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What is the association between ADI-R scores and final diagnosis of autism in an all IQ adult autism diagnostic service?

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Abstract

Purpose – The diagnosis of autism in adults often involves the use of tools recommended by NICE guidance but which are validated in children. The purpose of the paper is to establish the strength of the association between the Autism Diagnostic Interview-Revised (ADI-R) scores and the final clinical outcome in an all intellectual quotients adult autism diagnostic service and to establish if this in any way relates with gender and intellectual ability.

Design/methodology/approach – The sample includes referrals to Leeds Autism Diagnostic Service in 2015 that received a clinical outcome. Sensitivity, specificity and positive and negative predictive values were calculated to evaluate ADI-R and final clinical outcomes. Logistic regression model was used to predict the effect of the scores in all the domains of ADI-R and the two-way interactions with gender and intellectual ability.

Findings – ADI-R has a high sensitivity and low specificity and is useful to rule out the presence of autism, but if used alone, it can over diagnose. Restricted stereotyped behaviours are the strongest predictor for autism and suggests that the threshold should be increased to enhance its specificity.

Research limitations/implications – This is a single site study with small effect size, so results may not be replicable. It supports the combined use of ADI-R and Autism Diagnostic Observation Schedule and suggests increasing ADI-R cut-offs to increase the specificity.

Practical implications – The clinical team may consider piloting a modified ADI-R as suggested by the results.

Originality/value – To the authors' knowledge this is the only study of ADI-R in an adult population of all intellectual abilities.

Keywords Autism, Assessment, Diagnosis, Adults, Intellectual disability, ADI-R

Paper type Research paper

The diagnosis of autism in adults can be challenging and requires comprehensive, multidisciplinary assessments. Research has identified with good evidence, both the reliability and validity of the Autism Diagnostic Interview-Revised (ADI-R) as a diagnostic tool for autism diagnosis (Lord et al., 1997; Sappok et al., 2013).

ADI-R is a structured interview, usually involving the parent of the subject, which provides information relevant to the diagnosis of autism. The interviewer scores the information provided and feeds these scores into an algorithm that is scored in three domains:

1. A – qualitative abnormalities in reciprocal social interaction (cut-off ¼10);

2. B – qualitative abnormalities in communication (cut-off ¼7/8, verbal and non-verbal); and

3. C – restricted, repetitive, stereotyped behaviours (cut-off ¼3).

Elevated scores indicate problematic behaviour in a particular area and when the scores meet or exceed the cut-offs in all of the three areas and if onset is before three years of age, a diagnosis of autism may be made (Le Couteur et al., 2003). Although the ADI-R is commonly used, autism is a clinical diagnosis and this should only be made in combination with a clinical assessment of the person (Risi et al., 2006).

In the last decade, the standardised instrument has been used all over the world and translated into other languages including Brazilian, Portuguese (Becker et al., 2012), Polish (Chojnicka and Płoski, 2012) and Japanese (Kenji et al., 2013) with good effect.

Much of the existing research seems to be based on studies in children and adolescents, particularly those with intellectual disability (ID) (Papanikalou et al., 2009; Bildt et al., 2004). Papanikolau et al. studied 77 Greek children with a range of intellectual abilities using the Autism Observation Schedule-Generic (ADOS-G) and ADI-R and both instruments had excellent sensitivity (0.88) and satisfactory specificity (0.69) for the diagnosis of autism. The combination of ADI-R and Autism Diagnostic Observation Schedule (ADOS) was also reported as clinically relevant by Bildt et al. (2004) in the diagnosis of children and adolescents with ID with a reported percentage agreement of 63.6 per cent. Meilleur and Fombonne (2009) suggested that a more severe autistic symptom profile was indicated by ADI-R in children who regressed compared with those who did not.

There is a wide knowledge gap in relation to the use of ADI-R in the adult population across all intellectual abilities – much of the adult literature focusing on an additional and associated ID. Sappok et al. (2013) found the ADOS and ADI-R to be valuable diagnostic tools for a clinical sample of 79 adults with ID who were suspected of having ASD. In 68 per cent of testable cases, the ADOS was over-inclusive (specificity 45 per cent) but highly sensitive (100 per cent). In the ADI-R, the feasibility was 37 per cent, with a sensitivity of 88 per cent and a specificity of 80 per cent. Although they concluded that the ADI-R and ADOS were valuable, they suggested adjustments to the settings and tasks to improve feasibility and specificity. Saemundsen et al. (2010) used the ADI-R to research the prevalence of autism in individuals with ID in Reykjavik, Iceland and by so doing increased the number of cases of autism known to the service by a factor of two.

