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Welcome to the second edition of 2018. This issue features papers from a range of stakeholder perspectives.

The first paper by Chester et al. is one of the first to examine the clinical challenges associated with a diagnosis of psychosis in individuals with autism spectrum disorder (ASD), and intellectual disability (ID) using a case series approach. It is generally accepted that early identification and treatment is essential to reduce impact on both present and future quality of life. This case series examines four patients with psychosis, ASD and ID, who have been in receipt of forensic mental health and ID services. In all cases, the use of antipsychotic medication was associated with an improvement in both psychotic symptoms and quality of life. ASD symptoms that were exacerbated as part of the psychosis also fell back to the levels they were pre psychosis.

The second paper from Adamou et al. examines gender differences in autism, which are still poorly understood. Our first paper based in an NHS specialist service for ASD. This study examines the assessment process retrospectively, specifically for significant gender differences. Using the ADOS scores of patients referred, the study results provide support that autism diagnostic tools have a gender bias towards a traditional male presentation of autism. With women females reportedly under diagnosis, this paper adds evidence to support the move for diagnostic tools to be developed to address this bias.

The third paper from Razaei et al. investigated the effects of combined risperidone (RIS) and pivotal response treatment (PRT) in children with ASD. Using the child communication checklist (CCC), two groups, namely, an RIS treatment group and an RIS plus PRT, were compared. In both groups, the total score of the CCC improved and the authors concluded that treatment with RIS combined with PRT may result in a better outcome in communication skill for children with autism than RIS training alone.

The fourth paper from Tolchard and Stuhlmiller describes the evaluation of health and behavioural lifestyle outcomes of people diagnosed with ASD in a student-led clinic in rural/regional Australia. Although it is generally accepted that people with ASD are at greater risk of developing chronic health through lifestyle choices and problems, there are few studies that examine people living in rural communities in cultural backgrounds traditionally underserved by healthcare services. This paper confirmed an increased risk for people with ASD developing chronic conditions when compared to the general population and dominant culture. Evidence included higher BMI and blood sugar levels and anxiety disorders. In terms of lifestyle, smoking was an issue for people with ASD but there was not an increased risk of alcohol use. The paper not only gives an insight into ASD in a rural community, but it also offers an insight and solutions into how services can respond and adapt or modify care for people with ASD to improve health outcomes.

The final paper from Kannis-Dymand et al. examines metacognitive beliefs and processes in children and associations with anxiety and depression. In the general population, metacognitive beliefs have been found to maintain perpetuate symptoms of anxiety and depression. However, the association of metacognitive beliefs in children with ASD is still unclear. This area has attracted limited research attention, and this study offers an insight into the role of metacognitive beliefs in high functioning children with ASD and comorbid anxiety or low mood and future targets for research.
We hope you enjoy this edition of the journal and wish to thank you for your continuing support of *Advances in Autism*. We invite contributions from our readers to the journal and welcome a variety of papers on areas including innovative and evidence-based practice, research, case studies, service and policy-related issues and literature reviews. We welcome submissions from the range of health and social care professionals, but additionally those who use services and people who care for them. If you would like to know more about how to submit your work for publication, please contact us at: chapline@lsbu.ac.uk
A phenomenological approach to diagnosing psychosis in autism spectrum disorder and intellectual disability: a case series

Rahul Rai, Samuel Tromans, Chaya Kapugama, Verity Chester, Ignatius Gunaratna, Peter Langdon and Regi T. Alexander

Abstract

Purpose – The diagnosis of psychosis in individuals with autism spectrum disorder (ASD) poses a unique clinical challenge. The presence of intellectual disability (ID) further complicates the diagnostic picture. Reliable and timely diagnosis of psychosis in such individuals minimises the duration of untreated psychotic symptoms and the subsequent impact on the quality of life of the patients concerned. The paper aims to discuss this issue.

Design/methodology/approach – The authors present four patients with psychosis, ASD and ID, who have received care within forensic mental health and ID settings. These examples demonstrate the interaction between these conditions, as well as issues pertaining to diagnosis and management.

Findings – In all four patients, sustained use of antipsychotic medication was objectively associated with an improvement in psychotic symptoms and quality of life. In instances where autistic phenomena were accentuated upon development of psychosis, such features returned to the baseline levels evident prior to the onset of psychosis.

Practical implications – The discussion and related case examples could improve the understanding of the possibility of psychosis in individuals with ASD and ID, and increase awareness of this diagnostic possibility among healthcare professionals.

Originality/value – This is the first published case series illustrating the challenges of diagnosing psychosis in individuals with ASD and ID.

Keywords Mental health, Learning disability, Mental disorder, Schizophrenia, Autism spectrum condition, Comorbidity

Paper type Case study

Introduction

Autism spectrum disorders (ASD) and psychotic disorders have historically been considered as related diagnostic entities (Sugranyes et al., 2011). However, the nature of this relationship has been the subject of extensive debate (Padgett et al., 2010), alternating between the view that ASD is an early manifestation of childhood schizophrenia on the one hand (Kanner, 1949), and that people with ASD could not be diagnosed with schizophrenia on the other. It is now widely believed that ASD and psychosis are two distinct clinical entities. Kolvin et al. (1971) highlighted that children whose abnormal behaviours were apparent before the age of three years fitted with Kanner’s description of “early infantile autism” (Kanner, 1968). In contrast, those children whose development was essentially normal until school years, but then later developed hallucinations, delusions or other behavioural abnormalities, were felt to be more in keeping with a diagnosis of schizophrenia.
A number of studies have examined whether schizophrenia is significantly more prevalent amongst people with ASD, in comparison to the general population (Billstedt et al., 2005; Ghaziuddin et al., 1998; Volkmar and Cohen, 1991). A recent systematic review examined the rates of psychosis in individuals with ASD, reporting a prevalence which ranged from 0 to 53 per cent (Padgett et al., 2010). However, the authors noted marked differences and heterogeneity in terms of the methodological approaches of the included studies, which precluded a meaningful pooling of the findings. For example, the prevalence of psychosis in adults with ASD differed greatly between studies. Joshi et al. (2013) found a lifetime prevalence of 13 per cent and point prevalence of 8 per cent of psychosis in adults with ASD, whereas a study by Volkmar and Cohen (1991) found only 0.6 per cent from a sample of 163 patients with ASD. Stahlberg et al. (2004) reported that 7.8 per cent of participants with diagnosed ASD had comorbid schizophrenia, which is higher than the 1 per cent prevalence of schizophrenia in the general population (McGrath et al., 2004). A preliminary study reported a prevalence of 2.4-5.3 per cent across three secure hospitals, depending on whether equivocal cases were considered within the estimate (Hare et al., 1999).

A meta-analysis highlighted that childhood-onset schizophrenia is preceded by and comorbid with ASD in 30-50 per cent of cases (Rapoport et al., 2009). Likewise, it has been noted that in a population of people with childhood-onset schizophrenia, 25 per cent met the criteria for childhood ASD (Sporn et al., 2004).

Furthermore, Larson et al. (2017) conducted a study whereby 116 individuals with ASD and psychosis were compared with a group with psychosis only. They found that a diagnosis of atypical psychosis was more likely in individuals with ASD relative to those with psychosis only, who were more likely to receive a diagnosis of schizophrenia. This suggests that there may be a specific manifestation of ASD linked to comorbid psychosis, yielding an atypical clinical picture, especially with regard to affective disturbance.

Phenomenological overlap is a likely contributory factor to the wide variability in the reported rates of schizophrenia in people with ASD (Skokauskas and Gallagher, 2010). The neurodevelopmental hypothesis of schizophrenia suggests that its origins in adolescence are partially explained by the consequences of events in early development (Owen et al., 2011). Evidence suggesting a genetic overlap between neurodevelopmental disorders such as ASD, attention deficit-hyperactivity disorder (ADHD), bipolar affective disorder and major depressive disorder are emerging (Cross-Disorder Group of the Psychiatric Genomics, 2013). Such findings suggest viewing these functional psychoses as a group of related and overlapping syndromes, with origins partially based in the early developmental period.

**Diagnostic issues**

The diagnosis of a psychotic disorder in an individual with ASD poses a clinical challenge. In fact, numerous cases of misdiagnosis of psychosis and schizophrenia have been reported, especially in instances where the ASD was not previously diagnosed (Tantam, 2003). Through their ASD presentation alone, some people with ASD may meet the criteria for schizophrenia on clinical interview schedules, such as the Structured Clinical Interview for the Diagnostic and Statistical Manual of Mental Disorders (Konstantareas and Hewitt, 2001). More specifically, the negative symptoms of schizophrenia have an overlap with the symptoms of ASD (Frith and Frith, 1991). Carpenter (2007) reported that it is relatively common for more able individuals with ASD to be erroneously diagnosed with schizophrenia, and cited examples of how some common features of ASD may be misinterpreted, as summarised in Table I.

Conversely, there is also a risk that the diagnosis of schizophrenia can be missed among those diagnosed with ASD. It is therefore essential to carefully elicit the content and form of the person’s thoughts, which will help identify a change from the usual preoccupations/meanings attached to these thoughts. A thorough exploration of phenomenology can minimise the risk of psychosis being missed in people with ASD.

The presence of intellectual disability (ID) adds a further layer of complexity to the aforementioned difficulties of misdiagnosing ASD as schizophrenia, or vice versa, as well as establishing cases where there is a dual diagnosis of ASD and schizophrenia. This is further discussed in Box 1 (Fletcher et al., 2007). Such cases are rarely described within the literature, and thus there is a limited understanding
of the prevalence of coexisting ASD and schizophrenia within the ID population. Esan et al. (2015) assessed a sample of 138 patients treated over 6 years within an inpatient forensic ID service, finding that 6 of these patients had both ASD and schizophrenia. However, it is difficult to make assumptions of the representativeness of this single study (Esan et al., 2015).

It has been suggested that the reliability of diagnosing psychosis in people with ASD who also have ID is poorer than in those in the general population (Dossetor, 2007). This is unsurprising given that there is no consensus on the best way to assess psychopathology in adults with ASD and ID (Underwood et al., 2010). There is no guidance for the clinician on the diagnosis of psychosis in ASD in the 10th revision of the International Statistical Classification of Diseases and Related Health Problems (ICD-10) (World Health Organisation, 1992). The 5th edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-V) specifies that in individuals with a history of ASD, the diagnosis of schizophrenia can only be made if there is a presence of hallucinations and delusions for at least one-month duration (American Psychiatric Association, 2013). However, this seeming lack of guidance and literature on psychosis and ASD should not prevent the consideration of the diagnosis in patients where such clinical suspicion exists. The failure to correctly identify psychosis in individuals with ASD would mean a lack of provision of antipsychotic treatments, prolonging psychotic experiences and further compromising quality of life. Conversely, the failure to identify ASD in individuals with comorbid psychosis would lead to individuals receiving inadequate treatment for their actual clinical needs (Larson et al., 2017).

In this case series, we present four patients with mild ID who illustrate the clinical challenges of differentiating between diagnoses of ASD and psychosis, or may indeed have both conditions, with the overall aim of improving recognition, understanding of the interface between these conditions, as well as issues pertaining to diagnosis and management.

<table>
<thead>
<tr>
<th>Table I</th>
<th>Common reasons for the misdiagnosis of schizophrenia in ASD</th>
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<tbody>
<tr>
<td><strong>ASD feature</strong></td>
<td><strong>Reason for potential confusion with schizophrenia</strong></td>
</tr>
<tr>
<td>Imaginary friends</td>
<td>Can be interpreted as thought broadcasting and insertion</td>
</tr>
<tr>
<td>Preoccupation with control</td>
<td>Can lead to a person being said to have delusions of control or influence</td>
</tr>
<tr>
<td>Obsessional thoughts</td>
<td>Can also have a quasi-hallucinatory, inserted quality</td>
</tr>
<tr>
<td>Internal dialogue</td>
<td>Can be interpreted as hallucinatory voices giving a running commentary or discussing amongst themselves</td>
</tr>
<tr>
<td>Bizarre ideas</td>
<td>Can be held in such a fixed manner, that it has the quality of a delusion</td>
</tr>
<tr>
<td>Fleeting visual hallucinations</td>
<td>If such hallucinations are rare and fleeting, they do not merit a diagnosis of schizophrenia</td>
</tr>
<tr>
<td>Oddities of speech</td>
<td>The way that people with ASD may speak – with tangential comments and abbreviated explanations can mimic thought disorder</td>
</tr>
<tr>
<td>Stereotypes and catatonic symptoms</td>
<td>Can also lead to an erroneous diagnosis of schizophrenia</td>
</tr>
<tr>
<td>Social isolation</td>
<td>When undergoing time of change, social isolation can become more evident</td>
</tr>
<tr>
<td>Proneness to psychotic reactions under stress/anxiety</td>
<td>When stressed, internal experiences can become “out of control” and appear to merit the diagnosis of psychosis</td>
</tr>
<tr>
<td>Concrete thinking</td>
<td>May be present in both ASD and schizophrenia</td>
</tr>
</tbody>
</table>

Source: Carpenter (2007)
Method

Design

In this case series, four patients with psychosis, ASD and ID, who have received care within forensic mental health and ID settings, are presented.

