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Title: Conceptualising paranoia in ASD: A systematic review and development of a theoretical framework

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Abstract: Paranoia, unfounded ideation that others deliberately intend harm, has predominately been studied in schizophrenia. Increasingly, it is recognised that there is a spectrum of severity of excessive mistrust across the general population. Relatively little is known about paranoia in individuals with Autism Spectrum Disorders (ASD), but rates could be expected to be higher given both difficulties in understanding others' mental states and frequent experiences of negative social interactions. A systematic search of English-language peer-reviewed publications was undertaken to synthesise empirical research about paranoia in ASD. Seven studies, comprising a total of 180 ASD participants, met the inclusion criteria. All the studies were cross-sectional, thereby limiting causal interpretations. Individuals with ASD were consistently found to have higher levels of paranoia compared to non-clinical controls, and lower levels than individuals with current psychotic experiences manifesting in the context of schizophrenia. Furthermore, the initial evidence indicates that paranoia in ASD may be linked with theory of mind performance, negative affect, and jumping to conclusions, but not to attributional style. As in typically-developing populations, causal and maintaining mechanisms for paranoia in ASD, against a background of genetic and environmental risk, most likely include cognitive and affective processes interacting with social factors. We hypothesise, however, that core ASD characteristics and associated neurocognitive impairments also serve to precipitate and perpetuate paranoia. A framework to guide further investigation is outlined.

Conceptualising paranoia in ASD

Abstract

Paranoia, unfounded fears ideation that others deliberately intend harm, has predominately been studied in schizophrenia. Increasingly, it is recognised that there is a spectrum of severity of excessive mistrust across the general population. Relatively little is known about paranoia in individuals with Autism Spectrum Disorders (ASD), but rates could be expected to be higher given both difficulties in understanding others' mental states and frequent experiences of negative social interactions. A systematic search of English-language peer-reviewed publications was undertaken to synthesise empirical research about paranoia in ASD. Seven studies, comprising a total of 180 ASD participants, met the inclusion criteria. All the studies were cross-sectional, thereby limiting causal interpretations. Individuals with ASD were consistently found to have higher levels of paranoia compared to non-clinical controls, and lower levels than individuals with current psychotic experiences manifesting in the context of schizophrenia. Furthermore, the initial evidence indicates that paranoia in ASD may be linked with theory of mind performance, negative affect, and jumping to conclusions, but not to attributional style. As in typically-developing populations, causal and maintaining mechanisms for paranoia in ASD, against a background of genetic and environmental risk, most likely include cognitive and affective processes interacting with social factors. We hypothesise, however, that core ASD characteristics and associated neurocognitive impairments also serve to precipitate and perpetuate paranoia. A framework to guide further investigation is outlined.

1 Introduction

Autism spectrum disorders (ASD) are childhood onset, behaviourally defined, neurodevelopmental conditions. Core ASD characteristics include socio-communication impairments, adherence to non-functional rituals and routines, and engagement in a narrow repertoire of interests and activities (APA, 2014; WHO, 1992). ASD prevalence rates are reported to just exceed 1% of the population (Brugha et al., 2011), however given significant clinical heterogeneity, and potential sex differences in symptom profiles (Van Wijngaarden-Cremers et al., 2014), it is conceivable that rates are underestimated (NICE, 2011, 2012).

Although diagnostic overshadowing may mar a full assessment of psychopathology, studies consistently indicate that individuals with ASD are highly likely to experience psychiatric co-morbidities (Joshi et al., 2013; Russell et al., 2015; Simonoff et al., 2008),

including anxiety disorders (White, Oswald, Olendick & Scahill, 2009), obsessive compulsive disorder (OCD) (Cadman et al., 2015), low mood and depression (Ghaziuddin, Ghaziuddin & Greden, 2002), and psychosis (Chisholm, Lin, Abu-Akel & Wood, 2015). Similarly, transdiagnostic characteristics – i.e. symptoms which can manifest across a range of psychiatric disorders – occur commonly. Such characteristics can include problems with eating and 'food selectivity' (Marí-Bauset, Zazpe, Mari-Sanchis, Llopis-González & Morales-Suáres-Varella, 2014), sleep disturbance (Elrod & Hood, 2015), emotion dysregulation (Weiss, 2014), and paranoia (Blackshaw, Kinderman, Hare & Hatton, 2001).

