Systematic review of tools to measure outcomes for young children with autism spectrum disorder

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Scientific summary

Review of tools for autism spectrum disorder in children

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Scientific summary

Background

Autism spectrum disorders (ASDs) are neurodevelopmental, lifelong conditions that are diagnosed using a set of behavioural criteria. ASD is common, affecting at least 1% of the child and adult population. The ASD early intervention literature is largely focused on the promotion of social communication skills and management of coexisting behaviour problems. One difficulty for the interpretation of research findings is the multitude of different measurement tools that have been used in collecting evidence of progress and outcomes. The tools are of varying relevance and with limited evidence of their measurement properties when used with young children with ASD.

Review questions and objectives

The aims of the MeASURe (Measurement in Autism Spectrum disorder Under Review) review were to identify the validity of tools and outcome measures used in measuring and monitoring young children with ASD, and to consider how well these reflect and measure issues of importance for patients and carers. To achieve this, our objectives were to:

- identify the tools reported in literature on quantitative research involving children with ASD up to the age of 6 years
- conduct a detailed systematic review of the measurement properties of tools within the major domains of development and functioning
- synthesise evidence regarding the most robust and useful tools in these different domains
- identify gaps in measurement of outcomes and make research recommendations.

These steps were undertaken in the context of understanding what people with ASD, and parents, thought should be measured, and their perspectives about some of the better tools.

Methods

Framework for what outcomes to measure

To consider the outcomes of importance for parents and other key stakeholders, we consulted with people with ASD, parents and professionals. We were guided by the evidence-based procedures for developing a core outcome set outlined by the UK Medical Research Council-funded Core Outcome Measures in Effectiveness Trials initiative. As ASD is complex, and the review needed to take account of the developmental context of measuring outcomes up to the age of 6 years, we placed the findings of the consultation stages in a conceptual framework to guide the full review of tools for measurement. For the MeASURe conceptual framework, there were four primary domains, with subdomains in each of impairments, activity level indicators, participation and family measures.

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Understanding the views of people with autism spectrum disorder, parents and professionals around the measurement of outcomes that are of importance to them

We undertook the following steps:

- First, to identify the child- and/or family-specific outcomes that parents of children with ASD perceive as important, we undertook a scoping review of qualitative literature, using MEDLINE, the Cumulative Index to Nursing and Allied Health Literature (CINAHL) and PsycINFO (via Ovid).
- Second, we conducted a consultation – through groups and by e-mail – with young people on the autism spectrum regarding:
  - Outcomes – What do you think it is useful for health professionals and teachers to measure in young children (up to the age of 6 years) with autism?
  - Process – What is the best way of assessing these skills?
  - Where is the best place for observation to take place? What is it important for professionals to know about children with autism before they start to test them?
- Third, we undertook a survey through networks of health and education professionals to explore what constructs are most often measured by early years professionals in monitoring children's progress.
- Fourth, we consulted with parents at meetings (Exeter, London, Newcastle) on three occasions during the review process to establish which outcomes that parents consider to be most important.
- Finally, at the end of the review process, we held a discussion day with multiple stakeholders about the preliminary conclusions of the review, regarding what outcomes are important and how to assess them.

Systematic reviews

First systematic search

The *first systematic search* was undertaken to determine the range of tools used in observational and intervention evaluation studies in ASD, and relate these tools to the subdomains of the conceptual framework adopted for the MeASURe project.

Search strategy We included studies published from 1992 to coincide with the publication of the then-current international classifications, *International Classification of Diseases*, 10th Edition (ICD-10) and *Diagnostic and Statistical Manual of Mental Disorders*-Fourth Edition (DSM-IV).

Original searches were conducted in June and July 2012, and re-run in June and July 2013.

A total of 3059 papers were examined at full text and, from these, 255 papers were identified as appropriate for potential inclusion. There was a further stage of sifting of records found during the search of papers about measurement properties of tools, with searches completed by 9 September 2013. After exclusions, a total of 184 papers had information about tools extracted.

The following study types were included:

- all relevant randomised and quasi-randomised trials of early interventions
- cross-sectional and case–control studies of children
- descriptive cohort studies, including studies of baby siblings of children with autism, which provide information on tools to monitor developmental progress and follow early markers of ASD.

Child characteristics

We reviewed all studies in which at least 50% of children included had ASD operationalised as a 'best estimate' clinical diagnosis of ASD, including autism, ASD, atypical autism, Asperger syndrome and pervasive developmental disorder – not otherwise specified, according to either ICD-10 or DSM-IV criteria. All children were aged ≤ 6 years upon entering the study.

