21.5 (7.6)	25.9 (7.7)	30.9 (7.2)
Age equiv (mths)	Receptive language	15.8 (8.7)	22.2 (6.5)	28.5 (6.1)
	Expressive language	16.2 (8.9)	19.8 (6.9)	24.7 (6.9)
ADI-R	Social	17.5 (4.6)	7.9 (5.2)	2.8 (3.5)
Mean (SD)	Communication	11.1 (2.8)	6.2 (3.3)	3.5 (3.0)
	Repetitive	5.1 (1.4)	3.7 (2.2)	1.3 (1.3)
	Total[a]	33.7 (6.6)	17.8 (7.8)	7.6 (5.5)
ADOS	Social	10.4 (2.7)	4.7 (3.3)	0.3 (0.6)
Mean (SD)	Communication	5.6 (1.5)	2.4 (1.9)	0.7 (0.9)
	Repetitive	3.8 (1.4)	3.3 (1.3)	0.6 (0.8)
	Total[a]	19.8 (4.2)	10.4 (5.2)	1.6 (1.7)

[a] Social, communication and repetitive behaviours

[b] There are significant differences between the BECD groups for all assessment scores

Table 2 Levels of agreement and kappa statistics between the ADI-R and ADOS

Outcome	Agreement ADI-R versus ADOS (%)	Kappa statistic
Autism social interaction cut-off	78	0.56
Autism communication cut-off	74	0.48
Above Autism cut-off/below Autism cut-off	81	0.62
Above spectrum cut-off/below spectrum cut-off	78	0.54

on the ADI-R was below on both social interaction and repetitive behaviours by only one point.

Only two subjects were found to score below criteria for Autism on both the ADI-R and ADOS when the BECD indicated a diagnosis of Autism. However, both of these had scores above cut-off for ASD on the ADOS, and had scores above cut-off on Social Interaction and on Repetitive Behaviours on ADI-R (Table 3, marked c; see Table 4). Subject 2 had age appropriate language skills, though Subject 1 used less language on formal testing than reported by parents. Both children used language effectively for communication and to establish joint attention at the time of assessment, which may have affected how parents reported their skills. The BECD of Autism was arrived at because of the degree of impairment shown by the children in everyday settings such as playgroup or nursery.

In summary, the agreement between ADI-R and ADOS ratings, and a BECD of Autism appeared generally sound and for those individuals where there were differences between the BECD and the instruments' classifications, the differences in scores were marginal and there was consistent clinical evidence of difficulties across a range of settings and over time.

Best Estimate Clinical Diagnosis of ASD

The joint agreement of the ADI-R and ADOS with a BECD of **ASD** was only 14% (4/28). Four children met criteria for Autism on the ADOS but were not above cut-off on the

Table 3 Diagnosis based on BECD, ADI-R and ADOS

BECD	ADI-R	ADOS		
		Below spectrum cut-off	Above ASD cut-off	Above Autism cut-off
Autism	Below spectrum cut-off	0	0	1[b]
	Above ASD cut-off	0	2[c]	7
	Above autism cut-off	1[a]	5	**33**
ASD	Below spectrum cut-off	6[f]	8	4[d]
	Above ASD cut-off	5	**4**	**0**
	Above autism cut-off	1[e]	**0**	**0**
Other	Below spectrum cut-off	**22**	0	0
	Above ASD cut-off	2[g]	0	0
	Above autism cut-off	0	0	0

a, b, c, d, e, f, g = see text for explanation

c = see also Table 4

d = see also Table 5

Bold text indicates the expected area of joint agreement for each BECD

Table 4 Vignettes of two children with a best estimate clinical diagnosis of Autism, but not meeting criteria on all domains of ADI-R and ADOS