Procedure

Clinicians working within two forensic ID services (one national health service and one independent sector) in the East of England were invited to submit cases to the project, and provided with the procedure to adhere to. The clinicians were asked to provide key information which included the apparent age of onset of psychosis, relevant family history, clinical diagnoses according to diagnostic manuals, any diagnostic tools to assist in their decision making, evidence of impairment due to symptoms and the patient’s response to described treatment. All participants had diagnoses of ID and ASD according to established diagnostic criteria, made by consultant psychiatrists during their care.

Ethical considerations

Case studies are presented according to principles established by the British Medical Journal Ethics Committee (British Medical Journal, 2018). These include obtaining informed consent and removal of patient identifying details. Informed consent was obtained from all patients included as case studies within the paper.

Case Series

Four case studies are presented (cases A-D). Cases A and B are described in Tables II and III, and cases C and D are described in Tables IV and V.

<table>
<thead>
<tr>
<th>Table II</th>
<th>Case study A</th>
</tr>
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<tbody>
<tr>
<td>Age of onset of psychosis</td>
<td>20 years</td>
</tr>
</tbody>
</table>

Family history
A has a family history of schizophrenia in his brother, anxiety and depression in his mother, and harmful use of alcohol in his father.

Diagnoses
- Mild intellectual disability (F70.1)
- Autism spectrum disorder (F84.0)
- Schizoaffective disorder (F25.1)
- Harmful use of alcohol, cannabis and solvents (F10.1, 12.1 and 18.1) though currently abstinent in a forensic setting

Diagnostic tools used
- WAIS-III (Wechsler, 1997): full scale intelligence quotient = 61
- ICD-10 (consensus of diagnosis established between two psychiatrists) (World Health Organization, 1992)

Brief description
A is a 33-year-old male with mild ID. He experienced sexual abuse during his childhood. Though he attended mainstream school, he struggled with lessons and left school aged 14 years, with no formal qualifications. He was described as a loner, having only one close friend. After leaving school, he began work at an animal shelter. He struggled to understand instructions, often interpreting them in a concrete manner.

In his teens, A felt that he was homosexual, and also developed a sexual interest in animals. He fantasised about the latter, and occasionally acted on such fantasies, though after such encounters, he would ruminate and become distressed. At around 20 years of age, A started hearing voices commanding him to carry out bizarre sexual acts with animals. He firmly believed that if he did not comply with these commands, either he or his mother would be killed by the voices. A also experienced unpleasant bodily sensations, and was convinced that the individuals responsible for the voices were imposing these experiences on his body. Along with these symptoms, he had episodes of mood disturbances alternating between low mood and elation/irritability, lasting for few weeks at a time.

Evidence of impairment due to symptoms
- Subjective distress
- Risks of self-harm (alcohol abuse)
- Risks of sexually inappropriate behaviours (sexual acts against animals)
- Vulnerability (deterioration in level of functioning related to onset of symptoms)

Response to treatment
While on antipsychotic medication, the intensity of A’s hallucinatory experiences reduced considerably, alongside a subsequent reduction in distress, and sexually inappropriate behaviour towards animals.
Discussion

This paper has described the phenomenological approach to diagnosing psychosis in individuals with both ID and ASD. The paper has a number of drawbacks, including all of the associated difficulties with case studies, such as limited generalisability. Furthermore, all of the relevant cases identified during this study were male, and as such, future research on women with this combination of diagnostic comorbidity is required. Nevertheless, the paper has a number of implications. While there is a growing clinical ability in professionals to recognise and diagnose ASD, and an increasing recognition that mental disorder is more prevalent in people with ID, the differentiation between, and diagnosis of psychosis in ASD remains a clinical conundrum, with further complexity if ID is also present.

Clinical training should place appropriate emphasis on the ability to understand, describe and document the interface between ASD and psychosis. More research, including qualitative research, is needed to support the development and evaluation of reliable diagnostic criteria, and subsequent tools to support clinical practice. There are a number of issues currently affecting practice in this area. There appears to be a reluctance to diagnose psychosis (when clinically appropriate) which poses difficulty in early recognition and treatment (Larson et al., 2017). A delayed diagnosis, or a diagnosis not being made, is likely to result in a poor treatment response, as well as a need for higher doses of antipsychotics. Symptoms not appropriately treated may result in risky behaviour, subjective distress, mistrust in professionals and a poorer quality of life. In some cases, delayed diagnosis may increase the likelihood of being admitted to inpatient or secure placements.

There is currently a lack of guidance from diagnostic criteria, and, subsequently, tools to assist with the diagnosis of ASD and psychosis. As such, the clinical approach must be based on a phenomenological assessment, including differentiating objective reality, the “normal alternate reality” of autism and the “loss of reality contact” observed in psychosis, from one another.
The value of establishing a good picture of premorbid and baseline level of functioning, obtaining information from multiple reliable and consistent sources, conducting detailed observations in various settings and contexts, and seeking another opinion where possible to enhance reliability cannot be overemphasised. Until better tools and criteria are developed, a judicious clinical approach may separate reliable early diagnosis and treatment with therapeutic benefit, from progressive deterioration and therapeutic nihilism.

All the cases described in our series had a mild level of ID, and were capable of adequate communication and engaged well, allowing independent assessment of their mental states by at least two psychiatrists. The qualitative change in behaviour, new-onset symptoms including hallucinations and delusions, as well as the alteration or modulation of pre-existing autistic phenomena coupled with a decline in general functioning all supported the diagnosis of a psychotic illness. While a treatment response cannot be considered a diagnostic test, the treatment with antipsychotic medication alleviated the new-onset symptoms with minimal effects on pre-existing autistic symptoms.

The patient described in the first case had a pre-existing sexual interest in animals. With the onset of psychosis, the rationale behind committing sexual acts to animals was to prevent harm to himself and his mother by a persecutor. The accompanying distress was reduced after commencement of antipsychotic medications. In the second case, the onset of psychosis correlated with a clear accentuation of autistic phenomena, as well as the development of clear-cut delusions and affective symptoms. With treatment, the autistic symptoms returned back to baseline levels.
Similarly, in the third case, there was a clear onset of new symptoms during episodes of psychosis. Circumstances leading to the committing of a criminal offence may have been attributable to a combination of autistic and psychotic phenomenology. The patient appeared to have developed persecutory ideations towards his mother in the context of being destabilised by changing placement. In the last case of the series, the onset of psychosis was mistakenly attributed to ASD for a period of time. The clear delusional content and perceptual disturbances were eventually identified whilst the patient was in hospital following detailed psychiatric assessments and longitudinal observations.

In all of these cases, the diagnostic difficulty lay in determining if the patients’ preoccupations, thoughts and behaviours were qualitatively different from the features of ASD. For example, while the “content” of the psychopathology was similar to that of ASD, the “form” was different. Features suggestive of a comorbid psychosis include the degree of distress, an increase in the intensity and frequency of preoccupations, the appearance of morbid themes, new behaviours and a change in functioning. A careful exploration of phenomenology to differentiate between the content and form of thoughts is required.

Regarding management, it is important for clinicians to develop a clinically sensible, sensitive and balanced approach to ensure people with ASD with possible development of a comorbid psychotic disorder access the principles of early recognition, intervention and treatment. Psychosis diagnosed in a person with ASD should be treated in the same way as psychosis affecting any other individual. This includes prescribing adequate doses of antipsychotic medication, after ensuring clarity around the target symptoms that need to be addressed.

### Table V  Case study D

<table>
<thead>
<tr>
<th>Age of onset of psychosis</th>
<th>15 years</th>
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<tbody>
<tr>
<td><strong>Family history</strong></td>
<td>D had no known family history of mental illness or intellectual disability</td>
</tr>
</tbody>
</table>
| **Diagnoses**             | Mild intellectual disability (F70.1)  
|                           | Autism Spectrum Disorder (F84.0)  
|                           | Hyperkinetic Conduct Disorder (F90.1)  
|                           | Paranoid schizophrenia (F20.0)  |
| **Diagnostic tools used** | Schedules for Clinical Assessment in Neuropsychiatry (SCAN) (World Health Organization, 1994) |
| **Brief description**     | D is a 32-year-old male with mild ID, ASD and paranoid schizophrenia. His family reported that his early developmental milestones were delayed, particularly his speech. He attended special educational needs schooling and came to the attention of mental health services at around the age of 5 years. He was diagnosed with ADHD and conduct disorder during his childhood. Later on, he was diagnosed with paranoid schizophrenia during his adolescent years, at a time when he had been experiencing difficulties in affect regulation, an intense preoccupation in death, poor relationships with his peers and a history of fire setting. D was first admitted to hospital at the age of 15 years and had multiple subsequent readmissions. Most readmissions were due to relapses resulting from non-compliance with medication, as well as alcohol and illicit substance abuse.  
|                           | When initially admitted to hospital, D would typically report hearing the tweets of birds, with the conviction that they were directed specifically to him. He believed that such experiences represented warnings that people intended to harm him. D had previously worn a stab proof vest and carried a knife in order to protect himself from such perceived harm. D also complained that he was able to see small lights on walls and lights coming through windows, which he interpreted as an indication that there were men outside using torches, awaiting an opportunity to murder him. He additionally reported that women were communicating to him through the television to warn him of forthcoming danger. D specifically mentioned a well-known British television presenter doing this repeatedly. D also reported other firmly held beliefs consistent with a clinical picture of Capgras syndrome, stating that his family, and the nursing staff, had been taken and replaced by clones, who were intent on murdering him. He believed that they were talking exclusively about him into a recording device. He believed staff were “wearing wires” and spying on him and planned to murder him. D also disclosed that he thought these individuals were part of the mafia. |
| **Evidence of impairment due to symptoms** | Subjective distress  
|                           | Risks of harm to others (related to hallucinations/delusions)  
|                           | Risks of property damage/ fire setting (related to hallucinations/delusions)  
|                           | Risks of substance misuse (as a means of managing his distressing experiences)  
|                           | Vulnerability (deterioration in level of functioning related to onset of symptoms)  |
| **Response to treatment** | D initially responded well to Olanzapine but would frequently stop taking oral medication. His periods of non-compliance correlated with increases in illicit drug and alcohol abuse. Commencement of Olanzapine in depot form led to sustained symptomatic improvement  |
Non-pharmacological treatments, such as interventions to address maladaptive assumptions, the use of distraction or masking techniques to treat auditory hallucinations (Nelson et al., 1991) and interventions to reduce expressed emotions (Anderson and Adams, 1996) may also be used as adjunctive measures.

It should also be recognised that the combination of ASD and psychosis may have an impact on the length of stay in hospitals. Hare et al. (1999) reported that people with ASD stayed in hospital settings for an average of 8.5 years, which is 2-3 years longer than other patients. This may be because people with ASD pose unique challenges in terms of management, treatment and eventual placement (Alexander et al., 2016). When a comorbid psychotic disorder is present, this may potentially increase the length of stay even further. However, it is likely that improving the recognition of the needs of patients with comorbid ASD and psychosis will begin to improve the effectiveness of treatment in this area.

A key area of relevance is risk management of people with ASD and comorbid psychosis. Some of the features of ASD, such as a lack of understanding of social norms, concrete interpretation of rules, misinterpretations of others’ intentions, difficulties in expressing emotions and pursuit of special interests with morbid/ unusual qualities, may pose additional risks on patients when they are diagnosed with psychosis, potentially making them even more prone to offending. Future research should seek to clarify these issues, in order to more effectively assess and treat patients with complex presentations.

References


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Autism Diagnostic Observation Schedule (ADOS) scores in males and females diagnosed with autism: a naturalistic study

Marios Adamou, Maria Johnson and Bronwen Alty

Abstract

Purpose – Many tools are available for assessing autism in an adult population; however, few have been studied for the effects of gender on diagnostic scores. The purpose of this paper is to evaluate the Autism Diagnostic Observation Schedule (ADOS) assessment for gender bias in a clinical population, specifically whether the ADOS favours a “male-type” of autism.

Design/methodology/approach – The ADOS scores of patients referred to an NHS specialist autism assessment service were retrospectively examined for significant gender differences. The combined ADOS scores and diagnostic outcome were grouped by gender for each participant. The data were analysed in SPSS using independent t-tests to look for significant gender differences between combined ADOS scores and diagnostic outcomes.

Findings – A significant difference was observed in the mean combined ADOS scores for those participants who later received an autism diagnosis (male = 10, female = 6, t (13) = 3.34, p = 0.005). However, no significant difference was observed between mean scores of those who did not receive an autism diagnosis (t (26) = 1.21, p = 0.237).

Originality/value – The ADOS is a popular assessment used for autism diagnosis. These results provide support for a male gender bias. This could have clinical implications for autism assessment services, whereby lower diagnostic thresholds could be considered for female patients. This could allow more females with autism to receive a diagnosis, and access support services.

Keywords Assessment, Diagnosis, Autism spectrum disorder, Adults, Autism spectrum condition

Paper type Research paper

Background

Autism spectrum disorder (ASD) is a neurodevelopmental condition characterised and diagnosed by impairments in social communication and social interaction in the presence of restricted, repetitive behaviours, or interests (American Psychiatric Association, 2013). Current prevalence is estimated to be at least 1.5 per cent in developed countries, with recent increases primarily among those without comorbid intellectual disability (Baxter et al., 2015; Brugha et al., 2016). The onset of ASD symptoms typically occurs by age 3, although symptoms may not fully manifest until school age or later, and some research suggests symptoms can emerge between 6 and 18 months of age (Szatmari et al., 2016).