There is a wide gender disparity in the prevalence of autism with a high proportion of males in varying ratios of 3.3:1 to 5.5:1 as compared to females (Fombonne et al., 2011; Baird et al., 2006). Some studies have recommended the need to consider the extent to which females on the autistic spectrum present differently from males and suggest that this would then have implications for the

systems, instruments and processes used for diagnosis and for the types of interventions offered (Gould and Ashton-Smith, 2011).

Diagnosing autism for the first time in adulthood can be difficult for a variety of reasons – not least the differing presentations of males and females (Lai and Baron-Cohen, 2015). Wilson et al. (2016) suggested that sex had an impact on the diagnosis of adults with suspected autism as the sexes showed different presentations of the symptoms and Frazier et al. (2014) suggested a phenotype for female autism with associated under-identification. This persisting suggestion that girls are often under diagnosed and less likely to meet the diagnostic criteria (Dworzynski et al., 2012) has led to some researchers advocating for the development of instruments standardised for females (Goldman, 2013) A recent study by Lai et al. (2017) considered the well-established clinician observation that women with autism may "camouflage" their social communication difficulties. The study not only highlighted but also quantified this interesting aspect of "camouflaging", showing high scores in females when compared to age matched men with no significant correlation with age or intellectual quotients (IQ).

In order to study the variations in the adult population exclusively and across all intellectual abilities, we conducted a retrospective, post-diagnostic study at a specialist autism diagnostic service in Leeds. Our aim was to review and analyse all processed referrals in the service in a one-year period (2015), to establish the strength of the association between the ADI-R scores and the final outcome of the clinical decision meeting. In addition, we also sought to explore if the association between ADI-R and final outcome is different in people with and without ID and in males and females.

Method

Subjects

The sample included all adults above the age of 18 who were referred for a clinical assessment at Leeds Autism Diagnostic Service (LADS) during the year 2015, irrespective of ethnicity, gender and intellectual ability. The referral source included primary care, secondary mental health services, specialist ID services and self-referrals. Only those referrals that have progressed through the entire diagnostic pathway, i.e. have received a clinical outcome in the year 2015, have been included in this study. The subjects are therefore adults who received a decision as to whether or not they met diagnostic criteria for Autism Spectrum Disorder 299.00 (F84.0) – referred to in this paper as autism. The diagnosis is determined using the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5). Out of a total of 175 completed referrals, there were 120 males and 55 females.

LADS does not provide services for psychometric assessments including IQ assessments; some patients had documented IQ's at the time of referral.

Setting

LADS is an adult-specific specialist autism diagnostic service, which accepts referrals from all sources, amongst the local adult population. LADS was piloted in 2011 and has developed since (Davidson et al., 2015; Stansfield et al., 2017) This is a specialist autism diagnostic service catering to people across the IQ range. The service now provides not only assessment and diagnosis of autism but also consultancy support if a diagnosis of autism is made.

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In 2014 (the first year of permanent funding), LADS received 264 referrals out of which only 29 per cent received a diagnosis of autism. This data followed a consistent pattern of diagnostic rate (29-32 per cent) since the origin of the service in 2011 as a pilot, so we were keen to understand the clinical significance of the large proportion of undiagnosed cases.

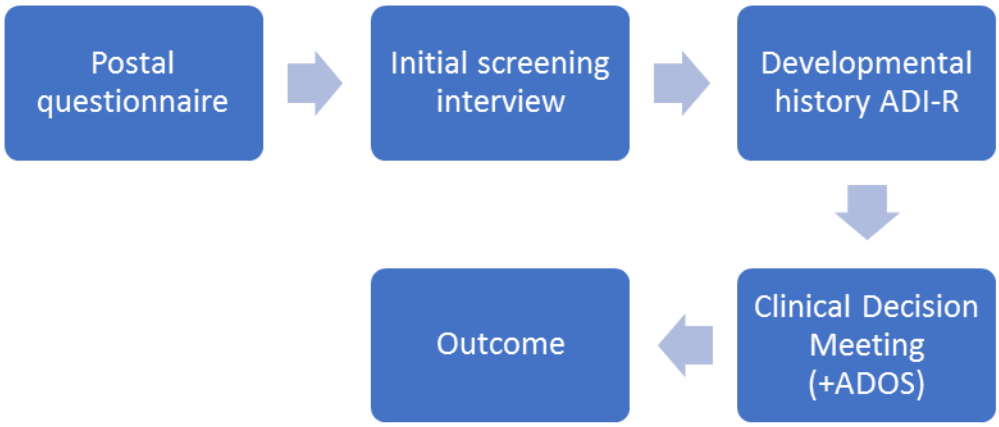
Ethical approval

The study did not require NHS research ethics review but did go through Health Research Authority approval which was issued on the 17 January 2017.

