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Review

Measurement properties of screening and diagnostic tools for autism spectrum adults of mean normal intelligence: A systematic review

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ABSTRACT

Background: The autism spectrum (AS) is a multifaceted neurodevelopmental variant associated with lifelong challenges. Despite the relevant importance of identifying AS in adults for epidemiological, public health, and quality of life issues, the measurement properties of the tools currently used to screen and diagnose adults without intellectual disabilities (ID) have not been assessed.
Objectives: This systematic review addresses the accuracy, reliability, and validity of the reported AS screening and diagnostic tools used in adults without ID.
Methods: Electronic databases and bibliographies were searched, and identified papers evaluated against inclusion criteria. The PRISMA statement was used for reporting the review. We evaluated the quality of the papers using the COSMIN Checklist for psychometric data, and QUADAS-2 for diagnostic data. For the COSMIN assessment, evidence was considered to be strong when several methodologically good articles, or one excellent article, reported consistent evidence for or against a measurement property. For the QUADAS ratings, evidence was considered to be "satisfactory" if at least one study was rated with a low risk of bias and low concern about applicability.
Results: We included 38 articles comprising 32 studies, five reviews, and one book chapter and assessed nine tools (three diagnostic and six screening, including eight of their short versions). Among screening tools, only AQ-50, AQ-S, and RAADS-R and RAADS-14 were found to provide satisfactory or intermediate values for their psychometric properties, supported by strong or moderate evidence. Nevertheless, risks of bias and concerns on the applicability of these tools limit the evidence on their diagnostic properties. We found that none of the gold standard diagnostic tools used for children had satisfactory measurement properties.
Conclusion: There is limited evidence for the measurement properties of the screening and diagnostic tools used for AS adults with a mean normal range of measured intelligence. This may lessen the validity of conclusions and public health decisions on an important fraction of the adult autistic population. This not only justifies further validation studies of screening and diagnostic tools for autistic adults, but also supports the parallel use of self-reported information and clinical expertise with these instruments during the diagnostic process.

1. Introduction

Autism Spectrum (AS) is a multifaceted neurodevelopmental condition for which the diagnosis is stable throughout its development [1]. Estimates of the prevalence of AS are regularly revised, following the recurrent updating of diagnostic guidelines. The prevalence of the autistic spectrum is estimated to be approximately 1%, with higher rates in men (1.8%) than in women (0.2%) [2,3]. AS is characterized in children, as well as in adults, by the coexistence of atypical communication and social interaction, with restricted and repetitive activities or behaviors (DSM-5) [4]. Current diagnosis criteria cover various symptom severity, language/speech, and intellectual levels. The reported proportion of autistic adults without intellectual disability (ID) is 50% [5], but this plausibly represents a conservative estimate, due to measurement issues [6], speech delay, or conversely, the absence of adaptive issues, limiting case ascertainment [7]. These individuals are frequently identified as "High Functioning", a potentially

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misleading label, confusing the level of adaptation with measured intelligence, and missing autistic people of normal intelligence with limited use of speech. Autistic adults of normal intelligence in these populations often manifest strong adaptive deficits that, in combination with poor societal adaptation, severely limit their socio-professional status [8,9]. They are often identified late, resulting in a 'lost generation' of adult autistic people [10]. Some may never even be diagnosed [2,11–13], with uncertain consequences for their well-being. Diagnostic challenges in adult autistics arise from a decreased magnitude of symptoms and atypicality with age and the presence of comorbid psychiatric conditions [14–17], and overlap between the signs of AS and those of other psychiatric or neurodevelopmental conditions [18]. One major obstacle to better identifying AS among adults is the lack of robust screening and diagnostic tools, as emphasized by most international guidelines [19–22].

In the absence of a biological gold standard method for the diagnosis of AS, its identification remains clinical, and requires multidisciplinary assessments from multiple sources [23,24]. The current state-of-the-art of adult AS diagnosis relies on self-report or informant questionnaires, observation guides, and clinical interviews. *Screening tools* are typically used to determine whether an individual is at risk for having AS and/or to justify a more formal assessment [25]. However, they are also used during the diagnostic process by primary care professionals or researchers with limited clinical expertise in the general or at-risk populations [20,26]. Screening tools designed for adults focus on the core symptoms of AS in different contexts, particularly among people referred for a medical diagnosis in clinical settings, notably in psychiatry units [20,27]. *Diagnostic tools* are used after a positive screening test to determine the presence or absence of AS when an individual displays signs of this condition. Trained professionals usually administer them during a multidisciplinary assessment [19,20,28]. They are more comprehensive, but also more time-consuming, and their use requires greater clinical expertise.