Recent research indicates that paranoia (ideas of reference and persecution) exists on a spectrum of severity in the general population (Freeman et al., 2005), similar to the profile of common mental health problems such as anxiety and depression (Plomin, Haworth & David, 2009). Paranoia comprises ideas of reference and persecution. The defining type of cognition is persecutory ideation, unfounded ideas that others deliberately intend to harm the person. It is This is unsurprising that there is a paranoia spectrum in the general population: every day, people make decisions about whether to trust or mistrust, but accurately judging the intentions of others is difficult. Many people have a few paranoid thoughts; a few people have many. One of the clearest demonstrations of this is that a significant minority of the non-clinical general population can have paranoid thoughts about neutral computer characters in immersive virtual reality social situations (Freeman et al., 2008). Paranoia is associated with youth, poverty, isolation, stress, use of cannabis, and a range of mental health disorders (Freeman et al., 2011), although the direction of these associations is yet to be definitively established. It is also associated linked with a range of adverse life experiences (e.g. Bentall et al., 2012). Detailed Aanalysis of data from a nationally representative population (N=8580) indicated that there is a single paranoia dimension, with ideas of persecution building on ideas of reference, which build on mistrust and interpersonal sensitivity (Bebbington et al., 2013). This notion of paranoia building on common concerns about the self in regard to interpersonal relationships is captured in the conceptualisation of a paranoia hierarchy (see Figure 1) (Freeman et al., 2005). The severity of paranoia varies in content, degree to which concerns are believed, and levels of distress and impairment. At the severe end are persecutory delusions, commonly seen in people who have psychosis or schizophrenia. The presence of persecutory delusions in those diagnosed with non-affective psychosis has led to paranoia predominately being studied in this patient population. However, paranoia is also associated with a range of other mental health disorders including social phobia (Schutters et

al., 2012), post traumatic stress disorder (PTSD) (Freeman et al., 2013), depression (Wigman et al., 2012), and bipolar disorder (Goodwin & Jamison, 1990).

The contribution of genetic and environmental causes to the occurrence of paranoia has been examined in one large classical twin design study (Zavos et al., 2014), whereby paranoia was assessed dimensionally in five thousand adolescent twin pairs; it was found that the contribution of genes and environment was approximately equal. Several psychological mechanisms have been hypothesised to precipitate and perpetuate paranoia (Frith, 1992; Bentall, Corcoran, Howard, Blackwood & Kinderman, 2001; Freeman & Garety, 2014). Since almost by definition mental states are being misread in paranoia, theory of mind impairments have been proposed as a cause (Frith, 1992). Similarly, the explanation of events in terms of malevolent others has implicated a generalised externalising and personalising attributional style linked to unstable and negative self-views (Bentall, Corcoran, Howard, Blackwood & Kinderman, 2001). Another view is that at the centre of paranoia are ideas about threat, linked to anxiety and depression, negative self-beliefs (including interpersonal sensitivity), and adverse life experiences (Freeman & Garety, 2014). Reasoning biases, such as 'jumping to conclusions' (reduced data-gathering), have also been considered a contributory causal factor to the levels of conviction and failure to consider alternative explanations seen in paranoia (Garety et al., 2015).

Figure 1 about here.