Approximately, four males are affected with ASD for every female (Szatmari et al., 2016; Lord et al., 1994; Volkmar et al., 1993; McLennan et al., 1993; Brugha et al., 2011; Loomes et al., 2017) though the sex ratio appears to decrease with increasing severity of the condition (Werling and Geschwind, 2013). The reasons behind this difference to the gender ratio are still unclear with suggestions made that it has a genetic origin (Skuse, 2000; Jacquemont et al., 2014; Iourov et al., 2008) based on the assumption that psychosocial environmental factors only rarely appear to be influential in bringing about phenotypic expression (Bailey et al., 1996).

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The data were initially gathered by Bronwen Alty as part of her work towards her MSc degree dissertation, with Leeds Beckett University. The authors would like to thank the team at the Adult ADHD and Autism Service for their help and support with this paper.
There is, however, a possibility that there are other causes that could explain this difference. It maybe that the diagnostic criteria which are gender agnostic do not capture accurately the qualitative differences between males and females with autism (Hiller et al., 2014; Gould and Ashton-Smith, 2011), the quantitative differences in the normative distribution of autistic traits between males and females (Constantino and Charman, 2012; Lai et al., 2013) and the developmental differences between males and females with (Lai et al., 2011; Kreiser and White, 2014) suggesting that males and females may require partly different criteria in defining “having” autism which is not currently the case.

For example, since females have more developed social skills than males with ASD (Wang et al., 2017; Head et al., 2014) or appear as better adapted (Dworzynski et al., 2012) may not present as impaired during a diagnostic assessment or even seek one in the first place (Dworzynski et al., 2012) leading to a lower diagnostic rate. To the contrary, because males show more restricted, repetitive, and stereotyped behaviour than females (Hartley and Sikora, 2009; Van Wijngaarden-Cremers et al., 2014) which are easier to identify, may be more likely to receive a diagnosis. This outcome is not desirable as potential under recognition of ASD in females may lead to them receiving inadequate care, masking of coexisting mental and physical health problems and lead to social and economic exclusion (National Institute for Health and Clinical Excellence, 2012).

Different rates between males and females have been reported in other mental health disorders such as major depressive disorder (Abate, 2013), bipolar affective disorder (Douglas and Scott, 2014) and schizophrenia (Ochoa et al., 2012) with similar reports of gender bias as in ASD with diagnosis (Hambrecht et al., 1993; Brommelhoff et al., 2004). As with psychosis (Thomas, 2001), the evolution of the understanding of ASD has changed (Robel, 2010) with early descriptions suggesting it was the result of childhood psychoses, or psychodynamic disturbances of parent-child relationships (Mintz, 2017), or caused by women (Kanner, 1949), pointing towards a gender bias to the development of the ASD concept itself.

Reports of gender bias in one of the screening tools for ASD (Murray et al., 2017) made us want to investigate if the same would apply to diagnostic tools. In our study, we examined if one of the commonly used diagnostic tools the Autism Diagnostic Observation Schedule (ADOS) (Lord et al., 1989) had the potential to introduce gender bias to the diagnosis of ASD.

ADOS is a standardised protocol for observation of social and communicative behaviour associated with ASD and consists of a series of structured and semi-structured processes for interaction accompanied by coding of specific target behaviours associated with particular tasks and by general ratings of the quality of behaviours. It is recommended by the National Institute for Health and Care Excellence (2012) and it is an important tool as it has been found to be objective in naturalistic settings (Zander et al., 2016) and demonstrated strong predictive validity against best estimate diagnoses (Gotham et al., 2007).

**Method**

The convenience sample was of 43 service users who underwent an assessment for ASD at the Service for Adults with Neurodevelopmental Disorders at an NHS Foundation Trust in Yorkshire between May 2015 and June 2016. As part of the normal service pathway, the ADOS-2 (Lord and Rutter, 2012) Module 4 is administered to all patients. The pathway includes obtaining a psychiatric history and multidisciplinary team discussion of every case which leads to a diagnostic classification according to DSM-5 criteria. No patients with an intellectual disability are accepted and whenever there is a suspicion, a formal assessment is made to exclude it using the Wechsler Adult Intelligence Scale (Skuse, 2000). The team includes medical doctors, clinical psychologists, social workers, occupational therapists, nurses and speech and language therapist. All clinicians who administered the ADOS were trained by a certified provider and underwent interrater peer validation. The ADOS-2 scores for the period of May 2015 and June 2016 were collected as part of a service evaluation project in March 2017, after the clinical diagnosis was made and were entered anonymised in an SPSS Version 24 database.

The ADOS-2 is a semi-structured, standardised assessment designed for use with individuals referred for possible ASD. Five ADOS modules accommodate various developmental
and language levels. In each, a protocol of activities or social processes is administered in approximately 45 minutes, and then items are scored on a four-point scale, with zero indicating "no abnormality of type specified" and three indicating "moderate to severe abnormality". To receive an ADOS classification of autism or ASD, an individual's scores on the original diagnostic algorithms must meet separate cut offs in the communication and social domains, and a summation of the two. If any or all of these thresholds are not met, then a nonspectrum classification is assigned. Item scores of 2 and 3 are collapsed in the algorithms to reduce the impact of individual items. In modules 1-4, algorithm scores are compared with cut-off scores to provide one of three classifications: autism, autism spectrum, and nonspectrum. The difference between autism and autism spectrum classifications is one of severity, with the former indicating more pronounced symptoms. In the Toddler Module, algorithms provide "ranges of concern" rather than classification scores.

Toddler Module is for children between 12 and 30 months of age who do not consistently use phrase speech. Module 1 is used with children who use little or no phrase speech. Subjects that do use phrase speech but do not speak fluently are administered Module 2. Since these modules both require the subject to move around the room, the ability to walk is generally taken as a minimum developmental requirement to use of the instrument as a whole. Module 3 is for younger subjects, who are verbally fluent, and Module 4 (Lord et al., 1994) is used with adolescents and adults who are verbally fluent and this was the Module used in this study.

Results
The sample consisted of 43 service users 31 males (72 per cent) and 12 females (28 per cent) with an average age of 34 years (SD = 9.89) (male = 32.2 years, female =36.6 years, non-significant difference \( t(39) = 1.312, p = 0.197 \)). Of these, 15 were diagnosed with ASD (34 per cent) 12 males (80 per cent) and 3 females (20 per cent). The diagnostic rate for men was 38 per cent and for females 25 per cent. The mean ADOS score for men who were diagnosed was 11.5 (SD = 2.4) and for women was 6 (SD = 2.5) whilst for men who were not diagnosed was 5.7 (SD = 3.9) and for women 4 (SD = 2.1). Correlational analysis showed a significant difference between the mean scores of males and females who received an ASD diagnosis \( t(13) = 3.34, p = 0.005 \) but no difference between the mean scores of males and females who were not diagnosed \( t(26) = 1.21, p = 0.237 \) (Tables I and II).

<table>
<thead>
<tr>
<th>Table I</th>
<th>Showing the descriptive statistics of patients who received an ASD diagnosis</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>n</td>
</tr>
<tr>
<td>Females who received a clinical diagnosis</td>
<td>3</td>
</tr>
<tr>
<td>Males who received a clinical diagnosis</td>
<td>12</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Table II</th>
<th>Showing the descriptive statistics for patients who did not receive an ASD diagnosis</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>n</td>
</tr>
<tr>
<td>Females who did not receive a clinical diagnosis</td>
<td>9</td>
</tr>
<tr>
<td>Males who did not receive a clinical diagnosis</td>
<td>19</td>
</tr>
</tbody>
</table>
Discussion

In terms of the age of the people presenting at the Service, there is no information relating to the UK to make comments about comparisons with other services. It is interesting though that the age range was similar between males and females who were in their mid-30s suggesting that this group maybe the one either more targeted by awareness campaigns or most in need.

The approximate ratio of males to females of 7:3 shows that more men present to diagnostic services and is consistent with other studies (Loomes et al., 2017). Of the people referred to this Service, only 34 per cent received a diagnosis suggesting that a better screening method of referrals before the clinical diagnosis process starts could increase the diagnostic rate. Although the diagnostic rate for men was higher than females, this was not by much and with such a small sample, cannot be said to reflect a Service bias in the diagnostic process.

In terms of the diagnostic process, it included the ADOS score and information collected through a psychiatric history resulting in an outcome based on the DSM-5 criteria of either receiving an ASD diagnosis or not. Our findings suggest that as part of this process, an overreliance on the ADOS scores may result in gender bias against females as we found that females who received a DSM-5 ASD diagnosis scored significantly lower on the ADOS than men.

As things stand, the ADOS diagnostic thresholds for ASD are the same for both males and females, i.e. $\geq 10$ for autism and 7-9 for autism spectrum. In our study, however, the females who received a clinical ASD diagnosis scored on average lower than the diagnostic threshold for autism spectrum whilst the same did not apply to men. The scores, however, for both men and women who did not receive an ASD diagnosis were clearly below the ADOS diagnostic threshold. As a result, it can be concluded that the sensitivity of the ADOS to ASD in females is lower than in men and as such, developing different thresholds for males and females is indicated.

As we discussed earlier, it is well known that there are gender differences in ASD that can either be due to nosological or diagnostic challenges (Lai et al., 2015). The idea that ASD was an example of the “extreme male brain” (Baron-Cohen, 2002) with males often demonstrating more repetitive behaviours and fixed interests than females (Gould and Ashton-Smith, 2011) with greater difficulties with communication (Rynkiewicz et al., 2016) was also highlighted. What this means in clinical practice is that because females tend to make greater use of facial emotions and gestures as a form of expression and are usually more open with discussing their feelings and emotions than males with ASD (Head et al., 2014) may receive lower scores than males during the ADOS assessment and as a result not reaching the ADOS diagnostic threshold and not receiving an ASD diagnosis.

Future studies should look to gather a greater sample size, using data from service users from different geographical locations and also looking to examine each ADOS profile (social behaviour and communication) as part of the analysis to show potential differential effects by domain.

Limitations

This study has limitations and the obvious one is the relatively small sample size. Given the variation in ADOS scores which can be observed in ASD assessments, the results would bear more statistical power and validity if a larger number of participants were recruited. A second limitation is that the sample was taken from a single geographical location (West Yorkshire) albeit large. For the purposes of this study though we do not expect this would have affected the results as gender differences in ASD are the same across geographical locations. A strength of this study is that the sample was taken from service users of an NHS Service reflecting day to day clinical practice. This contrasts with previous literature which typically recruits participants from known research databases and Autism networks, thereby reducing the ecological validity of their results.

Conclusions

In diagnosing ASD, it is likely that the diagnostic tools used for that purpose play a part in the gender bias favouring males receiving a diagnosis. In this study, we identified that the ADOS is less sensitive in identifying women than men and this maybe because that tool is designed to capture a type of ASD described by the diagnostic criteria which more commonly presents in men.
Future research should focus on refining current diagnostic tools to be more sensitive to women, developing new diagnostic tools for women and describing better the presentation of ASD in women so there can be different descriptors in the diagnostic criteria.

References


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A pilot study on combining risperidone and pivotal response treatment on communication difficulties in children with autism spectrum disorder

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Abstract
Purpose – The purpose of this paper is to investigate the effects of combined risperidone (RIS) and pivotal response treatment (PRT) in children with autism spectrum disorder (ASD).
Design/methodology/approach – In all, 34 children diagnosed with ASD (mean age of 12.36 years) were randomly divided into two groups: an RIS treatment group (n = 18) and an RIS plus PRT (n = 16).
Communication skills were evaluated with the child communication checklist (CCC).
Findings – Total score of the CCC was increased in both groups after three months compared with the score prior to treatment. The total score of the CCC was significantly higher in the combined treatment group than in the RIS group.
Originality/value – Treatment with RIS combined with PRT may result in a better outcome in communication skill for children with autism than RIS training alone.
Keywords Interventions, Autism spectrum disorder, Social communication disorder
Paper type Research paper

Introduction
Autism spectrum disorder (ASD) is a neuro-developmental disorder that is defined by impairments in social interaction and communication as well as repetitive and restricted patterns of behavior that begin in early childhood (Chakrabarti and Fombonne, 2001). An internationally increasing accepted prevalence estimate for ASD among school-aged children is approximately 1 percent (Baird et al., 2006).

Children with ASD may experience delays in the onset of verbal expressive language (Koegel et al., 2016), and some may have difficulty using communication effectively to accomplish social interactive goals (Donno et al., 2010). These communication and language abnormalities create considerable challenge for children and their parents (Landa, 2007), including increased disruptive behaviors (Carr and Durand, 1985), academic difficulties (Catts, 1993), reduced levels of play (Ungerer and Sigman, 1984), and so on and interfere with learning and activities of daily living. Thus, the need for effective interventions that address communication is critical.

Children with ASD often require multimodal treatment including specialized educational interventions (Council, 2001), behavior therapy (Schreibman, 2000) and medication (Volkmar et al., 2004).