Measures and procedure

The diagnostic pathway in LADS includes an initial questionnaire pack, followed consecutively by an initial screening interview, developmental assessment (using ADI-R) and clinical decision meeting (incorporating ADOS) (Lord et al., 2000) that ultimately results in a person receiving a clinical outcome, i.e. meets criteria for autism or does not meet criteria for autism (as per DSM-5). In a small number of cases, it is not possible to arrive at a clinical consensus and these are categorised as being "Unable to diagnose" (Figure 1).

Figure 1: Diagnostic pathway



Prior to the initial screening assessment, a postal pack including the Cambridge Behaviour scale and the Adult Asperger Assessment (AAA) questionnaire is sent out to the person who has been referred. The pack also includes a form requesting demographic and clinical information as well as information about co-morbidity and risk.

At the initial screening (usually undertaken by a nurse or doctor) these forms are analysed and additional information is collected to inform the assessment as well as detect any additional mental health or physical health issue that may need urgent attention or referral. This is followed by the ADI-R assessment. A relative (parent or carer) who knows the patient's childhood developmental history is interviewed by staff who have attained reliability to use the ADI-R. Although the default assessment

tool is ADI-R, Diagnostic Interview for Social and Communication Disorders (DISCO) (Leekam et al., 2002) is used occasionally. It is not uncommon for the parental history to be unavailable. In such circumstances, the team looks for additional sources of information from other relatives, family friends or employers as well as reports from education, health and social care if possible.

Finally, a clinical decision meeting, involving a multidisciplinary team (psychiatrist, autism nurse or speech and language therapist – but always at least two different disciplines with at least one psychiatrist) is conducted where all the information collected up to this point is considered and a further clinical assessment is conducted using the ADOS.

At the beginning of the clinic, before the person enters the room, the multidisciplinary team (MDT) reviews information obtained from the process thus far. This includes all the information from:

- the initial postal questionnaire and AAA scores;
- the initial screening assessment;
- the qualitative clinical information obtained from the developmental interview (ADI-R), which is typed up in full after each assessment, together with the associated scores; and
- any additional collateral information including school reports, reviews of child health/social service/Child and Adolescent Mental Health and psychiatric notes, questionnaires from relatives or employers, films or photographs from childhood and any additional telephone interviews that have been conducted.

This enables the team to form a multidimensional clinical and developmental perspective, prior to assessing the patient. The various autism diagnostic assessments using standardised instruments are conducted by members of the MDT who have been appropriately trained and attained reliability to use these.

The person is then invited into the clinical decisions meeting and the ADOS is conducted by one member of the MDT, while the others observe and evaluate. Finally, the patient undergoes a brief psychiatric assessment including any outstanding relevant questions before they are asked to return to the waiting area while the MDT deliberates on a clinical outcome.

Despite the extensive collection of collateral and developmental information, the process is not always straightforward and although a definitive clinical outcome is possible in most cases, some cases cause a lot of debate within the team before a clinical decision is reached.

Ultimately, the final decision is a clinical one, but takes into account all of the above information. As stated above, although a definitive clinical decision is possible in most cases, the study excludes the small number of cases where a unanimous decision was not possible.

Sample population

The sample studied consists of a total of 175 people who went through the whole diagnostic pathway and achieved a clinical outcome in 2015. Barring gender and a statement about intellectual ability, no other demographic information was collated for the purpose of this study.