Enhancing understanding of paranoia in ASD is important for several reasons. Our view is that it is theoretically plausible that this clinical population are at an increased risk of <u>developing</u> paranoid ideation. This increased risk may stem, in part, from the social consequences of having ASD. For example, individuals who have ASD desire friendships and intimate relationships, yet they are often rebuffed, and are prone to being bullied and victimised (Schroeder, Cappadocia, Bebko, Pepler & Weiss, 2014); experiences which can exacerbate social withdrawal, augment <u>concerns about otherssocial-evaluative concerns</u>, and encourage negative beliefs about the self and others (e.g. Gracie et al., 2007), all of which can <u>serve as risk factors for paranoia</u>. But it may also be that neurocognitive processes <u>commonly</u> <u>experienced by individualsassociated</u> with ASD enhance the likelihood of paranoia. For <u>example, dD</u>ifficulties in understanding others' intentions (Baron-Cohen, Wheelwright, Hill, Raste & Plumb, 2001a), a tendency for being detail-focused (commonly referred to as weak central coherence; Happé & Frith, 2006), and problems with cognitive flexibility (Wilson et

al., 2014) may render individuals with ASD vulnerable to information or interpretation biases, <u>thereby</u> encouraging the negative misinterpretation of others' actions, <u>which can also</u> serve as risk factors.

In summary, although paranoia may well co-occur with ASD, it is unclear whether rates and levels are comparable to those reported for typically-developing clinical and nonclinical populations. Also, it is uncertain whether there is a degree of ASD-specificity to paranoia, i.e. whether core ASD or associated characteristics serve to precipitate or maintain paranoid features. In the typically-developing population, there is increasing evidence to suggest that timely assessment and psychological interventions (specifically cognitive behaviour therapy (CBT)) can reduce paranoia, associated distress, and secondary symptoms (e.g. Freeman et al., 2015). Whether this is also the case for the ASD population warrants consideration. The aims of this review were threefold: (1) to synthesise empirical data about ASD and paranoia; (2) to propose a conceptual framework outlining mechanisms potentially contributing to the development and maintenance of paranoia; and (3) to highlight implications for clinical practice and research.

2 Method

2.1 Search strategy

Four databases were searched – Medline, PsycInfo, PubMed, and the Cochrane Central Register of Controlled Trials (CENTRAL) – from the date of inception until 19 April 2015. The search terms used were autis*- asperger*- pervasive development* disorder* AND paranoi*.

2.2 Study inclusion and exclusion criteria

We employed several search parameters, as follows: (1) primary observational, experimental or intervention studies; (2) published in peer-reviewed English language publications; and (3) focusing on paranoia (measured using clinical diagnostic criteria, or a subjective self-rating scale) in young people or adults with ASD (including all diagnostic sub-types). We allowed for the possibility that study participants may have additional co-morbidities and/or intellectual disability (ID). We excluded: (1) grey literature (including non-peer-reviewed and non-academic publications); and (2) studies that included participants diagnosed with ASD and psychosis (or schizophrenia), but where reports did not demarcate paranoid features from other positive symptoms of psychosis.

2.3 Analysis plan

A narrative approach to the analysis was undertaken, due to methodological heterogeneity.

3 **Results**

3.1 Search results and data extraction

The database searches were undertaken by JS; results were imported into EndNote version 7. See Figure 2 for an overview of the search results. An initial 228 studies were retrieved. Duplicates (n=103) were removed, and the titles of the remaining 125 papers were reviewed independently by JS and DS. Twenty-four papers were deemed to potentially meet the review inclusion criteria. Of these, 18 papers were excluded, either as these constituted reviews (Carpenter, 2007; Schneier, Blanco, Antia & Liebowitz, 2002), or as there were no paranoia data about individuals with ASD (Skokauskas & Gallagher, 2012), or because the studies did not report outcome measures pertaining to paranoia in individuals with ASD (Bara, Ciaramidaro, Walter & Adenzato, 2011; Collacott, Cooper & McGrother, 1992; Couture, Penn, Losh, Adolphs, Hurley & Piven, 2009; Gutkovich, Carlson & Carlson, 2007; Hallerbäck, Lugnegård & Gillberg, 2012; Konstantareas & Hewitt, 2001; Murphy, 2006; Pary, 1993; Pilowsky, Yirmiya, Arbelle & Mozes, 2000; Prado de Oliveira, 1999; Shastri, Alla & Sabaratnam, 2006; Siegel et al., 2014; Travé Rodriguez, Barreiro Marin, Galvez Borrero, del Olmo Romero-Nieva & Diaz Alvarez, 1994; Yan et al., 2000), or as paranoia outcome data about individuals with ASD were subsumed and subsequently analysed in a more recent paper (Pinkham, Hopfinger, Pelphrey, Piven & Penn, 2008). One further study was identified which was published during manuscript preparation, and sub-group data were obtained (Taylor et al., 2015; MT personal communication, July 2015). A total of seven studies, therefore, met the pre-specified inclusion criteria, which were read by all three authors. Data were extracted, summarised, and tabulated.