The most used intervention options for ASD are derived from the field of behavior analysis (ABA) based on theories of learning and operant conditioning (Lovaas, 1987), as they are...
evidence based. While the structured ABA procedures are very effective in producing behavioral
cchanges in a broad range of areas, these often require massive numbers of trials presented
repeatedly in an analog teaching paradigm for the children to show success. This can be
extremely time consuming for all involved (Koegel et al., 1998).

Pivotal response treatment (PRT), a child-directed naturalistic behavioral method,
develops from these structured ABA approaches (Koegel et al., 1999). This method targets
core pivotal areas and relies on operant teaching principles and has been used to target
a wide range of deficits, including behavioral problems, social skills and communication
(Handleman and Harris, 2001).

Recent evidence suggests that PRT intervention was more effective at improving disruptive
behavior and social communication skills for children with autism than the structured ABA

Numerous studies have attempted to explain the empirical basis for pharmacotherapy in children
with ASD (Volkmar et al., 2004). To date, the use of medication in ASD has mostly focused on
core symptoms with varying degrees of success (Hollander et al., 2005; Ghaeli et al., 2014).

Several studies have investigated the effectiveness of risperidone in the treatment of autism;
however, their results have been uncertain especially in the domain of the social interactions.
Some of these studies have reported a modest or even no effect of risperidone in improvement of
social interactions as well as in communication skills (McDougle et al., 2005; McCracken et al.,
2002; Shea et al., 2004). In contrast, several studies have concluded that risperidone may be
effective in improvement of social interactions (Masi et al., 2001; Ghaeli et al., 2014).

It seems that decreasing disruptive behavior with RIS alone does not result in improved language
and PRT alone would not be successful for communication improvement if the child’s disruptive
behavior was not treated. To expand these results, we attempt to evaluate the combined effects
of medication and PRT in children with ASD.

In this work, we proposed that combining a behavioral intervention and medication could
contribute to greater improvement in social interaction and communication skills when compared
to medication alone. The aims of this paper are: first, to evaluate the efficacy and safety of
combining risperidone plus PRT over a period of three months in 36 children with ASD; second,
to compare effects of combining risperidone plus PRT to risperidone alone. The finding of this
study will help professionals involved in treatment process of children with ASD to select best
model for intervention. The specific questions asked in this study were:

RQ1. Would the combined treatment group show greater improvement on disruptive behavior
and targeted language areas compared to medication only group?

RQ2. Would the combined treatment contribute to greater generalized gains in untreated areas
as measured by a standardized communication checklist (CCC) (Bishop, 2003) completed
by each participant’s parent gains?

Method

Participants

In all, 34 children, 22 boys and 14 girls, ranging in age from 7 to 16 years, participated in this
study. The participants were recruited from referred patients to a university rehabilitation center
in Iran, for treatment. All children were diagnosed with ASD by a child psychiatrist according to
the DSM-5® (AP Association, 2013). The diagnosis was supported with social communication
questionnaire and first author screened each child for symptoms of ASD prior to the start of the
study. In addition, each child was diagnosed by the public school system and placed in special
education classrooms for children with ASD. Other inclusion criteria were as follows:
medication free for two weeks for most psychotropic drugs; if taking anticonvulsant,
seizure-free for six months or greater and with stable dose for four weeks, Body weight ≥ 15 kg,
intellectual quotient of 50 or higher indicating that they exhibited mild intellectual impairments;
and using expressive verbal communication with a mean length of utterance (MLU) of at least
two words. Exclusion criteria included previous adequate trial of risperidone, other co-morbid psychiatric disorder, unstable seizure disorder, or significant abnormality on routine laboratory tests. An outside treatment record indicated that none of the children received any other type of intervention during this study.

**Measures and procedure**

This 12-week study was undertaken during March-June 2017 in which prior to the start of intervention, children were selected based on the predetermined list of criteria that was necessary for inclusion in the study. Then, a total of 18 pairs were matched by age, gender and MLU. Each subject in each pair was then randomly assigned to risperidone plus PRT or risperidone alone. Once randomized, all subjects were assessed one time every two weeks at both of intervention. Two clinicians who were unaware of treatment assignment followed each subject through all phases of the trial a treating clinician who reviewed side effects and adjusted the dose of drug and an independent evaluator who followed improvement response. The PRT was delivered by speech/language specialists who had at least five years of experience teaching children with ASD. They read “How to teach pivotal behaviors to children with autism: A training manual” (Koegel et al., 1989). All intervention sessions were implemented in play rooms that contained a table, chairs, and stimulus materials. This study was part of a primary trial that was performed in accordance with the Declaration of Helsinki and approved by the ethics committee at the case study university in Iran. Written informed consents were obtained from the parents of subjects before the study. Primary trial was registered with the Iranian Clinical Trials Registry (IRCT2017030532884N1).

During the study, two subjects gave up to participate in the study. For two subjects, this was related to socioeconomic problems. The baseline characteristics of participant are showed in Table I.

The randomization was based on an assumption that families would prefer the combined treatment over the drug only treatment.

After 12-week intervention, the medication was gradually withdrawn over a four-week period with regular monitoring for up to four additional weeks by a clinician blind to treatment group. Although not directly probed, no caregivers reported adverse effects of the medication and behavioral treatment and no participants discontinued medication due to adverse events during the study.

The study outcomes focused on improving communication skills by expanding the child’s MLU. For the children that participated in the PRT intervention, a variety of child-chosen foods, toys, and activities were provided for rewards. In order to assess each child’s gain on the behavior that was targeted (MLU[1]) during the intervention sessions, language samples were collected both prior and following intervention. ASD diagnostic measures, medical and intellectual

<table>
<thead>
<tr>
<th>Table I Baseline characteristics</th>
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<tbody>
<tr>
<td>Variable</td>
</tr>
<tr>
<td>Age</td>
</tr>
<tr>
<td>MLU</td>
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<tr>
<td>Age of onset</td>
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<tr>
<td>Weight</td>
</tr>
<tr>
<td>Speech</td>
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<td>Syntax</td>
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<td>Inappropriate initiation</td>
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<td>Coherence</td>
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<td>Social relationship</td>
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<td>Interests</td>
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</table>
assessments were conducted pre-treatment to confirm eligibility and to characterize the sample. In order to assess any generalized gains, each child’s parent was given Children’s CCC pre-treatment and Week 12.

**Clinical global impressions-improvement (CGI-I) scale**

The CGI-I measures overall symptomatic change compared to baseline (Guy, 1976). It is rated on a seven-point scale: 1 = very much improved since the initiation of treatment; 2 = much improved; 3 = minimally improved; 4 = no change from baseline (the initiation of treatment); 5 = minimally worse; 6 = much worse; 7 = very much worse since the initiation of treatment.

**CCC**

The CCC is suggested for children age 4–16; 11 years of age. It consists of 70 items that aims to probe communication difficulties using nine subscales. The first two include the structural characteristics of a verbal interaction: speech; and syntax. The pragmatic domain includes five subscales: inappropriate initiation, coherence, stereotyped language, use of context, and rapport. The sum of these scales is called the “pragmatic composite.” The last two scales are social relationship; and interests that present the child’s nonverbal skills in everyday contents. This checklist is particularly sensitive to children with ASD as it discovers deficits not identified by other communication assessments, as it identifies pragmatic language deficits that are not assessed by language tests that focus exclusively on language fundamentals (Bishop, 2003).

**Interventions**

*Risperidone dose schedule*

As in Schahil study, first started with 0.25 mg at night for children between 14 and 20 kg followed by 0.25 twice a day on day 4; children over 20 kg started on 0.5 mg. The dose was gradually increased over a four-week period as tolerated to a maximum of 2.5 mg/day for smaller children (< 45 kg) and up to 3.5 mg/day for larger children (≥ 45 kg) (Sandler et al., 2002). Treating clinicians could delay an increase or reduce the dose to manage adverse effects.

*Dose reduction*

After week 12, the medication was reduced to 75 percent of the maintenance dose for week 13; to 50 percent for weeks 14 through 15; to 25 percent for week 16, and to 0 for weeks 17 through 20. Subjects were monitored weekly for signs of relapse (i.e. a score of “much worse” or “very much worse” on the CGI-I).

**PRT**

The PRT intervention was based on the published manual, *PRT: Using Motivation as a Pivotal Response* (Koegel, 2011). In this intervention instead of the clinician randomly selecting a stimulus item, items were selected according to the child’s preference for any given item for any given trial. The task was varied so that the reward was provided both for responses that had previously been mastered (in this study, shorter utterance) diffused with rewards for acquisition tasks (in this study, longer utterances). The reinforcement contingency was broadened so that if the child imitated either the exact correct response or a successive approximation, or made any clear verbal attempt to respond, the child was reinforced. Instead of the child being reinforced with edibles and praise, the child was reinforced with the opportunity to play with the instructional stimulus, paired with verbal praise.

Treatment sessions were conducted triple weekly for 45 minutes per session over a 12-week-period. Thus, each child received a total of 27 hours of intervention. Parents were informed that their children would receive speech and language services, but were blinded to the specific target behavior (expanding MLU). None of the parents was present during the intervention sessions. For all children, the target behaviors were the same and involved
expanding the children’s MLU using recast procedures (Nelson et al., 1996). The materials in the PRT intervention consisted of child-chosen items and activities.

Throughout the study, each child was observed at least four times and was scored for Fidelity of Implementation (FoI) by the first author. FoI was scored for a total of 10 minutes in one-minute intervals, and each of the seven points were scored as correct (+) or incorrect (−) according to each of the variables outlined in the following definitions for FoI. As noted in the definitions below, the speech-language specialists had to obtain the child’s attention, provide a clear opportunity, and provide contingent consequences. FoI for treatment providers ranged from 80 to 90 percent and never fell below the required 80 percent:

Definitions for FoI:
1. Child attending
   - The interventionist must have the child’s attention prior to presenting an opportunity.
2. Clear opportunity
   - The question/instruction/opportunity to respond must be clear and appropriate to the task.
3. Child choice
   - The interventionist should follow the child’s choice with tasks and activities. However, he interventionist must always assume control should the child engage in hazardous (i.e. self-injury) or inappropriate (i.e. self-stimulation) activities. If child is not showing interest in the current task, interventionist should attempt to change the activity.
4. Maintenance tasks
   - The interventionist should intersperse tasks the child can already perform with acquisition (new) tasks.
5. Contingent
   - Reinforcement must be contingent upon child’s behavior. The interventionist’s response (i.e. giving the child a reinforcer) must be dependent upon the child’s response (i.e. saying “little toy”).
6. Natural
   - Reinforcement should be natural or directly related to the desired behavior.
7. Contingent on attempts
   - Any goal-directed attempt to respond to questions, instructions, or opportunities should be reinforced. Although an attempt does not necessarily need to be correct, it has to be reasonable.

Statistical analysis
All data were analyzed with SPSS 16.0 software. Baseline variables were analyzed between groups using the independent t-test. The measures used were mean, standard deviation, percentages, and frequencies. Analysis of covariance (ANCOVA) and paired t-tests were used to compare the CCC scores between treatment groups during the study. Categorical variables were analyzed using the χ² test. A p-value of less than 0.05 was considered to be significant.

Results
Analyses of demographic data were conducted to assess for the possible differences between the RIS plus PRT and RIS groups. An independent samples test showed no significant difference between the two groups in regard to age, weight, MLU and CCC subscales prior to the start of intervention.

Following the 12-week intervention, the result of ANCOVA shows that the MLU and the mean of CCC subscales score for the RIS group improved slightly, but non-significantly. On the other hand,
significant improvements were seen in the RIS plus PRT group. Thus, the RIS plus PRT group showed significantly greater gains in MLU and CCC following the three-month intervention (Table II).

Furthermore, the paired t-test revealed significant differences between baseline and endpoint measures of the most CCC subscales in the two groups, with the RIS plus PRT group showing more gains on this measure as well (Table III).