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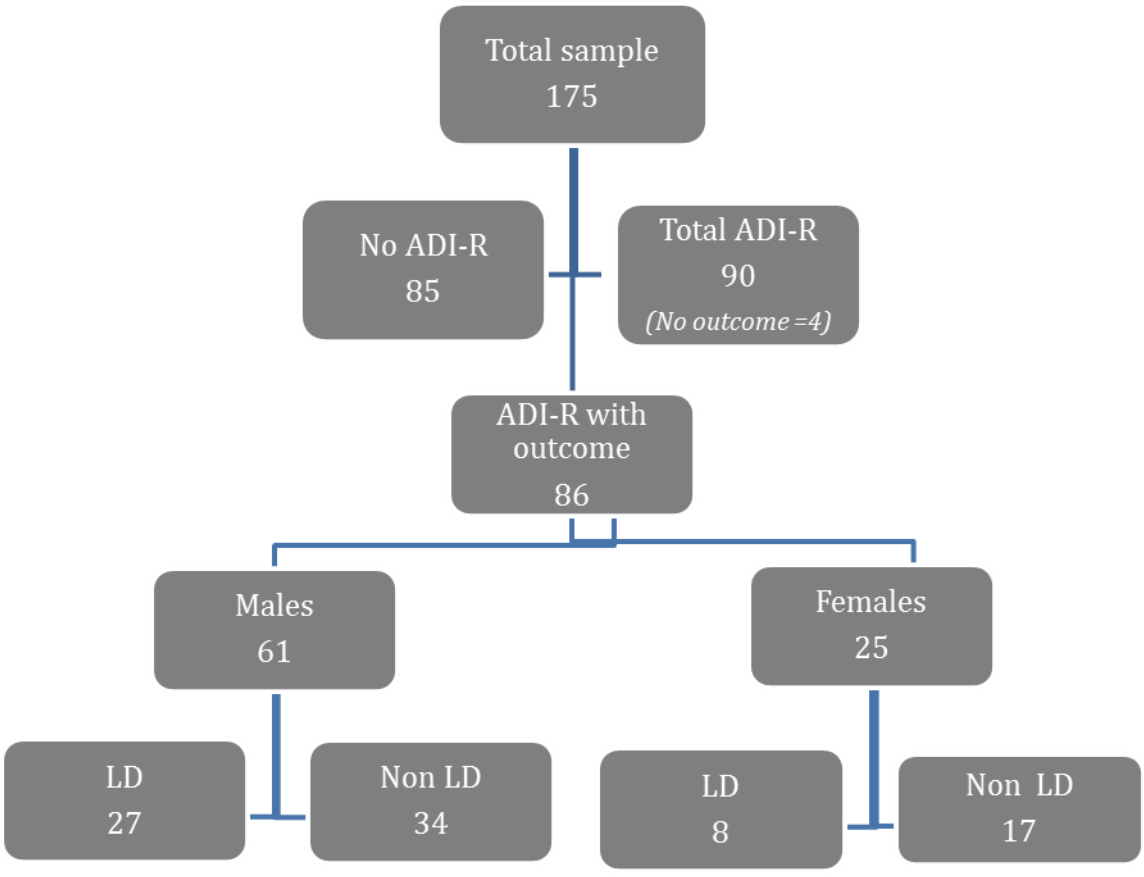
It was possible to complete a developmental history using the ADI-R in 90 cases. Of these, four did not receive any clinical outcome.

There were 85 people that did not have an ADI-R. They progressed to the clinical decisions meeting and of these, five did not receive any clinical outcome. In those that had an ADI-R and received an outcome, 61 were male and 25 were female, a ratio of 2.4:1.

Of the 61 males who had an ADI-R and received an outcome, 27 had an ID. Of the 25 females who had an ADI-R and received an outcome, eight had an ID (Figure 2).

Figure 2: Sample population flow chart



Statistical analysis

Sensitivity, specificity and positive and negative predictive values were calculated to evaluate the ADI-R outcomes compared to the diagnosis of autism, using the established cut-offs.

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Sensitivity

Sensitivity in this study is the number of true positives (ADI-R and the clinical decisions meeting both give diagnosis of autism) divided by the total number of people with autism in the population (true positives plus false negatives, i.e. ADI-R says NOT autism but clinical decisions meeting diagnoses autism).

Specificity

Specificity in this instance is the ADI-R's ability to correctly identify people who do not have autism. That is, the proportion of neurotypical people who will not score on the ADI (true negative rate). It is important to note that the ADI-R information informs the decision of the final clinical decision meeting.

Logistic regression

This is a predictive analysis exploring the effects of the individual scores of ADI-R (reciprocal social interaction (A), communication (B), stereotyped behaviours (C)), sex and ID. It explores the two-way relationship between sex and ID with the three domain scores of ADI-R (A, B, C) to determine if there is any relationship. Finally, receiver operating characteristic curves (ROC) were used to calculate the area under the curve (AUC).

A logistic regression model with high-discrimination ability will have high sensitivity and high specificity simultaneously. In such cases the ROC curve goes close to the top left corner of the plot. If the model has no discriminatory ability then the ROC curve will be a 45 degree diagonal line.

The discrimination ability of a model can be summarised as the AUC, ranging from 1 (perfect discrimination) to 0.5 (no discrimination ability).

Results

Of the total 175 processed referrals in 2015, 53 were diagnosed as having autism (30 per cent) which is consistent with diagnostic rates from the previous year.

Of the 86 who had an ADI-R as part of their assessment process, only 38 were diagnosed with autism (44.2 per cent) as shown in Table I. Out of the 80 without ADI-R, only 15 were diagnosed with autism (18.8 per cent).

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Table I: 2 x 2 Contingency table for the ADI-R

		Autism confirmed at LADS clinical decisions meeting		
		AUTISM DIAGNOSED	AUTISM NOT DIAGNOSED	
ADI-R Result	ADI suggests Autism	True positive TP 38	False positive FP 30	Positive Predictive value =55.9% TP/TP+FP
	ADI does not suggest autism	False Negative FN 0	True negative TN 18	Negative Predictive value =100% TN/FN+TN
		Sensitivity = 100% TP/TP+FN	Specificity = 37.5% TN/FP+TN	

Sensitivity, specificity and positive and negative predictive values of the ADI-R for males, females and people with and without intellectual ability were estimated. Table II illustrates high sensitivity amongst all groups, highest specificity for females and highest positive predictive value for those with ID.