Figure 2 about here.

3.2 Overview of studies

Seven studies, all employing cross-sectional designs, were included in the review (Blackshaw, Kinderman, Hare & Hatton, 2001; Craig, Hatton, Craig & Bentall, 2004; Jänsch & Hare, 2014; Maras & Bowler, 2012; North, Russell & Gudjusson, 2008; Pinkham et al.,

2012; Taylor et al., 2015). See Table 1 for a summary of study details. All studies bar one (Pinkham et al., 2012), were undertaken in the UK.

Table 1 about here.

Each study described levels of paranoia in individuals diagnosed with ASD compared to control groups. The aims of studies differed somewhat: one study examined associations between autistic traits and psychotic experiences in adolescents (Taylor et al., 2015); three studies explored the link between theory of mind and paranoia (Blackshaw, Kinderman, Hare & Hatton, 2001; Craig, Hatton, Craig & Bentall, 2004; Jänsch & Hare, 2014); two studies investigated relationships between attributional style and paranoia (Blackshaw, Kinderman, Hare & Hatton, 2001; Craig, Hatton, Craig & Bentall, 2004); one study examined selfrepresentation and self-awareness in conjunction with paranoia (Blackshaw, Kinderman, Hare & Hatton, 2001); one study considered associations between probabilistic reasoning style and paranoia (Jänsch & Hare, 2014); one study assessed similarities and differences in paranoid ideation between clinical and non-clinical populations (Pinkham et al., 2012); three studies described levels of anxiety and depression, co-occurring with paranoia (Blackshaw, Kinderman, Hare & Hatton, 2001; Maras & Bowler, 2012; North, Russell & Gudjusson, 2008); and finally, two studies measured paranoia in the context of interrogative suggestibility, compliance and vulnerability (Maras and Bowler, 2012; North, Russell & Gudjusson, 2008).

3.3 Participant characteristics

A total of 180 individuals with ASD (152 males and 28 females) participated across studies, most of whom were diagnosed with Asperger's Syndrome. ASD diagnosis was primarily confirmed via clinical criteria. Additionally, three studies (Jänsch & Hare, 2014; Maras & Bowler, 2012; Taylor et al., 2015) asked participants to complete the Autism Quotient (AQ; Baron-Cohen, Wheelwright, Skinner, Martin & Clubley, 2001b), four studies undertook the Autism Diagnostic Observation Schedule (ADOS-g; Lord et al., 2000) with a proportion of participants (Maras & Bowler, 2012; North, Russell & Gudjusson, 2008; Pinkham et al., 2012; Taylor et al., 2015), and three studies (North, Russell & Gudjusson, 2008; Pinkham et al., 2012; Taylor et al., 2015) obtained data from the Autism Diagnostic Interview-revised (ADI-r; Lord, Rutter & Lecouteur, 1994).

IQ data were estimated from the National Adult Reading Scale (NARS; Nelson, 1982), the National Adult Reading Test (NART; Nelson, 1991), the Wechsler Abbreviated Scales of Intelligence (WASI; Wechsler, 1991), the Wechsler Adult Intelligence Scale (WAIS, Wechsler, 1999), or subscales from the Wechsler Intelligence Test for Children (WISC; Wechsler, 2004). At group level, mean full scale IQs fell within the average range.

There were limited data reported about other demographic characteristics (such as ethnicity), and clinical characteristics (such as psychiatric co-morbidity, or receipt of mental health services).