Types of measurement included

1. Direct assessment of child ASD symptoms by trained assessor.
2. Direct measurement of developmental skills, i.e. language, cognition, fine and gross motor skills, by trained assessor.
3. Observational measures of social interaction skills.
4. Interview or self-completed (parent, teacher or other professional) questionnaire report of child ASD symptoms.
5. Interview or self-completed questionnaire report of developmental skills – for example, language or adaptive skills – with/by parent, teacher or other professional.
6. Interview or self-completed (parent, teacher or other professional) questionnaire report of co-existing problems, including behaviour, aggression, sleeping, eating, toileting, anxiety, hyperactivity and others identified through parent consultation.
7. Idiographic measures focused on particular behaviours (e.g. goal attainment scaling, target behaviours).
8. Measures of impact on parent or family.

Types of measurement not included

- Economic impact on home and family.
- Experimental tasks and measures, for example barrier tasks, reaction time.
- Biophysical measures, medical investigations.
- Process measures.

Second systematic search

The *second systematic search* was undertaken to find papers that report the measurement properties of identified tools.

Not all tools identified for monitoring or outcome measurement could be searched for by name. First, a number of tools had been developed for a particular study (such as a coding system for parent–child interaction). Second, some tools were translations or adaptations of tools for use in another country, or had been used only up to 1994, and these were not pursued further for the purposes of this review. Original searches for papers describing measurement properties were conducted in March and April 2013, with follow-up searches completed in November 2013. The databases searched were Education Resources Information Center (ProQuest) – 1966 to present; MEDLINE (Ovid) – 1946 to present; EMBASE (Ovid) – 1988 to present; CINAHL (EBSCO*host*) – 1981 to present; and PsycINFO (Ovid) – 1987 to present.

In order to search for papers describing studies of measurement properties of tools, a specific search filter developed by the COSMIN (COnsensus-based Standards for the selection of health status Measurement INstruments) group was applied.

Each search consisted of four components: autism terms, age group terms, COSMIN filter and tool name. Searches were limited to English language only, and papers published from 1992 to present.

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Inclusion criteria

1. Tool identified in first search was the focus.
2. Tool (or subscales) measured a domain from the 'conceptual framework'.
3. Study published as 'full-text original article'.
4. The study sample overlapped with the age range of 0–6 years.
5. The study sample could be individuals who were being monitored for ASD symptoms even if they had another primary diagnosis (e.g. a paper monitoring ASD symptoms in a Fragile X population could be eligible if exploring measurement properties of a tool used as an outcome).
6. The aim of the study was the development of a measurement tool or the evaluation of one or more of its measurement properties.

Exclusion criteria

1. Papers in which the measurement tool was tested only for its properties in diagnostic assessment or screening.
2. A sample drawn from only the general population of children.
3. Sample size of < 20.
4. With regard to papers on translated tools, if the purpose was simply to validate the translated version then it was not eligible. If the purpose was to explore the tool's validity in a different culture/country, the focus was on the properties of the tool and the findings appeared relevant for use in UK then it was included.

Results

Understanding the views of people with autism spectrum disorder, parents and professionals around the measurement of outcomes of importance to them

We found a striking difference between the constructs rated important by parents, and the constructs most frequently measured by health and education professionals. We found that parents' experience with their children leads them to emphasise outcomes such as child emotional well-being as affecting the whole family. Professionals acknowledged that they measure what they have the tools for, and that their practice is influenced by an emphasis on the core impairments in autism and behaviour that challenge, rather than necessarily seeing the broader picture and measuring how the child is affected by their environment. Thus the consultation did not produce, at this stage, 'consensus' across stakeholder groups about what outcomes are most important to measure in young children with ASD.

Systematic reviews to determine tools in use, and their measurement properties

Of the 132 named tools that were identified as eligible for inclusion in searches about papers on their measurement properties, no papers meeting inclusion criteria were found for 75 tools and therefore their measurement properties in use with children with ASD could not be examined further. Fifty-seven tools (43%) remained, for which evidence on measurement properties was obtained.