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Table II Indices of diagnostic accuracy for the ADI-R

	Total Sample n=86	**Males n=61**	**Females n=25**	**With Intellectual disability n=35**	**Without Intellectual disability n=51**
Sensitivity	100	100	100	100	100
Specificity	37.5	32.35	50	33.33	39.39
Positive Predictive Value	55.88	54	61.11	62.96	51.22
Negative predictive Value	100	100	100	100	100

In logistic regression modelling, p-values help in deciding whether there is a relationship between two variables and the smaller the p-value, the greater the confidence that the relationship exists. A p-value of less than 0.05 indicates a good relationship.

In our study none of the two-way interactions were significant (all pW0.1), nor were there significance in the main effects of sex (p¼0.44) or ID (p¼0.68), i.e. there was no particular relationship between the individual domain scores of ADI-R and either the patients' gender or intellectual ability.

The final model, incorporating only the three domain scores (A, B, C), is presented in Table III.

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Table III Logistic regression model for the outcome of autism diagnosis, incorporating the three domain scores of the ADI-R

Variable	Coefficient	SE	z	P>|z|	95% CI
Reciprocal social interaction (A)	0.084	0.059	1.43	0.154	(-0.031, 0.199)
Communication(B)	0.179	0.105	1.71	0.088	(-0.027, 0.385)
Stereotyped Behaviours(C)	0.586	0.166	3.52	<0.001	(0.29, 0.912)
Constant	-4.488	0.922	-4.87	<0.001	(-6.295, -2.680)

To determine if any of the individual domains was strongly associated with the final autism diagnosis, the p-values of each of the domains must be considered. The smaller the p-value, the greater its association with autism diagnosis and hence the propensity to be a predictor for indicating either the presence or absence of autism.

The strongest predictor for the presence of autism was C (restricted, repetitive, stereotyped behaviours) with a very small p-value (p¼0.001), followed, respectively, by B (communication) with a p-value of 0.088 and A (reciprocal social interactions) with a p-value of 0.154.

When controlling for restricted, repetitive, stereotyped behaviour, neither of the other two domain scores were statistically significant; however, they were retained in the model because the compound cut-off used in clinical practice incorporates all three scores.

The ROC curve (Figure 3) produced an AUC of 0.908, indicating good discrimination of the ADI-R. However, considering the sensitivity and specificity for the whole sample, illustrated in Table I, the ROC curve suggests that a slightly more stringent threshold may improve the specificity without adversely affecting the sensitivity of the test.

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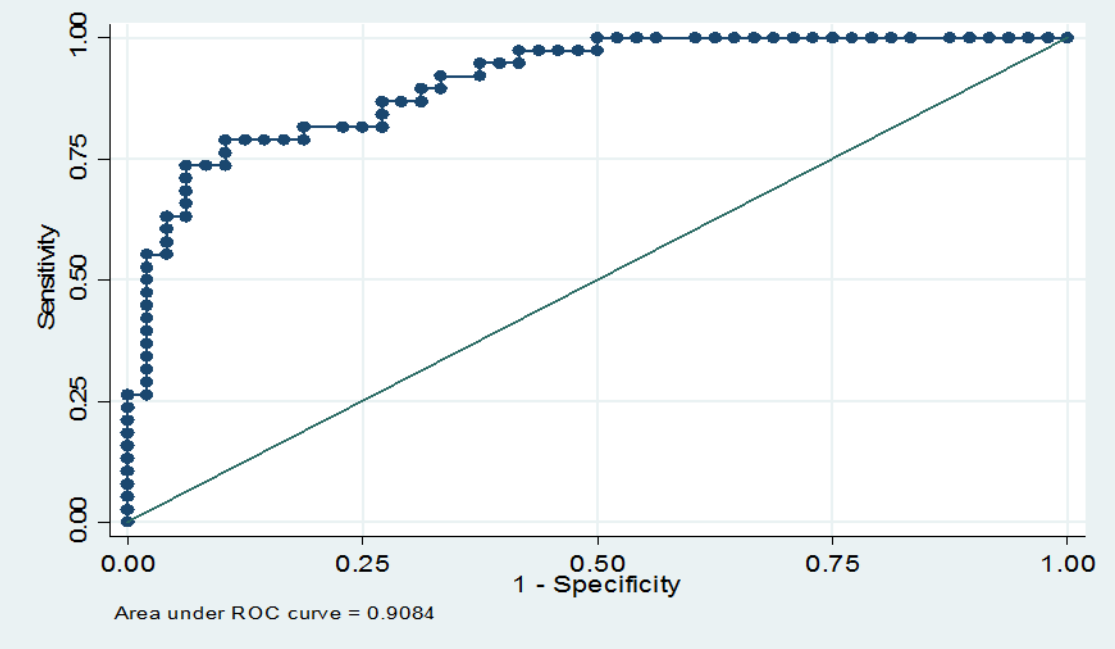
Figure 3 ROC curve for the ADI-R