3.4 Quality assessment of included studies

We did not use a standardised checklist to formally assess methodological rigour of included studies. Nonetheless, several considerations are noteworthy when appraising the study methods and results. First, while ASD participants were recruited from clinical and non-clinical settings, response rates (i.e. the percentage of participants who agreed to take part versus those who declined) were not provided. It may be that some individuals were deterred from taking part, given the study remits, thereby skewing the representativeness of samples. Second, ASD was not necessarily assessed using 'gold-standard' objective instruments: as such, the severity and spread of symptoms, i.e. whether participants presented with more social or communication impairments were not documented. Hence, the degree to which ASD participants within each study and across studies represent a homogeneous (or heterogeneous) group is not known, with implications for the generalisability of study findings. Third, each study relied on a single self-report measure of paranoia, completed at one time point. Given that the utility of self-report questionnaires in ASD remains a contentious issue, study methods could have been enhanced through inclusion of a clinicianadministered measure, or a second self-report paranoia scale, i.e. to establish construct validity or inter-rater reliability. It is also the case that self-report measures are unable to determine if paranoid ideas endorsed, are actually unfounded. Additionally, psychological variables examined in studies, including theory of mind, attributional and reasoning styles, self-esteem, and affect, were also primarily measured using one form of assessment; some of which were self-report instruments, and as such their validity and reliability for the ASD population is uncertain. Confidence in study results could therefore have been augmented with the addition of an alexithymia measure to ascertain the extent to which participants were able to describe their internal states, as well as further tests of neuropsychological functioning (e.g. local versus global processing styles, cognitive flexibility, recall and memory). Fourth,

while appropriate choice of comparison group clearly depends on the key hypothesis tested, it is of note that each study included at least one comparison group (a clinical and/or nonclinical control group), but no study included an ASD control group, i.e. either individuals with ASD who presented with no paranoia, or a comparison between individuals who have low and high levels of paranoia. As there are hypothesised differences between ASD and typically-developing individuals – for example, in terms of genetics (Miles, 2011), neuroanatomy (Stanfield et al., 2008), and neuropsychological functioning (Wilson et al., 2014) – it is possible that direct comparison with other ASD samples, or individuals with developmental disorders, would prove useful. Similarly, not all studies matched participant groups on baseline variables, including age, IQ, and potential past or current mental health. Given that these factors may influence or confound performance on tests, the implication is that results and inferences drawn may be called into question.

3.5 Establishing rates of paranoia

Three self-report measures of paranoia were used: the Green et al. Paranoid Thoughts Scale (GPTS; Green et al., 2008) (Jänsch & Hare, 2014); the Paranoia Scale (PS; Fenigstein & Vanable, 1992) (Blackshaw, Kinderman, Hare & Hatton, 2001; Craig, Hatton, Craig & Bentall, 2004; Maras & Bowler, 2012; Pinkham et al., 2012), and the Specific Psychotic Experiences Questionnaire (SPEQ; Ronald et al., 2014) (Taylor et al., 2015). Each of the measures has previously been used with clinical and non-clinical populations, although only the GPTS was designed to be used for clinical purposes. While each questionnaire assesses thoughts and beliefs associated with, and indicative of paranoia, the PS focuses more specifically on mistrust, resentment and ideas of reference, having considerable overlap with anxious and depressive concerns, the GPTS investigates the conviction, preoccupation, and distress associated with ideas of reference and persecution (see Freeman (2008a) for a comprehensive overview), and the SPEQ examines a range of paranoid content but mainly milder more common ideas of reference and persecution in the general population. All of the measures are reported to have good psychometric properties, but none are specifically validated for the ASD population.

In each study, ASD participants – at a group level – endorsed higher levels of paranoia, compared with non-clinical controls (Blackshaw, Kinderman, Hare & Hatton, 2001; Jänsch & Hare, 2014; Maras & Bowler, 2012; North, Russell & Gudjusson, 2008; Pinkham et al., 2012; Taylor et al., 2015). Conversely, ASD participants self-reported lower levels of paranoia compared to individuals diagnosed with paranoid schizophrenia (Craig, Hatton,

Craig & Bentall, 2004; Pinkham et al., 2012), and higher levels compared with a schizophrenia non-paranoid group (Pinkham et al., 2012). Pinkham et al. (2012) also put forward evidence that paranoia in ASD included a higher degree of 'social cynicism' compared to patients with schizophrenia, which they considered as evidence of qualitatively different factors leading to paranoia in ASD.