ADI-R domain scores[a]	Subject 1 scores	Subject 2 scores
Communication (8)	5	4
	First word 'apple' at 2–2.5 years. Currently uses a few two-word and three-word phrases. Attempts pointing to express interest. No pretend play	Single words from age 10 months. Currently uses a wide vocabulary and is able to point successfully to express interest. Said to have a 'fabulous imagination'
Social interaction (10)	10	13
	No eye-contact until 3 years 3 months. Finds everything funny. Sometimes mixes with others. Prefers older children, especially girls	Described as affectionate but does not like to mix with other children. Continues to have difficulty with direct eye contact. Tantrums if he cannot do as he wants
Repetitive behaviours (3)	4	6
	Upset by one particular advert on TV, but now watches warily. Repetitive drumming behaviour on all flat surfaces	Shows an unusual fear of hats. Is very sensitive to noise and puts his hands over his ears. Compulsively touches objects in his surroundings
ADOS		
Communication (4)	3	2
	Babbled jargon to mother, with gestures. A few single words	Used lots of appropriate language
Social interaction (7)	7	7
	Made lots of requests. Eye contact not well integrated but did initiate joint attention	Showed some appropriate eye contact but very active and easily distressed
Social–communication (12)	10	9
Repetitive behaviours	3	2
	Distracted during ADOS by repetitive interest in videos on shelf	Hands nearly always moving
Mullen scales of early learning		
Chronological age	3 years 6 months	3 years 10 months
Visual reception age equiv.	1 year 10 months	3 years 5 months
Receptive Language age eq.	1 year 6 months	3 years 11 months
Expressive Language age eq	1 year 3 months	3 years 0 month
Notes	Attends specialist language unit. Parents first concerned when he was 2–2.5 years because of slow development of speech	Attends mainstream school. Teachers describe difficulties coping with his behaviour especially during lunch break.
		Parents first concerned at 9 months because of constant crying and lack of interest in toys

[a] Autism cut-off score in brackets

ADI-R (Table 3, marked d; see Table 5). Three attended mainstream school and one a specialist language class. Subject 3 illustrates the complexity of the diagnostic process, and clinical judgements to be made. During the ADOS, carried out at home, he repeatedly requested that the television be switched back on. This fixed idea may have increased (i.e. made more abnormal) the scoring of the ADOS on social interaction on that occasion (when assessed again 4 months later, his social–communication algorithm score was below the Autism cut-off and above the ASD cut-off, in line with his BECD). He is an only child of relatively 'elderly' parents, and described by them as a 'happy lad' with no behaviour problems. His easy temperament and his parents' lack of comparison may offer a partial explanation for the ADI scores not meeting algorithm cut-off in any domain. For subject 4, the ADI-R ratings may again represent under-reporting by parents; their child was only 2 years old at the time of the assessment, and they stated the belief that his lack of spoken language is 'his only problem'. Subjects 5 and 6 are both girls. For both families the early social developmental histories did not alert professionals to the girls' complex developmental difficulties. Subject 5 was described by her parents as placid and compliant whilst Subject 6 showed definite behavioural difficulties described as severe temper tantrums. For both these girls the diagnostic picture became clearer over the next year, having perhaps been masked by their particular temperamental characteristics.

Table 5 Vignettes of four children with a best estimate clinical diagnosis of ASD, but meeting criteria for Autism on ADOS

ADI-R domain scores[a]	Subject 3 scores	Subject 4 scores	Subject 5 scores	Subject 6 scores
Communication (8)	3	5	6	6
	Despite using single words from age 12 months he then lost vocabulary of over 20 words. Useful speech returned aged 3 years. At age 2 years was able to point to express interest	Had single words from age 10 months, then stopped using these from 20 months. Uses words now and again	Placid child. Delayed use of single words. Phrase speech 39 months.No protodeclarative pointing, few gestures.unable to nod head for yes	No initial speech delay but limited use. Tends to be demanding with odd intonation and repetitive questions & statements
Social interaction (10)	5	4	5	7
	Has always been able to use brief eye contact	Eye contact and social smiling appropriate. Shy but interested in other children	Cheerful demeanour. Brief eye contact but no understanding of others' distress	Always on the go. Difficult with other children. Parents report behaviour difficulties and temper tantrums
Repetitive behaviours (3)	2	2	2	4
			Some mild unusual sensory interests and brief hand mannerisms reported	Difficulties with changes to routine at home and with routes to school/shops. Some complex whole body mannerisms
ADOS domain scores[b]				
Communication (2)	6	6	6	4
	Speech consisted mainly of babbled phrases and echoing	Few vocalisations, no pointing or gestures	Used single words but not clearly directed. Intonation showed little variation. Pushed adult's hand to object	Little spontaneous speech and poor co-operation- tends to try to refuse activities
Social interaction (4)	8	10	8	10
	Showed occasional eye-contact	Difficult to get eye contact. Gave objects but did not request	Limited eye contact. No requests, Put her finger in her ears but compliant	Tantrums and disruptive repetitive behaviour interfered with tasks
Social-communication (7)	14	16	14	14
Repetitive behaviours	5	3	4	4
	Showed overreaction to sounds and repeatedly requested TV on	Held on to one toy throughout	Definite hand mannerisms and some sensory exploration of toys	Resistance to change and other repetitive interests affected task performance in addition to poor concentration
Notes	Boy aged 4 years 1 month. Attends mainstream school with support teacher. Parents first concerned when he was over 2 years of age because of loss of speech	Boy aged 2 years 8 months. Attends specialist language group at Child Development Centre. Parents first concerned around 20 months of age because of loss of speech	Girl aged 3 years 3 months. Attends mainstream school with full-time support. Parents concerned at 18 months because of delay in speech and walking	Girl aged 3 years 0 months. Attends mainstream school with support. No speech delay; parents concerned about unco-operative behaviour and lack of social use of speech