Discussions

Sensitivity

A previous study by Sappok et al. (2013) concluded that the sensitivity of ADI-R is higher when compared to specificity. This is replicated in our study, although to a greater extent. The sensitivity of the ADI-R in this study was shown to be high (100 per cent) across all groups. The highest sensitivity obtained means that the ADI-R will recognise everyone with autism by giving a positive ADI-R score – but it will also suggest autism in people who do not meet the clinical criteria for autism. This is helpful both operationally and clinically as it supports the clinical pathway used by LADS which uses a developmental interview and a multidisciplinary clinical interview to make the final decision. The results clearly show that the ADI-R in this population, on its own, is not useful for identifying with certainty the presence of autism. The high sensitivity does not account for the high rate of false positives – i.e. it will suggest autism in people who are not autistic and could lead to over diagnosing.

These results suggest that providing diagnosis by using the ADI-R alone would not be appropriate. In addition, the result of this study supported the anecdotal impression of clinicians in the field that the ADI-R has a tendency to produce a high rate of false positives.

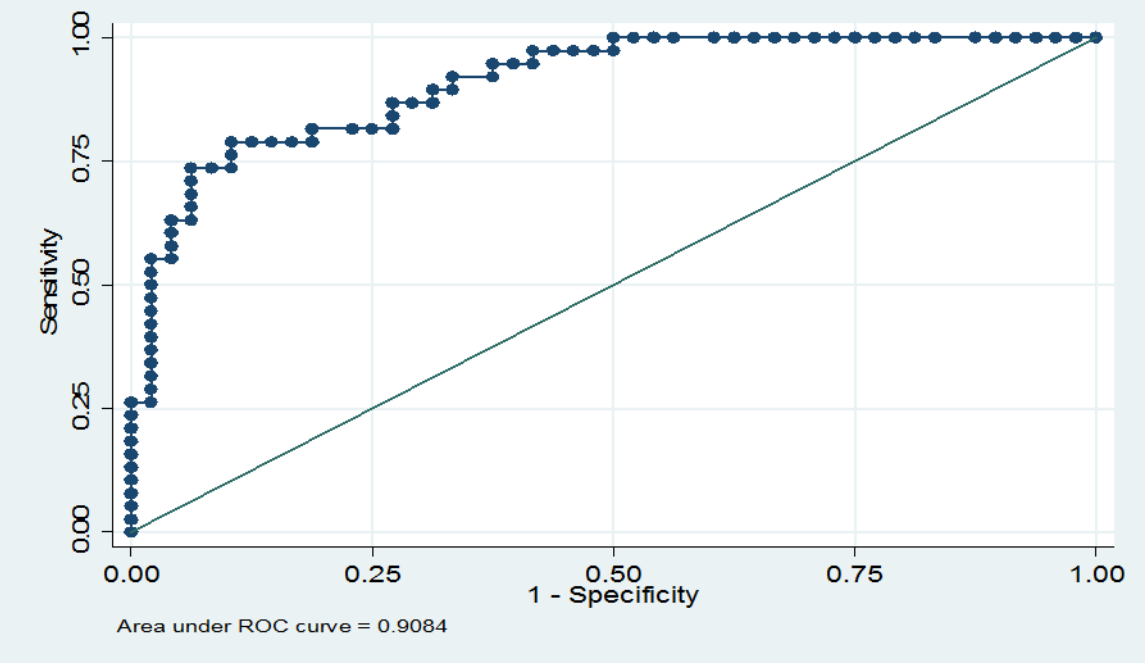
On the other hand, this high sensitivity will produce few false negatives which from a service perspective is extremely useful as it means that an ADI-R with scores failing to achieve thresholds in at least one of the domains is likely to be useful in ruling out autism. Obviously with the caveat that it is not negative due to lack of recall on the part of the interviewee.

Specificity

Previous studies suggest that the specificity of ADI-R is generally less than its sensitivity. The results of our study concur with this evidence, with the specificity ranging from 32.35 and 50 per cent. Clinically this finding is useful as the low specificity could be elevated slightly without compromising the integrity as such, of the diagnostic accuracy of the ADI-R.

From the ROC curve if the threshold is raised by 4 points the specificity could be increased up to 50 per cent, while keeping sensitivity to 100 per cent as shown in Figure 4. The blue arrow demonstrates the current specificity and the red arrow the potential increment in specificity by slightly increasing the thresholds. This could potentially help to avoid the false positive rates/over diagnosing people with autism based on the scores of ADI-R alone.

Figure 4 ROC curve for the ADI-R



Statistical analysis strongly points to the fact that while high sensitivity is obtained with the current ADI-R, increasing the stringency of the tool can positively increase the specificity without impairing the diagnostic accuracy of the test.