<table>
<thead>
<tr>
<th>Table II</th>
<th>MLU and CCC subscale scores for RIS and RIS plus PRT groups</th>
</tr>
</thead>
<tbody>
<tr>
<td>Variables</td>
<td>RIS group (n = 17)</td>
</tr>
<tr>
<td>MLU</td>
<td>591/0 ± 60/3</td>
</tr>
<tr>
<td>Speech</td>
<td>20/3 ± 53/30</td>
</tr>
<tr>
<td>Syntax</td>
<td>87/1 ± 53/27</td>
</tr>
<tr>
<td>Inappropriate initiation</td>
<td>30/2 ± 06/24</td>
</tr>
<tr>
<td>Coherence</td>
<td>36/3 ± 94/26</td>
</tr>
<tr>
<td>Stereotyped language</td>
<td>71/3 ± 24/26</td>
</tr>
<tr>
<td>Use of context</td>
<td>23/2 ± 88/23</td>
</tr>
<tr>
<td>Rapport</td>
<td>106/4 ± 12/26</td>
</tr>
<tr>
<td>Social relationship</td>
<td>34/2 ± 53/24</td>
</tr>
<tr>
<td>Interests</td>
<td>70/1 ± 53/27</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Table III</th>
<th>Mean ± SD of the two treatment groups on the subscales of CCC and MLU</th>
</tr>
</thead>
<tbody>
<tr>
<td>Variable</td>
<td>Baseline mean ± SD</td>
</tr>
<tr>
<td>MLU</td>
<td></td>
</tr>
<tr>
<td>Risperidone</td>
<td>60/0 ± 54/3</td>
</tr>
<tr>
<td>Risperidone + PRT</td>
<td>80/0 ± 23/3</td>
</tr>
<tr>
<td>Speech</td>
<td>15/3 ± 29/30</td>
</tr>
<tr>
<td>Risperidone + PRT</td>
<td>29/4 ± 88/28</td>
</tr>
<tr>
<td>Syntax</td>
<td>80/1 ± 35/27</td>
</tr>
<tr>
<td>Risperidone + PRT</td>
<td>78/1 ± 94/26</td>
</tr>
<tr>
<td>Inappropriate initiation</td>
<td>38/2 ± 76/23</td>
</tr>
<tr>
<td>Coherence</td>
<td>22/3 ± 65/26</td>
</tr>
<tr>
<td>Risperidone + PRT</td>
<td>27/3 ± 76/26</td>
</tr>
<tr>
<td>Stereotyped language</td>
<td>04/4 ± 41/25</td>
</tr>
<tr>
<td>Risperidone + PRT</td>
<td>70/3 ± 71/24</td>
</tr>
<tr>
<td>Use of context</td>
<td>29/2 ± 65/23</td>
</tr>
<tr>
<td>Risperidone + PRT</td>
<td>14/2 ± 71/22</td>
</tr>
<tr>
<td>Rapport</td>
<td>95/3 ± 53/25</td>
</tr>
<tr>
<td>Risperidone + PRT</td>
<td>23/3 ± 65/25</td>
</tr>
<tr>
<td>Social relationship</td>
<td>29/2 ± 18/24</td>
</tr>
<tr>
<td>Risperidone + PRT</td>
<td>05/3 ± 29/25</td>
</tr>
<tr>
<td>Interests</td>
<td>82/1 ± 41/27</td>
</tr>
<tr>
<td>Risperidone + PRT</td>
<td>92/1 ± 41/27</td>
</tr>
</tbody>
</table>
The frequencies of adverse events are summarized in Table IV. There was no significant difference between the two treatment groups.

The model proposed in this study indicates that RIS plus PRT will promote greater gains in communication skills than RIS alone.

Discussion

Studies aimed to compare medication with combined medication and a psychosocial intervention are becoming more common in child mental health (Abikoff et al., 2004; Pediatric, 2004).

The results of this study showed that both of the groups were effective in increasing communication skills; however, the PRT intervention plus RIS was more effective at improving language abilities and social communication skills for children with autism than the RIS alone. The children who participated in PRT plus RIS group demonstrated greater gains in both the targeted area (MLU) as well as in pragmatic skills, including inappropriate initiation, coherence, stereotyped language, use of context, and rapport, as measured by the CCC. Thus, the motivational components of PRT were more effective in improving in social communication (Koegel et al., 1987).

To date, studies of combined treatments in child psychiatry have directed both treatments at the same outcome. If the drug treatment has a large effect, it may be difficult to show an additive effect of the psychosocial intervention simply because there is little or no possibility for improvement on the primary outcome measure. Thus, a fundamental design question for this study was whether medication and PRT should be directed at the same (communication skills) or separate outcomes.

The clear superiority of risperidone over placebo in previous studies obviated the need for a placebo group on scientific and ethical grounds (Scahill et al., 2008; Scahill et al., 2009). The remarkable effect of PRT also challenged the inclusion of a PRT only comparison group (Mohammad Zaheri et al., 2014, 2015).

Despite prior evidence showed the significant and enduring reduction in serious behavioral problems with risperidone over three months; however, only modest improvements on social and communication skills observed after six months of treatment (Williams et al., 2006; Aman et al., 2009). The improvement in disruptive behaviors is a prerequisite, or a mediator, for improvement in adaptive functioning (Shrout and Bolger, 2002). Therefore, the rationale could be that RIS or PRT alone (for communication improvement) would not be successful if the child’s disruptive behavior was not treated and increasing improvement in disruptive behavior suggests that improvement in communication skills depends, at least in part, on change in behavior. It seems decreased disruptive behavior can facilitate learning language and communication skills.

Consequently, we proposed a model in which behavioral problems interfere with the acquisition of language and new communication skills. Medication plays a necessary role by reducing the serious behavioral problems (tantrums, aggression and self-injury) and sets the stage for the success of PRT on improving communication skill. We predicted that, compared to medication alone, medication plus PRT will be associated with significantly greater improvement in communication skills as measured by the CCC.

### Table IV

| Variables            | RIS group (n = 17) N (%) | PRT + RIS group (n = 17) N (%) | P  
|----------------------|--------------------------|-------------------------------|---
| Increased appetite   | 4 (23.5)                 | 3 (17.6)                      | 0.671  
| Nausea               | 2 (11.8)                 | 2 (11.8)                      | 1  
| Urinary problems     | 3 (17.6)                 | 2 (11.8)                      | 0.682  
| Increased weight     | 2 (11.8)                 | 2 (11.8)                      | 1  
| Sedation             | 1 (5.9)                  | 1 (5.9)                       | 1  
| Dizziness            | 1 (5.9)                  | 2 (11.8)                      | 0.545  
| Constipation         | 3 (17.6)                 | 3 (17.6)                      | 1  |
This study offers an approach to the study of combining medication and behavioral intervention in which treatments are not directed at the same outcome.

There are some limitations in our study. The present study was performed on a small number of participants, based on an open-label design and used only two instruments for its outcome measures. Therefore, although our findings illustrate that risperidone treatment fails to improve communication skills, we cannot conclude that risperidone has no efficacy.

Note
1. The total number of words the child emitted was divided by the total number of utterances, to yield an MLU.

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Chronic health and lifestyle problems for people diagnosed with autism in a student-led clinic

Barry Tolchard and Cynthia Stuhlmiller

Abstract

Purpose – People with autism spectrum disorder (ASD) are at greater risk of developing chronic health and risky lifestyle problems. This is exaggerated further for people living in rural settings and from cultural backgrounds traditionally underserved by healthcare services. The purpose of this paper is to describe an evaluation of health and behavioural lifestyle outcomes of people diagnosed with ASD in a student-led clinic in rural/regional Australia.

Design/methodology/approach – Routine clinical outcomes and lifestyle measures were routinely collected at a primary acre student-led Clinic in rural/regional Australia. Participants were all attending the clinic who provided consent for their routine date to be reported. Participants ranged in age from new born to 100 years and were representative of the local community.

Findings – The results indicate there is an increased risk for people with ASD developing chronic conditions compared to those without a diagnosis. This also resulted in higher body mass index and blood sugar levels linked to diabetes and hypertension. Mental health problems were common in people diagnosed with ASD especially anxiety disorders. Smoking was problematic for people with ASD but mainly in non-Aboriginal and Torres Strait Islanders. Alcohol use was not an increase risk in ASD.

Originality/value – Little is reported on the health and lifestyle experiences of people with ASD in rural/regional settings, especially from Aboriginal and Torres Strait Islander communities. This paper gives an initial insight to the presentation of chronic conditions and harmful lifestyle choices. Possible insights into adapting or modifying care for people with ASD in rural/regional Australia are given.

Keywords Health, Autism spectrum disorder, Complex needs, Rural, Comorbidity, Student-led clinic

Purpose

There are complex structural and social determinant of health issues affecting people living in regional and rural communities in Australia. Residents have lower life expectancy and increased risks of chronic disease. The health disparities of vulnerable groups in rural and regional Australia are pronounced especially among the indigenous nations (Marmot, 2011; McDonald et al., 2013). People with mental health problems including diagnoses of autism spectrum disorder (ASD) experience greater health disparities when living in rural and remote settings (Kelly et al., 2011) further heightened in indigenous peoples with mental health problems (Sayers et al., 2017). In addition to specific health inequities, regional and rural Australians experience problems of poor housing, limited educational opportunities and higher rates of unemployment or frequent low-paid work (National Rural Health Alliance, 2013).

The 2012 prevalence of ASD in Australia was 0.5 per cent or > 115,000 people. This is an increase of over 60,000 from the 2009 prevalence figure (Australian Bureau of Statistics, 2014). There are no formal mechanisms by which general practitioners (GPs) can diagnose autism in primary care in Australia which leads to inconsistencies in caring for people with such a diagnosis (Australian Medical Association, 2016). Formal diagnoses of autism in Australia varies by state.
In New South Wales, paediatricians, psychiatrists or a multi-disciplinary team provide ASD diagnoses. Once diagnosed with Autism the person can receive care through a GP mental health treatment plan (Australian Government Department of Health and Aging, 2011).

It is purported that people with autism experience increased rates of co-morbid chronic conditions including mental health problems (Croen et al., 2015; Hinckson et al., 2013; Tyler et al., 2011). Simoff et al. (2008) found 84 per cent of people with ASD had a co-morbid mental health problem especially anxiety disorders and other developments issues. This is supported in a retrospective case review of 474 adults diagnosed with autism which found a significant risk of anxiety disorders and obsessive-compulsive disorder (Russell et al., 2016). People diagnosed with ASD also experience co-morbid health problems including diabetes (Chen et al., 2016), cardiac, hypertension (Heffernan et al., 2017) and respiratory diseases (Axmon et al., 2017).

In most cases, the co-morbid conditions are those associated with social issues including lower physical activity and obesity.

There is a disparity between people with autism receiving adequate help in rural areas compared to urban settings and indigenous peoples receive further inconsistent care (Farmer and Reupert, 2013; Murphy and Ruble, 2012). In addition, people with ASD often report unhealthy or risky behaviours including inactivity, over-eating and anti-social contact with others (Hill et al., 2014; Jones et al., 2017; McCoy et al., 2016). Alcohol and tobacco use varies in ASD. It is reported that people with ASD experience lower rates of smoking and drinking of alcohol (Mangerud et al., 2014). However, in an Australian twin study those with ASD traits had an elevated risk of both smoking and drinking (Sizoo et al., 2010). The inability to access public awareness or community support programmes are reasons for poor integration of people with autism into local life especially when living in regional and rural settings where there are limited choices for the whole population.

The primary aim of this study was to examine whether people with autism experience higher rates of chronic health problems. A secondary aim was to determine if people with a diagnosis of autism experience greater lifestyle problems including obesity and increased risk of drinking and smoking. Finally, the study aimed to determine if there were specific demographic differences in both chronic health presentations and harmful behavioural choices.

Methods

Setting and services

All subjects attended a student-led primary care clinic (SLC) based in a local community health centre. This clinic addressed the shortage of health professional student places in regional and rural Australia, especially nursing (Stuhlmiller and Tolchard, 2015, 2017). Clinic attendee data were entered into a patient management database – Best Practice, which provides output, outcomes and financial reporting (www.bpsoftware.net). A local Aboriginal Health Service managed the service.

Clinic participants

Clinic attendees considered at risk of ASD received a diagnosis prior to receiving care in the SLC. A GP, Nurse practitioner or mental health specialist supported student-led treatments. Management of all attendees with a diagnosis of ASD was through their GP mental health Treatment plan. Distinctions are made between gender and age as well as if someone identifies as Aboriginal or Torres Strait Islander. The percentage of people identifying as Aboriginal was high compared to the surrounding region. No other ethnic distinctions were made as those not identifying as indigenous were white Australians.

Table I gives the demographic profile of the clinic compared with the local community. There were no significant differences between demographic profile and whether someone was diagnosed with autism. Compared to the wider community, clinic attendees were more likely to be Aboriginal or Torres Strait Islander, female and young (Table I).

Of one 1,564 clinic attendees assessed 3.84 per cent or 60 people were diagnosed with autism. This figure is from a sub-sample at risk of ASD, therefore, the actual percentage in the clinic is likely
much lower. The demographic profiles of people with autism, not surprisingly, were younger ($\chi^2(1, 564) = 327.49, p < 0.001$) and male ($\chi^2(1, 564) = 36.10, p < 0.001$). There were no differences on whether someone identified as Aboriginal or Torres Strait Islander.

**Statistical analysis**

Comparisons were made between those with and without ASD. Continuous variables were treated as dependent with ANOVA statistics being performed to determine differences. All categorical and nominal data were reported using either descriptive or non-parametric statistics. Risk of chronic disease with ASD was established using odds ratios with lower and upper limits.

**Findings**

**Health problems**

Overall, people diagnosed with ASD were at greater risk of co-morbid health and mental health problems compared with those without a diagnosis (Table II). The increased risks were 4.52 times higher for mental health, 4.37 times higher for cardiac problems, 3.34 times higher for diabetes and 6.27 times higher for hypertension. There was no increased risk for renal or respiratory problems. When examining the nature of the mental health problem, all diagnosed with ASD had an anxiety disorder and were over 18 years old.