[a] Autism cut-off score in brackets

[b] ASD cut-off score in brackets

One child (Table 3, marked e) showed markedly discrepant ADI-R and ADOS ratings. The assessor commented that he was somewhat of a contradiction. During the ADOS he seemed able to engage and to be social, but without a definite 'social press' he did not take any initiative. Social difficulties were reported in a variety of situations and thus scored on the ADI-R items. In addition his ADOS repetitive behaviour score was 3. When assessed 1 year later, he met the ADOS criterion for autism on social interaction, for ASD on communication, and had a total rating of four on repetitive behaviours. This probably means that the interactions with the experimenter during the ADOS, on the first occasion, were unrepresentative.

There were six children below spectrum cut-off on both ADI-R and ADOS, with a BECD of ASD (Table 3, marked f). It was noticeable that all these children had significant history of repetitive behaviours as reported by parents during the ADI-R developmental history and as rated on ADOS (2 or more), and/or an ADI-R social interaction score of 8 or 9, i.e. just below the cut-off score of 10. In one case the clinical diagnosis had been influenced strongly by additional observations made in a nursery setting, where the child's social difficulties and self-absorbed and stereotyped behaviours were particularly evident.

In summary, there is evidence that some of the apparent disagreement between the instruments and between the two instrument classifications and BECD arises from scores that are just below cut-off.

Best Estimate Clinical Diagnosis of 'Other'

Those subjects given a BECD of **Other** all had scores below algorithm cut-offs on both instruments, with the exception of two, giving a joint agreement of 92%. The two children (Table 3, marked g) both scored above cut-off on two out of three ADI-R domains.

Finally, we examined the repetitive behaviours scores for the children with BECD of **ASD** and **Other** ($n = 52$). Using a cut-off value of 2 or 3 on the ADI-R repetitive behaviours domain did not significantly predict a BECD of ASD (respectively, $\chi^2 = 1.77$, $p = 0.18$; $\chi^2 = 1.30$, $p = 0.25$). However, a value of 2 or more for ADOS repetitive behaviours was highly predictive of ASD ($\chi^2 = 13.52$, $p = 0.00$). That is, all but one of the children with ADOS social/communication scores above ASD cut-off had a rating of 2 or more on ADOS repetitive behaviours.

Discussion

The findings from this study of pre-school children demonstrated good agreement between the ADI-R and ADOS, especially in relation to core Autism. The results demonstrated a similar level of agreement between the instruments in pre-school children to that reported in school aged children for autism (74–75%: Bishop & Norbury, 2002; Conti-Ramsden, Simkin, & Botting, 2006; de Bildt et al., 2004), The correlation co-efficient for total domain scores across the two instruments was greatest for the social domain. For these young children the developmental history would have been relatively fresh for parents, rather than having to recall information about the past, as would be the case for parents of older individuals. This meant that the social behaviours as reported by parents and as observed during the ADOS and other assessments were contemporaneous. Parental report of communication was also strongly related to rating of observed communication, but the association was weaker for repetitive behaviours. This may have been as a consequence of the limited time period for observation during the administration of the ADOS, or that the algorithm threshold for repetitive behaviours in young pre-school children may need to be reviewed (Ventola et al., 2006).