Gender

The differing presentations of males and females with autism causes some contention with regard to the current diagnostic approaches used to diagnose autism in females.

Although the number of females in our sample is less than males, nevertheless it is a significant number. It is therefore interesting to note that in our study, there is no difference in performance of the ADI-R in either males or females. It is uncertain if the presence of ID has any influence here because as compared to males the female sample had a higher proportion of people with ID. The ratio

of ID in males, diagnosed with autism via ADI-R, is 1:1.25, whereas it is 1:2.12 in females. However, there is a good association between ADI-R scores and the final outcome in both sexes.

While some have discussed gender disparity in autism and have suggested that females present differently and would need further diagnostic interventions (Fombonne et al., 2011; Baird et al., 2006; Goldman, 2013), our statistical analysis indicates that the ADI-R has the best combination of both sensitivity and specificity amongst females (sensitivity ¼100, specificity ¼50).

The results from this study show that the rate of diagnosis is similar in both males and females, although there are a higher number of male referrals by 2.4:1 as compared to females. The greater prevalence in males is in keeping with recent national figures in children (Taylor et al., 2013), although interestingly the adult rate in this population is lower than annual prevalence rates for each year as described by Taylor et al. (2013) which were steady at approximately 3.8/1,000 boys and 0.8/1,000 girls (4.75:1).

As diagnostic services develop nationally, although it is known that diagnostic pathways differ, it is important to conduct comparative studies in adults to further explore the gender differences in terms of referral rates and presentation differences. Mandy and Lai (2017) have captured the issue of this gender difference in a statistically eloquent and evidence rich article, calling for a sex and gender informed autism research.

Intellectual ability

Many adult autism diagnostic services only see people with intellectual ability within the mainstream range. LADS assesses people irrespective of intellectual ability so it was important to establish any differences when using the ADI-R within this population and this is depicted in Table IV.

Table IV Diagnosis by gender and intellectual ability

	Males	Females	With ID	Without ID
ADI-R	61	25	32	54
Diagnosed	27	11	17	21
Percentage diagnosed	44%	44%	53%	38%

During the period of study, there have been more referrals for people without ID; however, the rate of diagnosis is higher in people with ID. This is consistent with the pattern of referrals over recent years. Referrals of people with ID, often come from the community ID teams with good awareness and knowledge of autism. However, this higher rate of diagnosis is in keeping with the already well-recognised fact that the prevalence of autism is higher in people with ID (Brugha et al., 2012).

Improving diagnostic assessment

Diagnosing autism in adulthood remains (quite rightly) a clinical process using standardised instruments where practicable to aid this process. It is generally accepted that the combination of ADI-R and ADOS are the most helpful in yielding a clinical decision.

Logistic regression has enabled the evaluation of p-values for individual domains to indicate their relationship with the final diagnosis of autism. Statistically speaking, domain C (restricted, repetitive, stereotyped behaviours) showed the strongest predictability for the clinical diagnosis of autism with the smallest p-value (p¼0.001). This significant result is upheld by a growing body of evidence, both clinically (Halladay et al., 2017) and statistically (Duvekot et al., 2017) that scores on repetitive stereotyped behaviours domain of ADI-R are more indicative of autism, especially in males.

It is possible that the other domains of the ADI-R may also present in conditions such as attachment disorders, mental health problems or even ID, whereas restricted repertoires and circumscribed behaviours are either unique or more specific to autism. The team at LADS have made clinical observations prior to the study period that many people who appeared to meet the diagnostic threshold for autism had not scored on the restricted, repetitive and stereotyped behaviours on the ADI-R and in such cases the DISCO questions had to be used to ask more demanding questions pertaining to this domain. Taking both the statistical analysis and the clinical evidence into consideration, we suggest that the requirement for increasing the stringency of the tool could be performed by modifying this domain.

Wiggins and Robins (2008) studied the ADI-R behavioural domains in toddlers and suggested that stereotyped interests and behaviours are not as relevant to ADI-R as other criteria when evaluating for autism. However Le Couteur et al. (2007) studied the assessment tools in pre-schoolers (both ADI-R and ADOS) and found the presence of repetitive behaviours observed during ADOS to be of diagnostic significance.

A recent study by Larson et al. (2016) suggested that people with psychosis and autism have significantly fewer restricted, repetitive and stereotyped behaviours and interests than those with autism alone which may represent the genetic fractionation of the parts of the autism triad as proposed by Happé et al. (2008).

Further work needs to be done to analyse the diagnostic relevance of the different domains in the adult population and to find out if it is the same in both male and female genders and for people of all intellectual abilities.