The detailed data extraction using the COSMIN checklist provided some positive evidence with regard to at least one measurement property for 41 tools (seven with various versions/editions) identified as being used to measure outcome at stage 2 of the review. The tools are grouped by primary conceptual framework domain:

Autism symptom severity: Autism Behavior Checklist; Autism Diagnostic Interview-Revised (ADI-R); Autism Diagnostic Observation Schedule (ADOS, including Toddler Module and Calibrated Severity Score); Autism Observation Scale for Infants; The Baby and Infant Screen for Children with aUtIsm Traits-Part 1

(BISCUIT); Behavioral Summarized Evaluation (BSE-R; including Revised and Infant); Childhood Autism Rating Scale; Gilliam Autism Rating Scale (GARS and GARS-2); Modified Checklist for Autism in Toddlers; Parent Observation of Early Markers Scale; Pervasive Developmental Disorders Rating Scale; Social Communication Questionnaire; Social Responsiveness Scale (SRS).

Global measure of outcome Autism Treatment and Evaluation Checklist; Pervasive Developmental Disorders Behavior Inventory (PDDBI).

Social awareness Imitation Battery; Preschool Imitation and Praxis Scale (PIPS).

Restricted and repetitive behaviour and interests Repetitive Behavior Scale-Revised.

Sensory processing Sense and Self-Regulation Checklist; Sensory Profile including Short Sensory Profile.

Language MacArthur–Bates Communicative Development Inventories (MCDI); Preschool Language Scale-Fourth Edition.

Cognitive ability Leiter International Performance Scale-Revised; Mullen Scales of Early Learning; Stanford–Binet Intelligence Scales-Fifth Edition.

Emotional regulation Baby and Infant Screen for Children with aUtIsm Traits-Part 2 (BISCUIT-Part 2); Children's Global Assessment Scale; Infant–Toddler Social–Emotional Assessment (including Brief form).

Play Test of Pretend Play.

Behaviour Problems Child Behavior Checklist (CBCL 1.5–5 and CBCL 6–18); Aberrant Behavior Checklist; BISCUIT-Part 3; Home Situations Questionnaire-Pervasive Developmental Disorders (HSQ-PDD) version; Nisonger Child Behavior Rating Form.

Global measure of functioning Behavior Assessment System for Children-Second Edition; Psychoeducational Profile-Revised (and Third Edition); Scales of Independent Behavior-Revised; Vineland Adaptive Behavior Scales (VABS; including Classroom and Screener versions).

Parent stress Autism Parenting Stress Index; Parenting Stress Index-Short Form (PSI-SF); Questionnaire on Resources and Stress-Friedrich Short Form.

The most evidence was gathered for tools that were developed especially for use with children with ASD. Content validity of these tools was accepted for this review as strong. Unfortunately, given the focus of the review, there was minimal evidence about which tools have capacity to track children's progress over time or in response to an intervention. In the case of standardised assessments (e.g. of language, cognition and play) and many questionnaires (e.g. assessing behaviour, attention and emotional regulation) developed for the general population, there was limited evidence of their measurement properties when used with or about young children with ASD. We found no evidence concerning tools that can describe and measure some of the aspects of children's social participation and well-being (valued by parents as important). Also, we have no evidence about measures of family quality of life, although there is some evidence about measures of parent stress.

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Conclusions

The review has provided, for the first time, not only a list of tools used in measuring outcomes for children with ASD up to the age of 6 years, but also a systematic evaluation of their measurement properties and qualities. A tension between the diagnostic process in ASD, and the focus on parent and professional valued outcomes, was evident. The synthesis of evidence took into account the availability of tools, stakeholder views about the presentation of tools, the age range covered and the extent of the positive evidence about measurement properties in use with children with ASD. In summary, just 12 tools were considered the most valid overall; however, given their scope and limitations, these should not be considered a 'recommended battery'. These tools were ADOS; BSE-R; CARS; SRS; PDDBI; PIPS; MCDI; BISCUIT-Part 2 (co-occurring symptoms); CBCL; HSQ-PDD version; PEP; and the PSI-SF.

Research recommendations in order of priority

1. Development of a tool to measure child quality of life, with careful content validation for children with ASD.
2. Assessment of the measurement properties of a newly developed tool, the Brief Observation of Social Communication Change, by research group(s) in the UK, which has apparent promise as a primary outcome for early intervention trials focused on improving social communication in young children with ASD.
3. Further studies of the measurement properties of the VABS in young children with ASD in the UK.
4. Assessment of the measurement properties of the UK Early Years Foundation Stage Profile for use with young children with ASD.
5. Development of a questionnaire tool appropriate for young children with ASD to measure repetitive behaviour and circumscribed interests, which can be used across settings.
6. Assessment of the measurement properties of tools developed for young children with ASD which focus on problems such as anxiety and sleep.
7. Establishment of an agreed core set of outcomes to be measured in effectiveness trials of early intervention in ASD.

Study registration

This study is registered as PROSPERO CRD42012002223.

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