The choice of a diagnostic or screening tool depends, among several factors, on its measurement properties [29]: reliability, accuracy (or validity), sensitivity, specificity, and generalizability to the population for which they are intended to be used [20,30,31]. Systematic reviews of research evaluating tools devoted to the diagnosis of AS may guide clinicians and researchers in the selection of the best tools. The previous reviews of AS screening or diagnostic tools cover the entire range of age and/or IQ's [32–35]. Two reviews focus specifically on tools to assess AS adults without ID, but one is not systematic [36], and the other does not fully explore their measurement properties [37]. The aim of this study was to evaluate the measurement properties of the tools used for the screening and diagnosis of AS in adults without ID, focusing on their psychometric measurements and diagnostic accuracy.

2. Materials and methods

2.1. Search strategy

We performed a literature search for articles published in English or French in PsycInfo-Esbco (Psycinfo, Eric, PsycARTICLES, Psychology and Behavioral Sciences Collection), PubMed, Web of science (Web of Science™ Core Collection, KCI-Korean Journal Database, MEDLINE®, SciELO Citation Index), Cochrane Library, Science Direct, and Springer Linkin. The search was conducted in May 2016 and updated in September 2016, without limitation on the publication year. We followed the PRISMA standards, a 27-item checklist, and a four-phase process including identification, screening, eligibility, and inclusion of studies [38]. The keywords used were: "adult*", Diagnos*/Screen*, "Tool"/ "scale"/ "questionnaire", AS/autism*/asperger*. The algorithm used in each database was (diagnos* OR screen* OR assess*) AND (autism* OR AS OR Asperger*) AND adult* AND (tool* OR scale* OR questionnaire). We applied this algorithm to Abstracts for PsycInfo-Esbco; to all fields for PubMed, to Title, Abstract and Keywords for the Cochrane Library and Science Direct; to Topic for the Web of science; and to all the words for SpringerLink. We performed a complementary search using the reference lists of the studies selected for the review. Additionally, we searched the "grey literature" via Internet (Google and Google Scholar), according to the same keywords used in the database search. When a paper was not available, the authors were contacted via ResearchGate. The screening and selection processes are detailed in Fig. 1.

2.2. Inclusion criteria

Inclusion criteria for the selected papers were:

- documentation of AS screening and diagnostic tools focused primarily on AS core signs;
- reporting at least reliability, validity, or diagnostic accuracy of AS screening and diagnostic tools;
- a mean age of over 18 years at study entry, and a mean IQ over 70 for at least half of the participant sample. Papers in which the chronological age and intellectual level of their participants were not reported were excluded;
- having their participants defined through a "best estimate" diagnosis of autism, atypical autism, Asperger Syndrome, or PDD-NOS (Pervasive Developmental Disorder-Not Otherwise Specified), according to ICD-10 or DSM-IV criteria or to ASD DSM-5 criteria and a multidisciplinary assessment. The use of specific diagnostic tools, such as the Autism Diagnostic Observation Schedule-Generic (ADOS-G), or the Autism Diagnostic Interview-Revised (ADI-R), was not required. Autistic individuals with another physical or mental health condition were included;
- minimum sample size of 10 per group [20].

2.3. Data extraction

Two authors (AB &FR) read abstracts, and selected them if they were broadly consistent with the inclusion criteria. If consensus was not reached, the abstracts were set aside for further evaluation. Then, AB reviewed full-text articles of the selected abstracts against the inclusion criteria. Data were extracted from full-text articles (FR) and reviewed (AB), with regular verification and discussion to ensure consistency. Data extraction from full texts was organized into the following sections:

- tools (authors; type (e.g. questionnaire); targeted population; short description);
- information about each article (author(s); year; sample characteristics/sample size, age, cognitive level, gender, control groups);
- psychometric properties, including reliability (internal consistency, test-retest reliability, inter-rater reliability), construct validity (content validity, internal validity, criterion validity), and diagnostic validity (sensitivity, specificity, and accuracy measured by the Area Under the ROC Curve [AUC].

2.4. Data analysis

Measurement properties were independently assessed according to thresholds reported in the literature, such as satisfactory, intermediate, unsatisfactory, or no information available [25,27,29,39–49]. Sensitivity was considered to be satisfactory if the value