**Lifestyle problems**

Obesity: Body mass index (BMI) was recorded on two occasions – assessment, then at 12-week follow-up. When comparing BMI at risk groups, people diagnosed with ASD were at greater risk

---

**Table I** Demographic profile of clinic attendees

<table>
<thead>
<tr>
<th>Demographic Profile</th>
<th>Clinic (%)</th>
<th>Region (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Gender</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>728 (43.1)</td>
<td>2,600 (48.6)</td>
</tr>
<tr>
<td>Female</td>
<td>961**</td>
<td>2,747 (51.4)</td>
</tr>
<tr>
<td><strong>Aboriginal or Torres Strait Islander</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Yes</td>
<td>1,053***</td>
<td>1,066 (19.9)</td>
</tr>
<tr>
<td>No</td>
<td>515 (32.8)</td>
<td>4,281 (80.1)</td>
</tr>
<tr>
<td><strong>Age</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>0-14</td>
<td>927***</td>
<td>26.11 (26.11)</td>
</tr>
<tr>
<td>15-24</td>
<td>515 (19.95)</td>
<td>15.38 (15.38)</td>
</tr>
<tr>
<td>25-44</td>
<td>513 (19.87)</td>
<td>18.62 (18.62)</td>
</tr>
<tr>
<td>45-64</td>
<td>479 (18.55)</td>
<td>23.18 (23.18)</td>
</tr>
<tr>
<td>65+</td>
<td>148 (5.73)</td>
<td>16.70 (16.70)</td>
</tr>
</tbody>
</table>

Notes: Gender $\chi^2(1) = 53.50, p < 0.001$; ATSI $\chi^2(1) = 489.05, p < 0.001$; Age $\chi^2(1) = 99.20, p < 0.001$. ***$p < 0.001$

---

**Table II** Odds ratios for diagnoses of ASD and other health-related problems

<table>
<thead>
<tr>
<th>Health-related Problem</th>
<th>OR</th>
<th>LL</th>
<th>UL</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mental Health (MH)</td>
<td>4.52</td>
<td>1.79</td>
<td>11.42</td>
</tr>
<tr>
<td>Cardiac (C)</td>
<td>4.37</td>
<td>1.43</td>
<td>13.29</td>
</tr>
<tr>
<td>Diabetes (D)</td>
<td>3.34</td>
<td>1.12</td>
<td>9.96</td>
</tr>
<tr>
<td>Hypertension (H)</td>
<td>6.27</td>
<td>1.99</td>
<td>19.81</td>
</tr>
<tr>
<td>Renal (R)</td>
<td></td>
<td></td>
<td>ns</td>
</tr>
<tr>
<td>Respiratory (Re)</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Notes: MH: $\chi^2(1, 564) = 12.12, p < 0.001$; C: $\chi^2(1, 564) = 7.98, p < 0.01$; D: $\chi^2(1, 564) = 5.24, p < 0.05$; H: $\chi^2(1, 564) = 12.73, p < 0.000$. OR, odds ratio; LL, lower limit; UL, upper limit.
of being overweight or obese (ASD: Odds ratio = 1.44 (0.73-2.86)). Therefore, people with ASD were 1.44 times more likely to be overweight or obese. All clinic attendees with a BMI recording showed reductions in their scores (Table III). A one-way analysis of variance of the BMI mean difference revealed no significant difference between changes in BMI for those with or without ASD. However, those without ASD had a larger mean difference. In both groups, there was a shift from obese to overweight.

**Blood sugar levels (BSL).** A one-way multivariate analysis of variance (one-way MANOVA) was run to determine the effect of a diagnosis of ASD and blood sugar levels. Two measures of blood sugar were assessed: BSL and fasting blood glucose (FBG). There was homogeneity of variance-covariance’s matrices, as assessed by Box’s test of equality of covariance matrices ($p = 0.085$) and there was homogeneity of variances, as assessed by Levene’s test of homogeneity of variance ($p > 0.05$), therefore the one-way MANOVA was able to be carried out. Clinic attendees diagnosed with ASD scored higher in their BSL ($M = 7.56$, $SD = 3.2$ and $M = 6.89$, $SD = 3.8$) and FBG ($M = 6.34$, $SD = 3.0$ and $M = 6.14$, $SD = 4.1$) compared to those without a diagnosis. There was no statistically significant difference between a diagnosis of ASD or no ASD on the combined dependent variables, $F(2, 784) = 0.536$, $p = 0.585$; Pillai’s Trace = 0.001; partial $\eta^2 = 0.001$.

**Tobacco and alcohol use.** There was no difference between having a diagnosis of ASD and alcohol use. There was an increased risk for smoking in people with ASD compared to those without a diagnosis (OR = 2.82, 1.70-4.69; Fishers exact $p = 0.04$). People with ASD were 2.9 times more likely to be smokers (Table IV).

**Discussion**

In summary, people diagnosed with ASD are at increased risk of chronic health problems and the effects of harmful lifestyle choices. Overall, the increased risks ranged between 3.34 and 6.27 times higher than people without ASD. People with ASD were 1.44 times more likely to be overweight or obese. Overall, people with and without ASD showed improvements in their BMI levels while attending the clinic. While there were no differences between someone diagnosed with ASD or not and drinking, people with ASD were 2.9 times at greater risk of being a smoker.

The risk of experiencing co-morbid health and mental health problems in people diagnosed with ASD is higher than those without a diagnosis. In this sample, all diagnosed as such were

### Table III  BMI at assessment, follow-up and mean difference

<table>
<thead>
<tr>
<th></th>
<th>$n$</th>
<th>$M$ (SD)</th>
<th>$M$ (SD)</th>
<th>$M$ (SD)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Autism</td>
<td>60</td>
<td>29.0 (0.16)</td>
<td>26.7 (0.49)</td>
<td>2.2 (0.65)</td>
</tr>
<tr>
<td>No autism</td>
<td>966</td>
<td>28.8 (2.84)</td>
<td>25.8 (2.83)</td>
<td>2.8 (3.84)</td>
</tr>
</tbody>
</table>

**Note:** ANOVA of mean difference ($F(1, 1,023) = 1.17$, $p = 0.28$)

### Table IV  Alcohol and smoking percentages between those with ASD and without ASD

<table>
<thead>
<tr>
<th>Alcohol use</th>
<th>Smoker</th>
<th>Drinker</th>
<th>Non-drinker</th>
<th>Smoker</th>
<th>Non-smoker</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>$n$ (%)</td>
<td>$n$ (%)</td>
<td>$n$ (%)</td>
<td>$n$ (%)</td>
<td>$n$ (%)</td>
</tr>
<tr>
<td>Autism</td>
<td>Yes 12 (20.00)</td>
<td>48 (80.00)</td>
<td>30 (50.00)</td>
<td>30 (50.00)</td>
<td></td>
</tr>
<tr>
<td></td>
<td>No 126 (25.00)</td>
<td>378 (75.00)</td>
<td>120 (23.26)</td>
<td>396 (76.74)</td>
<td></td>
</tr>
</tbody>
</table>

**Notes:** Alcohol use: $\chi^2(1, 564) = 0.346$, $p = 0.56$; Smoking: $\chi^2(1, 564) = 17.49$, $p < 0.001$
experiencing anxiety problems. In terms of mental health, having an anxiety disorder is reported in a number of studies and so these findings confirm this (Reaven et al., 2016; Vasa et al., 2016).

It was also found in this sample that all of those with a co-morbid diagnosis experienced higher BMI and blood sugar levels. In all cases, those with ASD fell within the overweight or obese risk areas. This again has been reported in a small number of studies where being overweight or obese are at least as high as other children and adolescents and in many cases higher (Curtin et al., 2014; McCoy et al., 2016). The raised blood sugar levels and a greater risk of diabetes in people with ASD are likely linked. Therefore, identifying those at risk from this group and targeting the health prevention strategies may require modified or specific approaches. One example, in this study population was to provide 24-hour blood sugar monitoring with lifestyle diaries. This approach helped people understand changes in their blood sugar levels related to their lifestyle choices such as alcohol use, eating and exercise. A future strategy of the SLC could be to provide this monitoring for people with ASD and offer individual or group feedback to try and encourage lifestyle changes.

The clinic offered a number of services to try and alleviate chronic health problems through early intervention including a walking club, after school healthy eating, social club, yoga and smoking cessation classes. These programmes are designed to engage the local community in healthier lifestyle and disease management strategies. Such approaches are known to be effective in people with intellectual disabilities and ASD (Roll, 2018). The various healthy eating and exercise programmes of the clinic appear to have had a positive effect overall on levels of obesity as measured by BMI. While clinic attendees diagnosed with and without ASD showed improved BMI scores over the 12-week period of measurement, those with ASD had a lower BMI change score. Therefore, working with those identified with ASD on better nutrition and exercise on specific goals may prove beneficial. While the evidence is limited, there are studies examining their benefits of such interventions (Ferreira et al., 2018; Srinivasan et al., 2014).

While the SLC provides smoking cessation opportunities, it appears those with ASD continue to smoke. The SLC professionals may need to consider how the cessation messages are being given to this group and modify them accordingly. Little is known about smoking cessation programmes for people with ASD. However, examples of specific programmes have been reported (Tracy and Hosken, 1997).

An unexpected finding was that people identifying as Aboriginal or Torres Strait Islander with a diagnosis of ASD did not appear to be at greater risk than those not identifying as such. Those without a diagnosis of ASD and having smoking and drinking problems were from the Aboriginal and Torres Strait Islander group. This community was nearly 75 per cent Aboriginal and even allowing for weighting of cultural background these difference remained the same. It would be valuable to examine any cultural differences in how Aboriginal clinic attendees with ASD are somehow protected from these lifestyle choices. Perhaps they are receiving more services that help them in this area, or the Aboriginal community which is traditionally close knit may ensure they are not encouraged to smoke and drink. Further investigation will be needed to tease out these issues.

Overall, the risks and protective factors described in this population may reflect the wider health and lifestyle disparities associated with rural and regional settings and social deprivation especially poverty. The area from which the SLC served was considered was of the most deprived in Australia (Australian Bureau of Statistics, 2011). Therefore, inflated risk of chronic disease, obesity and smoking may simply reflect the reality of such a community, whom re are largely underserved and experience multiple deprivations such as high crime, poor housing and limited education. In addition, a community predominantly identifying as Aboriginal and Torres Strait Islander face even greater burdens including vastly reduced life expectancy and ill health. There was a sense of ownership by this part of the community of the SLC and a desire for the clinic to work. This may have been reflected in the people diagnosed with ASD being on certain measures no worse than those without a diagnosis and in some situations being better such as with alcohol use. Parents from Aboriginal and Torres Strait Island backgrounds often volunteered in the clinic activities including offering a soup kitchen in the evenings, providing fresh fruit to children after school and a mother and baby support group, all of which directly addressed the concerns of the community. It is believed this direct community engagement alongside the SLC activities was responsible for reduced petty crime and improved school attendance, through a greater civic pride.
References


Further reading


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A preliminary evaluation of metacognitive beliefs in high functioning children with autism spectrum disorder

Breeanna Campbell, Michelle Curran, Raymond Inkpen, Mary Katsikitis and Lee Kannis-Dymand

Abstract

Purpose – Metacognitive beliefs and processes have been found to perpetuate anxiety and depression in youth and adults. However, the presence of metacognitive beliefs in children with autism spectrum disorder is somewhat unclear and has received limited research attention to date. The purpose of this paper is to explore metacognitive beliefs in children with autism and associations with anxiety and depression.

Design/methodology/approach – In total, 23 high functioning participants (17 male and 6 female) between the ages of 8 and 12 (M = 10.38) diagnosed on the autism spectrum completed the study. Participants completed the Revised Children’s Scale of Anxiety and Depression and the Metacognitions Questionnaire for Children.

Findings – Correlation analyses revealed that positive and negative metacognitive beliefs were found, as hypothesised, to be prevalent in this sample.

Originality/value – Despite methodological limitations, this is one of the first research evaluations to provide evidence for metacognitive beliefs in high functioning children with autism and comorbid anxiety or low mood.

Keywords Anxiety, Autism spectrum disorder, Depression, Metacognitive beliefs

Introduction

The diagnosis of autism spectrum disorder (ASD) appears to be increasing, with rates climbing from 1:150 children in 2000 (Centers for Disease Control and Prevention, 2007), to 1:50 in 2013 (Blumberg et al., 2013), with the CDC reporting rates of 1:68 in 2014. Growing research has established that people with ASD may experience various co-occurring psychiatric comorbidities, with some studies reporting higher prevalence rates of comorbidity in ASD than in non-ASD populations (Mazzone et al., 2012; Vasa and Mazurek, 2015). Further, the management of behavioural problems in children with ASD continues to be a challenge for those who care for and provide treatment for those with ASD, which is often complicated by comorbidity (Mazzone et al., 2012). Research has found that rates of worry, anxiety, and depression are prevalent and significantly elevated in individuals with ASD and substantially impacts their daily functioning and quality of life (Gillott et al., 2001; Jang et al., 2013; Kim et al., 2000; Matson and Nebel-Schwalm, 2007; Simonoff et al., 2008; Strang et al., 2012; Tantam, 2013; White et al., 2009).

A recent meta-analysis found that 39.6 per cent of 2,121 children and adolescents with ASD had at least one comorbid anxiety disorder according to DSM-IV criteria (van Steensel et al., 2011), including specific phobia, obsessive-compulsive disorder (OCD), social anxiety disorder, generalised anxiety disorder (GAD), panic disorder, and separation anxiety disorder. Simonoff et al. (2008) found that 41.90 per cent of 112 children with ASD meet DSM-IV-TR criteria for the following anxiety disorders: specific phobia (8.50 per cent), OCD (8.20 per cent), GAD (13.40 per cent), panic disorder (10.10 per cent), and social anxiety (29.20 per cent). White et al. (2009), in a review of 11 prevalence studies, reported that 11–84 per cent of 1,353 children with ASD experienced anxiety

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to an impairing degree. Depression rates have been estimated between 15 and 24 per cent in children with ASD; notably higher than in children without ASD (Rieffe et al., 2014). Further, worry, anxiety, and depression in ASD populations has been reported to be more prevalent in individuals with functional language and an intellectual quotient (IQ) above 70 (Caamaño et al., 2013; De-la-Iglesia and Olivar, 2015; Hallett et al., 2013).