Further, the findings demonstrated that the ADI-R and ADOS have a complementary effect in aiding diagnosis, i.e. when their results were taken together they provided a greater level of diagnostic clarity but used as a single assessment instrument in isolation each could over or under score particular behaviours. This study of pre-school children confirms the importance of a multidisciplinary assessment and the need to consider the developmental history alongside direct assessments of current level of functioning and evidence of co-morbidity. However, there is a complexity to the diagnostic process especially for the 'just subthreshold' cases. The best agreement between ADI-R, ADOS and BECD was for the non-ASD or 'other' group.

Turning to the 'disagreement' between ADI-R and ADOS algorithm scores: These were greatest for children with a BECD of ASD. For the 14 children with none or only one above algorithm cut-off score for ASD on ADI-R, and/or ADOS algorithm scores, the clinical information available in addition to the research assessments suggested patterns of three common influences. First, there could have been a tendency for some parents/carers to underreport unusual features in communication in young children with good language skills i.e. useful expressive speech (Noterdaeme et al., 2002). Second, the clinicians' BECD was influenced by the findings from the direct assessments of a child's skills and difficulties and other clinical observations in settings such as nursery, thus taking into account the impact of potentially unpredictable, noisy and unstructured social environments upon children's function and behaviour. Third, the analysis showed that the presence of repetitive behaviours observed during the course of the ADOS, contributed to the consideration of an ASD diagnosis.

To date, the diagnostic algorithm for the ADOS has not included repetitive behaviours, i.e. it is a composite of communication and social interaction ratings. This was because the assessment is a snapshot in time, and repetitive behaviours may not be triggered in a one-to-one play session with an attentive adult. However, the examples presented in this paper suggest that where repetitive behaviours are noted, they may be considered significant. This recommendation is consistent with the recently published revised ADOS algorithms (Gotham, Risi, Pickles, & Lord, 2006). Further, with respect to the ADI-R our findings concur with other recent publications (Mazefsky & Oswald, 2006; Risi et al., 2006) that further research is needed, to address the potential use of the ADI-R for identifying ASD and to generate joint revised algorithms for pre-school children and across the lifespan.

Limitations

This cohort has been carefully described in previous papers (Honey et al., 2006; McConachie et al., 2005). The children were recruited to two research studies and as such may not be representative of all pre-school children with suspected ASD from the local population. The research group may for example be more advantaged economically than the families that declined to take part. In the light of recent findings of the potential impact of parents' socio–economic status or ethnicity on local case identification (Baird et al., 2006; Mandell, Listerud, Levy, & Pinto-Martin, 2002), this possible trend will inevitably impact on the generalisability of the study findings but may also have other as yet unknown influences on the research diagnostic process. The senior authors agreed the best estimate clinical diagnoses (BECD) based on all available information. However, although it is accepted that expert clinical diagnosis (including using information gathered from the use of systematic diagnostic assessment tools) is predictive of a stable diagnosis, there are as yet no specific guidelines for the development of algorithms for individuals at different developmental stages across the lifespan.

Clinical Implications

Finally, Filipek et al. (1999) and de Bildt et al. (2004) have commented on the time taken both to train in the use of and to administer the ADI-R and ADOS diagnostic tools in the context of limited clinical resources. This study used these assessment tools in a research context and provided summary assessment reports for the family and responsible clinicians with the prior consent of the children's parents. Diagnostic evaluation for ASDs and all complex developmental disorders is time consuming (Wing, 1996). For both clinical and research practice, with the increasing emphasis on early detection and early intervention, and as the diagnostic criteria have broadened from a narrow definition of autism to the broader Autism spectrum, it is important that a clear assessment framework is used. Standardised structured instruments (interviews and observations) have both advantages and limitations when used in clinical practice. However, one of the principal advantages is the opportunity to gather clinically relevant information in a systematic and comparable fashion. For example, change over time is hard to predict, but where a query of possible ASD is raised in a child with language delay, there is good evidence that the characteristics may become clearer in middle childhood (Michelotti, Charman, Slonims, & Baird, 2002; Miniscalco, Nygren, Hagberg, Kadesjö, & Gillberg, 2006). Therefore, in complex cases, especially where there is diagnostic uncertainty, detailed recording of behaviour with specific instruments such as ADI-R and ADOS is valuable in the diagnostic process.

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