Strengths and limitations

The study has been conducted in a naturalistic setting and is inclusive of the spectrum of intellectual ability. The sample reflects the general population and contains a good mix by gender and intellectual abilities. The study sample is robust in size for a single site study. However in general, the numbers are small and the test characteristics are dependent upon the sample size. In order to reduce the error a larger population is required and a multi-site study could potentially add more information.

This study is based solely on an adult population and for clinicians and researchers who use the ADI-R for confirmation of a diagnosis of autism this work is significant for understanding the specificity

and sensitivity of the ADI-R and the importance of not using it alone either in research studies or clinical services.

This study only looked at the referrals where a structured developmental history was possible. The reasons for not being able to undertake a developmental history are varied in an adult population – either parents/carers are no longer alive, live too far away or the person doesn't want their family involved for personal reasons. In this study about 49 per cent of the referrals did not have an ADI-R but the reasons for this were not examined. When such circumstances arise in daily clinical practice at LADS, we endeavour to find alternative sources of information to inform the final clinical decision. For example, getting collateral information from employers, siblings, family friends or reading child health/social care records. Sometimes we also arrange to meet the person for coffee in a less clinical setting so that we can more thoroughly assess current socio-communication difficulties and try to establish whether these difficulties are developmental by asking more questions of the person about their school experiences and childhood in general.

While the referral pool is from multiple sources, the study did not consider where the referrals came from, although a previous study in the same service (Davidson et al., 2015) showed that the majority of referrals were from primary care (40 per cent), followed by ID services (23.8 per cent).

The predictive values are functions not only of the test but also of the population in which it is used and is therefore prone to selection bias. This study was conducted in a specialist diagnostic service for autism and it is to be expected that the results may vary when compared to a different sample such as paediatrics. The presence of other confounding factors could be further explored.

It could be argued that the final outcome of the diagnostic pathway is in fact heavily influenced by the ADI result and these are not therefore independent variables, which is a potential source of bias. However, many factors are taken into account before making a final decision. The diagnostic conclusion is based on a series of assessments (by varying clinicians) including contemporaneous information like school reports, etc. and even employers viewpoints. This in combination with the ADI qualitative history, including the algorithm, is discussed prior to an ADOS assessment and all is finally contextualised within the remit of DSM-5.

The clinical team is of the viewpoint that the particularly robust diagnostic pathway is responsible for the service's relatively low conversion rate (the proportion of people who are assessed who receive a diagnosis of autism) (Davidson et al., 2015). This viewpoint has been upheld by this study which shows that the ADI-R has high sensitivity and low specificity. This means that if the ADI-R is used exclusively for diagnosis there will be a high number of false positives, i.e. people will be wrongly diagnosed with autism when they do not meet the clinical criteria for autism. This study therefore supports the continuance of the current pathway as it is a strong argument for the combined use of the ADOS and ADI-R as described by Risi et al. (2006).

Implications

The use of the ADI-R within the autism diagnostic service in Leeds can be continued with a better appreciation of its clinical value in day-to-day practice. Evidence supporting the anecdotal clinical view that it produces false positives can help alleviate anxiety when the developmental history is

clearly at odds with the clinical presentation. Furthermore, the knowledge that a negative ADI-R is likely to be an accurate predictor of the absence of autism will be helpful in those cases where clinical presentation is not clearly indicative of autism but there are several neurodevelopmental features that cause diagnostic confusion.

The team at LADS have previously tested clinical tools suggested by guidance to ensure their usefulness in the population catered for by the team. Kenny and Stansfield (2016) looked at the use of the AAA questionnaires in a similar population. (The AAA is part of the postal questionnaires sent prior to a screening assessment, consisting of Autism Quotient (AQ), Empathy Quotient (EQ), and Relatives Questionnaire (RQ). This study concluded that although AAA scores should not be used to influence the final outcomes the RQ (which is informant based) is a useful indicator of final outcome.

Most importantly our study reveals the significance of domain C – restricted and repetitive, stereotyped interests and behaviours (which again supports the clinical anecdotal observations, including the difficulties in ascertaining whether these features are present in adulthood). The service is currently producing brief educational films about autism using a patient expert. During one of these films, the expert describes the complex repetitive behaviours he uses to relax in the privacy of his own home – activities that many of his family and colleagues may be completely unaware of.

The clinical team will use the information obtained in the study and consider piloting raising of the threshold cut-off in this domain. In this way, we should be able to compare the consistency and concurrence of the ADI-R scores and clinical decisions diagnostic outcome. If this can be achieved, then the continued use of the time intensive ADI-R in conjunction with the present clinical pathway, can be justified in an increasingly constrained NHS (in terms of both time and finances), to provide the best possible clinical care and accurate autism diagnosis.

Contribution to authorship

AS conceptualised the research and took a substantial role in critically reviewing and revising the paper. ST and KB undertook the whole project and contributed significantly to the concept, drafting and revising the paper. All authors were involved in the approval of the final version.

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