There is mounting agreement that anxiety and depression are common in ASD; however, there is a limited understanding of the underlying mechanisms (De-la-Iglesia and Olivar, 2015; Mazefsky and Herrington, 2014; White et al., 2009). Further, it is likely that the relationship between anxiety and ASD is bidirectional, in that ASD exacerbates anxiety symptoms and vice versa (Mazefsky and Herrington, 2014; van Steensel et al., 2013). There has also been some debate in the research that the presence of both ASD and depression may exacerbate each other’s condition, as well as increase symptoms of anxiety and OCD (De-la-Iglesia and Olivar, 2015).

Research contends that ASD is a heterogeneous disorder that may be associated with various etiological factors and that psychological treatment outcomes in ASD populations are notably variable in comparison to non-ASD populations (Damiano et al., 2014; Matson, 2016; Mazefsky et al., 2012; Zafeiriou et al., 2007; Zafeiriou et al., 2013). Therefore, it has been recommended that future research needs to explore the various mechanisms that may contribute to comorbid conditions, such as anxiety, in those with ASD; particularly since research to date into interventions has primarily focussed on mechanisms identified to maintain psychological distress in children without ASD (Damiano et al., 2014). One such mechanism that has been identified as a transdiagnostic process that contributes to comorbidity in ASD is poor emotion regulation (Damiano et al., 2014).

Metacognition

The metacognitive model of emotional disorder – the self-regulatory executive function model (S-REF; Wells, 2000; Wells, 2009; Wells and Matthews, 1996) is a transdiagnostic model that asserts that emotional dysregulation and associated negative thoughts are maintained by four interacting constructs: the cognitive attentional syndrome (CAS), metacognitive beliefs, executive control, and mental modes (Wells, 2013). While a detailed account of this model is beyond the scope of this paper (see Wells, 2009, 2013; Wells and Matthews, 1996), the S-REF model proposes that psychological disorder and emotional distress results from the CAS, which is controlled by metacognitive beliefs. The CAS comprises of repetitive and perseverative thinking, in the form of worry, rumination, focussing on threat, and the utilisation of maladaptive coping behaviours (e.g. avoidance, thought suppression) that fail, leading to a paradoxical impact on self-regulation and continued negative emotional experience (Wells, 2009, 2013).

Metacognitive beliefs, or beliefs about one’s thinking, are conceptualised to control and maintain the CAS and are categorised under two main domains: positive metacognitive beliefs and negative metacognitive beliefs (Wells, 2009). Positive metacognitive beliefs typically occur prior to the development of negative metacognitive beliefs. Positive metacognitive beliefs are concerned with the benefits or advantages of engaging in cognitive activities (worry, rumination); for example, “worrying helps me to stop bad things from happening” (Wells, 2009). Such beliefs may involve worry being viewed as a helpful strategy. This results in a reliance and increased use of worry as a coping strategy, for example, an individual with GAD may hold the positive metacognitive belief “worrying means I will be prepared” (Wells, 2009). Negative metacognitive beliefs, which are more potent in psychological disorder, are concerned with either the harmfullness or uncontrollability of one’s thoughts (Wells, 2009, 2013). For example, “worrying is going to give me a heart attack” or “my worry is uncontrollable”. In turn, metacognitive beliefs result in the employment of maladative metacognitive strategies such as thought suppression and avoidance, which maintain worry or rumination and the associated anxiety or negative affect (Cartwright-Hatton and Wells, 1997; Spada et al., 2008; Wells and Butler, 1997).

Several studies have substantiated Wells’ metacognitive theory (Halvorsen et al., 2015; Normann et al., 2014; Wells, 2013). Davis and Valentiner (2000) found in a study of 175 adults that individuals with GAD demonstrated significantly higher levels of metacognitions than non-anxious and non-worried/anxious participants. Wells (2005), in a sample of 174 university students, found that metacognitions are not only evident in GAD, but that metacognitive strategies that result from
negative metacognitive beliefs (e.g. thought suppression, avoidance) further perpetuated worry. More recently, research \((n = 230)\) has suggested that negative metacognitive beliefs mediate the relationship between trait worry and GAD symptoms (Penney et al., 2013).

Further, Halvorsen et al. (2015) reported that metacognitive beliefs, in a sample of 168 adults, were endorsed more by currently depressed individuals than those who were previously depressed or those who had not experienced depression. Additionally, several studies that have examined the outcomes of treatment on metacognitive processes, including metacognitive beliefs, have demonstrated that changes in such beliefs were associated with reductions in worry and rumination and associated emotional distress (Papageorgiou, 2015; Wells, 2013). The role of metacognitive beliefs in emotional disturbance in other forms of anxiety, such as OCD, social anxiety, panic, and health anxiety, has also been demonstrated (Bailey and Wells, 2015; Cucchi et al., 2012; Vassilopoulos et al., 2015; Wells, 2000, 2013; Wells and Papageorgiou, 1998).

**Metacognition in children and adolescents**

Regarding metacognition in children and adolescents and its relationship with emotional difficulties, there has been limited research. Flavell et al. (1995, 2000) found metacognitive processes in children as young as five years of age, and reported that the capacity for introspection increases with age, and that children’s ability to hold metacognitive beliefs about anxiety may begin around ages seven to eight. Further research found that children and adolescents report metacognitive beliefs that are correlated with emotional distress (Cartwright-Hatton et al., 2004; Smith and Hudson, 2013). White and Hudson (2015), in a sample of 187 children, aged 7-12 years, reported that metacognitive beliefs were associated with levels of GAD, OCD, panic attacks, separation anxiety, and social anxiety. Research into 7-17 year olds \((n = 98)\) demonstrated that metacognitive beliefs were also present in this age group and were associated with anxiety and depression symptoms, and levels of excessive worry (Bacow et al., 2009).

Only a small number of studies have explored the role of worry and rumination in children with ASD, demonstrating that such repetitive and perseverative thinking is associated with anxiety and depression (Mazefsky et al., 2014; Rieffe et al., 2014). However, there appears to have been no published studies of the role of metacognitive beliefs in high functioning children with ASD.

**Metacognitive beliefs in children with ASD**

The role of metacognitive beliefs and processes in anxiety experienced by ASD populations currently remains unclear due to a lack of investigative research. Only one study to date appears to have specifically evaluated metacognitive beliefs in ASD (Grainger et al., 2014). This study \((n = 18)\) found that high functioning adults with ASD endorsed higher levels of metacognitive beliefs about monitoring their own thoughts than matched adults without ASD; however, the study did not explore metacognitive beliefs in regard to emotional distress.

Considering the high prevalence of anxiety, including worry, and depression in ASD, it may be theorised that children with ASD have developed metacognitive beliefs in line with the findings in individuals who are not on the autism spectrum. Thus, the aim of the current research was to explore the presence of metacognitive beliefs in high functioning children with ASD and the associations of such beliefs with emotional distress. It was hypothesised that there would be a positive correlation between anxiety and metacognitive beliefs in children with ASD. Primarily, that elevated metacognitive beliefs would be associated with increased levels of anxiety symptoms. It was further hypothesised that metacognitive beliefs in this sample would be associated with depressive symptoms.

**Method**

**Sample**

Following ethical approval from the University of the Sunshine Coast (USC), Australia Human Research Ethics Committee, participants were recruited through the University of the Sunshine Coast (USC), Australia ASD Support Group, radio, and local newspapers. Consistent with research determining the development and presence of metacognitive abilities in children (Flavell et al., 1995, 2000), the age of eight years was defined as the minimum age criteria for the
current study. Because the focus of this study was children, the upper age limit of 12 years was established due to the onset of adolescence at 13 years. In total, 35 individuals expressed interest in participating in the study. Of these participants, 12 did not meet the inclusion criteria: that is, they did not have an ASD diagnosis from a psychiatrist or paediatrician, or were outside the age range, or had a comorbid diagnosis of oppositional defiant disorder or an intellectual disability. Participants were not excluded if they had comorbid diagnoses such as attention deficit hyperactivity disorder \( (n = 6) \), a diagnosed anxiety disorder \( (n = 1) \), Tourette’s syndrome \( (n = 1) \), or a hearing impairment \( (n = 1) \). Consequently, 23 participants \( (17 \text{ male and 6 female}) \) aged between 8 and 12 years \( (M = 10.38 \text{ years}, SD = 1.39) \) voluntarily participated in the study. All participants had a confirmed diagnosis of ASD from a paediatrician or child psychiatrist. The process of confirming this included review of current diagnostic information (process of obtaining diagnosis, e.g. paediatrician/psychiatrist name) and confirmation of the State of Queensland (Department of Education and Training) verification status of the participant (name of school and relevant support information) from parent/guardian and review of the individual case to ascertain if further diagnostic clarification was required. All diagnoses were made prior to the release of DSM-5 and as such included: Asperger syndrome or pervasive developmental disorder – not otherwise specified (PDD-NOS). High functioning was determined by one of the researchers, an experienced clinical psychologist, with expertise in diagnosing ASD. High functioning autism is the terminology used when referring to an individual on the spectrum who is deemed to be functioning at a higher cognitive level (in terms of a cognitive capacity (IQ) greater than 70; Carpenter \textit{et al.}, 2009; Sanders, 2009). An individual who is considered to be High Functioning, in the context of this study, is the one who is, in cases, able to function with consistent adjustment plans, in a mainstream educational setting, without the requirement of further adjustments.

All children included in the study were currently attending mainstream schooling and were State of Queensland verified (requiring sign off of their diagnosis as being on the spectrum under the categories of pervasive developmental disorder – not otherwise specified or Aspergers syndrome (DSM-IV-TR) under their categories entitling them to an Educational Adjustment Programme) as signed off by a paediatrician or psychiatrist.

**Measures**

Demographic information was gathered prior to the commencement of the study. The study questionnaires were completed on an iPad using Survey Monkey® and included: the Metacognitions Questionnaire for Children (MCQ-C30; Esbjørn \textit{et al.}, 2013) and the Revised Children’s Scale of Anxiety and Depression (RCADS; Chorpita \textit{et al.}, 2000).

**Metacognitions Questionnaire for Children (MCQ).** The MCQ-C30 (Esbjørn \textit{et al.}, 2013) is a 30-item self-report questionnaire used to measure metacognitions in youth aged 7-17 years. It was adapted for children from the MCQ (Cartwright-Hatton and Wells, 1997) and Metacognitions Questionnaire for Adolescents (Cartwright-Hatton \textit{et al.}, 2004). Scores are measured on a four-point Likert scale \( (1 = \text{not at all}, 2 = \text{a little}, 3 = \text{very}, 4 = \text{totally}) \), and there are five subscales. The positive metacognitions subscale (POS) measures the belief that worrying may be helpful in preventing or avoiding problems in the future (e.g. “if I worry now, then I will have fewer problems later”). The negative metacognitions subscale (NEG) measures beliefs that worry is harmful and uncontrollable (e.g. “worrying is bad for me”). The cognitive confidence subscale (CC) evaluates the confidence a person has in their memory and attention (e.g. “I think I am bad at remembering names”). The need for control subscale (NC) measures beliefs about needing to control one’s thoughts and being responsible for the negative consequences of not doing so (e.g. “If I cannot control a worry, and it comes true, then it is my fault”). Lastly, the cognitive self-consciousness subscale (CSC) assesses the extent to which a person is aware of, and focusses on, their thinking (e.g. “I think a lot about my thoughts”). The scores for each subscale are summed to provide the overall MCQ-C30 total score, where higher sum scores of the total score and each of the five subscales indicate a greater number and strength of metacognitions (Esbjørn \textit{et al.}, 2013).

Esbjørn \textit{et al.} (2013) found that the Cronbach’s \( \alpha \) coefficient for the total scale was good \( (\alpha = 0.87) \). In addition, the internal consistency of the total scale score and four of the five subscales scores was good with \( \alpha \)s ranging from 0.75 to 0.87. Some concerns were raised...
with the NC subscale which was found to exhibit an internal consistency of $\alpha = 0.60$ (Esbjørn et al., 2013). It should be noted that consistency and reliability may be impeded by the child’s metacognitive ability and understanding. Research has suggested that although children display metacognitive understanding from five years old, their understanding develops throughout middle and later childhood (Bolton, 2004). Consequently, metacognitive understanding varies with age and should be considered when interpreting this measure.

RCADS. The RCADS (Chorpita et al., 2000) is a 47-item self-report questionnaire used to measure symptoms corresponding to selected DSM-IV anxiety and major depressive disorders in children aged 6-18 years. Answers are recorded on a four-point Likert scale ($0 = \text{never}$, $1 = \text{sometimes}$, $2 = \text{often}$, $3 = \text{always}$). There are eight scales, including generalised anxiety (GAD), separation anxiety (SA), obsessive-compulsive (OCD), social phobia (SP), panic disorder (PD), major depression (MDD), total anxiety (TOTA), and total anxiety and depression (TOTAD).

Psychometric examination has revealed that the RCADS scales have good internal consistency and reliability in clinical and Australian samples: GAD$\alpha = 0.84$, MDD$\alpha = 0.87$, SAD$\alpha = 0.78$, OCD$\alpha = 0.82$, SOC$\alpha = 0.87$ and PD$\alpha = 0.88$ (Chorpita et al., 2005; de Ross et al., 2002). The RCADS has recently received support, including from a systematic review, for clinical and research use in young people with ASD (Kaat and Lecavalier, 2015; Sterling et al., 2015; Wigham and McConachie, 2014).

**Procedure**

Potential participants’ guardians responded by phone or e-mail to local advertisements for children with ASD to participate in research on anxiety. Once the contact was made, a senior clinical psychologist (author two) invited the participants to attend a research appointment at the university’s clinical psychology clinic. Both the guardian and the child attended this appointment. At that time, the details of the research project were explained to all parties; separate and developmentally appropriate child and adult research project information sheets and consent forms were provided to the participants and were further verbally explained by the researcher. Consent was provided by both the child and the parent individually, ensuring both the child and parent understood that they could withdraw from the research at any time. The senior clinical psychologist then verified the participant had a formal diagnosis of ASD from a paediatrician or child psychiatrist. Following this, the self-report measures were completed by the child using Survey Monkey® on an iPad™. Participants completed the MCQ-C30 first, followed by the RCADS; the completion of these questionnaires took approximately 20 minutes. The data were collected in an appropriately lit, well-ventilated therapy room at the university’s clinical psychology clinic at a time when the clinic was not heavily attended in order to reduce potential anxiety for the child. All ethical requirements granted by the university’s Human Research Ethics Committee were adhered to.

**Statistical analysis**

As this was an exploratory study, with a limited sample size, a cross-sectional correlation design was utilised. We used IBM SPSS Statistics version 21 to compute descriptive statistics, internal consistency of scales, and correlation analyses. Preliminary analysis screened the data for outliers and missing values. Missing scores were established as random occurrences and replaced by substituting the mean score of that item (Somasundaram and Nedunchezhian, 2012). To explore the relationship between metacognitive beliefs and anxiety and depression conditions, a series of Pearson’s correlation coefficients ($r$) were calculated. The analysis included all RCADS and MCQ-C30 subscales and total scores for both measures.

In our data, the Cronbach’s $\alpha$ coefficients for the MCQ-C30 ranged from 0.65 to 0.87 across the subscales except for the CSC subscale that had an internal consistency of $\alpha = 0.62$. In relation to the RCADS, the Cronbach’s $\alpha$ coefficients ranged from 0.84 to 0.92, except for the obsessive-compulsive (OCD) subscale that produced 0.63.

**Results**

Regarding our hypotheses that there would be positive correlations between anxiety, depression, and metacognitive beliefs in children with ASD the following was found. Correlational analysis identified
seven significant, medium, positive correlations and 20 significant, large, positive correlations (see Table I). On the MCQ-C30 subscales, negative metacognitions (NEG), need for control (NC), and total MCQ-C30 (TOT) were correlated with the full range of RCADS anxiety subscales. Further, positive metacognitions (POS) were significantly correlated with the RCADS generalised anxiety (GAD) and obsessive-compulsive (OCD) subscales; interestingly, it was the only MCQ-C30 subscale significantly related to the major depression (MDD) subscale. The less frequently correlated subscales of the MCQ-C30 in relation to the RCADS in descending order included the positive metacognitions (POS), cognitive confidence (CC), and cognitive self-consciousness (CSC). Furthermore, from the RCADS subscales, it was evident that separation anxiety (SA), generalised anxiety (GAD), and obsessive-compulsive (OCD) were more frequently correlated with MCQ-C30 subscales than the remaining RCADS subscales. These trends suggest that metacognitive beliefs related to worrying, as measured by the MCQ-C30, are associated with anxiety and, less so, with depression in high functioning children with ASD. Further, specific types of metacognitive beliefs, negative metacognitions (NEG) and need for control (NC), were more prevalent than others in this ASD sample.

Discussion
The aim of the current research was to explore metacognitive beliefs in high functioning children with ASD and the relationship with anxiety and depression symptoms. It was predicted that a positive correlation between metacognitive beliefs and anxiety and depressive symptoms would be evident. The current study is one of the first to demonstrate the presence of metacognitive beliefs, associated with anxiety and depressive symptoms, in high functioning children with ASD.

Consistent with other research into metacognitive beliefs in children and adolescents (Bacow et al., 2009; Benedetto et al., 2013), a positive correlation was found between anxiety and metacognitive beliefs. Further, a positive correlation was found between the metacognitive subscales negative metacognitions (NEG), need for control (NC), and the total MCQ-C30 and all the anxiety subscales on the RCADS. These findings provide further support that young people with ASD experience metacognitive processes and monitor their own thoughts, consistent with previous studies (Semrud-Clikeman et al., 2010; Wojcik et al., 2014). Overall, a strong prevalence for negative metacognitions (NEG) and need for control was found, which was coherent with the findings of the research of Smith and Hudson (2013) into the occurrence of metacognitive beliefs in children and preadolescents without ASD.

The large positive correlation found between total anxiety (TOTA, RCADS), total anxiety and depression (TOTAD, RCADS), and the total MCQ-C30 provides further support for the presence of metacognitive beliefs in the current sample and their relationship with symptomology found in the comorbid anxiety and depression conditions. However, when reviewed at a subscale level, it was observed that the major depression (MDD) subscale yielded a moderate significant

<table>
<thead>
<tr>
<th>Table I</th>
<th>Pearson R correlation coefficients MCQ-C30 subscales and the RCADS subscales</th>
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<tbody>
<tr>
<td>Metacognitions Questionnaire for Children: MCQ-C30</td>
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<tr>
<td>Metacognitions Questionnaire for Children: MCQ-C30</td>
<td></td>
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<tr>
<td>POS</td>
<td>NEG</td>
</tr>
<tr>
<td>Revised Children’s Scale of Anxiety and Depression: RCADS</td>
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<tr>
<td>SP</td>
<td>0.301</td>
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<tr>
<td>PD</td>
<td>0.168</td>
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<tr>
<td>MDD</td>
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</tr>
<tr>
<td>SA</td>
<td>0.274</td>
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<tr>
<td>GAD</td>
<td>0.507**</td>
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<tr>
<td>OCD</td>
<td>0.467**</td>
</tr>
<tr>
<td>TOTA</td>
<td>0.356</td>
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<tr>
<td>TOTAD</td>
<td>0.386</td>
</tr>
</tbody>
</table>

Notes: n = 23. MCQ-C30: positive metacognitions (POS), negative metacognitions (NEG), cognitive confidence (CC), need for control (NC); cognitive self-consciousness (CSC); RCADS: generalised anxiety (GAD), separation anxiety (SA), obsessive-compulsive (OCD), social phobia (SP), panic disorder (PD), major depression (MDD), total anxiety (TOTA), and total anxiety and depression (TOTAD). **Medium strength effect size; **large strength effect size. *Significant at the 0.05 and 0.01 levels (two-tailed)
correlation with only the positive metacognitions (POS) and total MCQ-C30. It was interpreted that this is a result of the emphasis on the measurement of worry-related metacognitions in the MCQ-C30 (consistent with the MCQ-30), a core mental activity within anxiety-related disorders, compared to the mental activity of rumination as found more commonly in depressive related disorders (Papageorgiou, 2006). As such, future research into metacognitive beliefs in children with ASD would likely yield findings of a stronger relationship between metacognitive factors and depressive mood if a measure of metacognitive beliefs about rumination was utilised. Importantly, our findings contribute to the empirical need identified by Damiano and colleagues (2014), for research on the mechanisms that maintain anxiety in children with ASD; that is, signifying that metacognitive beliefs are associated with anxiety and depressive symptoms in those with ASD. An evaluation of the anxiety subscales in the RCADS revealed that generalised anxiety (GAD) was the only subscale that yielded a significant positive correlation with positive metacognitions. These findings provide preliminary support that the role of positive metacognitive beliefs may in fact be more prominent in GAD than other forms of anxiety, further supporting theories developed by Wells (2005). The finding that positive (POS) and negative metacognitions (NEG), and the need for control (NC) metacognitions were correlated with GAD was consistent with Wells’ (1995) metacognitive theories of GAD and psychological disorder (Wells and Matthews, 1996). The finding that positive metacognitions (POS) was correlated with the generalised anxiety (GAD) subscale and lesser so to obsessive-compulsive (OCD) subscale was consistent with White and Hudson (2015), who noted that positive metacognitions in their sample of children without ASD was related to higher levels of GAD and OCD symptoms. The need for control (NEC) subscale was found to be positively correlated to all the anxiety conditions. This finding was unsurprising when considering that ASD is often characterised by inflexible and rigid behaviour and thinking (American Psychiatric Association, 2013). Research has found evidence of responsibility of thoughts in children and adolescents with GAD (Bacow et al., 2009). However, studies relating to the role of need for control of thoughts specifically in ASD are scarce. Nonetheless, research on the need for routine in children with ASD suggests that disturbances to routine outside of the child’s control is associated with more externalising behaviour problems and can cause significant distress (Henderson et al., 2011). This suggests that children with ASD may experience a need to control their internal cognitive events also, and that failure to do this results in anxiety, which would be consistent with our results regarding metacognitive beliefs about the need for control.

Interestingly, the CSC subscale, which evaluates beliefs related to the tendency to be aware of, and to focus on, one’s thoughts was only significant in respect to the obsessive-compulsive (OCD) subscale. This was consistent with previous research that examined adults with anxiety and demonstrated that individuals with OCD tended to score higher on the CSC subscale in comparison to other anxiety conditions and nonclinical controls (Cartwright-Hatton and Wells, 1997; Janeck et al., 2003). This suggests that in high functioning children with ASD, those with OCD symptoms, likely direct their attention to their thoughts excessively. Thus, as Janeck et al. (2003) asserted, such hyperawareness of thoughts may promote importance being placed on such thoughts, for example, thought-action fusion.

Limitations and future directions
Regarding limitations of the current research, the findings in this study need to be interpreted with caution, given that individuals with ASD may have reduced capacity to use the meta-representational concepts that are required to organise their introspections (i.e. the capacity to have self-understanding of their own thoughts and feelings; Frith and Happé, 1999; Grainger et al., 2014; Schriber et al., 2014). Further to this, research has found that individuals with ASD often have alexithymia – “having no words for emotions” (Ben Shalom et al., 2006; Berthoz and Hill, 2005), thus limiting their ability to accurately attend to the emotional content that is intertwined with their cognitive processes. Additionally, the study was cross-sectional in nature and the recruitment method of convenience sampling was required due to the rural location; consequently, only a small sample of participants was achieved, and they were generally comprised of Australian, Caucasian individuals from middle-class socioeconomic backgrounds. Therefore, the results of this study must be viewed as preliminary and may not necessarily be generalisable to individuals with ASD of other backgrounds or ages. Lastly, a comparison group of children without ASD was not utilised and,
therefore, differences regarding metacognitive beliefs for children with and without ASD cannot be established from the current findings.

Based on the present findings, more evidence is required to gain a full understanding of the metacognitive processes in children with ASD; including utilising a larger sample with the addition of a comparison group of peers without ASD, as well as longitudinal studies. Areas of research could include establishing the accuracy of self-reported metacognitions and the associated validity of psychometric measures for metacognitions in ASD populations. This could give greater insight into the role of metacognition in anxiety disorders, mood disorders, coping strategies, and adjustment in those with ASD and could inform treatment modality.

Clinical implications

The current findings that psychological distress in our sample was positively correlated with metacognitive beliefs were consistent with Wells’ metacognitive conceptualisation of psychological disorder, the S-REF model (Wells and Matthews, 1996). Given that anxiety disorders have been found to endure over time (Esbjörn et al., 2015) and are frequently comorbid in children with ASD (van Steensel et al., 2011; White et al., 2009), our findings on metacognitive beliefs provide initial insight into potential maintaining factors that could be identified during clinical assessment and may be responsive to appropriate treatment. If future research establishes the present results in larger, more demographically diverse populations of children with ASD, this would further support the theoretical applicability of the metacognitive model of emotional disorder (the S-REF model) for these individuals. Consequently, indicating the possible utility of metacognitive therapy for these individuals; however, such an intervention would need rigorous, clinical evaluation of its effectiveness.

Conclusions

This research explored the role of metacognitive beliefs in ASD. Metacognitive beliefs were found to be prevalent in this population, particularly associated with anxiety-related conditions such as GAD, OCD, social phobia and separation anxiety. Specific metacognitions endorsed with these anxiety symptoms included negative and the need for control metacognitive beliefs. Despite methodological limitations, the results of this study provide direction for future research, initial evidence for metacognitive beliefs in ASD, and revealed that different metacognitive beliefs were associated with various psychological symptoms. However, more research in this area is necessary to substantiate and elaborate the current research. This will ideally enhance knowledge in the area of ASD in terms of the processes and interventions that may be useful in reducing symptoms and improving the quality of life.

References


**Further reading**


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