3.6 Associations between autistic traits and paranoia

Taylor and colleagues (2015) examined relationships between autistic traits and paranoia in an adolescent sample, a proportion of which (n=32) were also diagnosed with ASD. Study findings indicated that associations between autistic traits (measured by the AQ; Baron-Cohen, Wheelwright, Skinner, Martin & Clubley, 2001b) and paranoia (measured by the SPEQ; Ronald et al., 2014) narrowly failed to reach significance (r = 0.09, p = 0.06).

3.7 Psychological variables associated with paranoia

3.7.1 Theory of mind

Three studies investigated theory of mind, either using the Reading the Mind in the Eyes Task (RMET; Baron-Cohen, Wheelwright, Hill, Raste & Plumb, 2001a) (Craig, Hatton, Craig & Bentall, 2004; Jänsch & Hare, 2014), the Hints Task (Corcoran, Mercer & Frith, 1995) (Craig, Hatton, Craig & Bentall, 2004), or the Projective Imagination Test (unpublished; Blackshaw, Kinderman, Hare & Hatton, 2001). Of these measures, the RMET has been most commonly used in ASD research, and has good psychometric properties (Baron-Cohen et al., 2015). In each study, theory of mind deficits were greater for individuals with ASD compared with non-clinical controls but comparable to participants with schizophrenia. Furthermore, Craig and colleagues (2004) found a negative correlation between scores on the PS (Fenigstein & Vanable, 1992) and theory of mind measures (RMET, r = -0.37, p = <0.01; Hints task, r = -0.25, p = <0.05), suggesting that neuropsychological functioning impairments may be implicated in paranoia in this group.

3.7.2 Attributional style

Attributional style was measured using the Internal, Personal and Situational Attributions Questionnaire (IPSAQ; Kinderman & Bentall, 1996) (Blackshaw, Kinderman, Hare & Hatton, 2001) and the Attributional Style Structured Interview (ASSI; unpublished; Craig, Hatton, Craig & Bentall, 2004). At group level – and in both studies – there were no significant differences between attributional styles of participants with ASD, and non-clinical

controls. Individuals with schizophrenia however, were more likely to 'externalise attributions for negative events', and display a global attributional style, compared with ASD participants (Craig, Hatton, Craig & Bentall, 2004). Blackshaw and colleagues (2001) also examined correlations between attributional style, paranoia and theory of mind, and found no significant relationships between these variables ('overall model' - F (6, 24) = 1.75, p = 0.152).

3.7.3 Self-representation and self-awareness

Self-awareness and self-representation were explored in one study via the Self-Discrepancies Questionnaire (SDQ; adapted from Higgins, 1986, 1987) and the Self-Consciousness Scale (SCS; Fenigstein, Scheier & Buss, 1975) (Blackshaw, Kinderman, Hare & Hatton, 2001). Given that individuals with ASD may experience innate difficulties in their ability to distinguish between self and others, and self-awareness, assessment of these constructs can prove complex but not insurmountable (see Hobson, 2010). Nonetheless, there were no significant differences in SDQ scores, between the ASD and non-clinical control groups. However, individuals with ASD displayed significantly higher levels of private self-consciousness, and comparable levels of public self-consciousness and social anxiety. Using regression analyses, it was also reported that private self-consciousness was a potential predictor variable (in this cross-sectional study) for paranoia (beta = 0.529, t = 3.42, p = 0.0019).

3.7.4 Reasoning style

The data-gathering 'jumping to conclusions' (JTC) reasoning style was assessed in one study (Jänsch & Hare, 2014). Using a computerised version of the 'beads task' (Garety et al., 2005), participants were asked to gather data in a reasoning task until they were sure in their judgement. There were significant differences, at group level, in the number of beads requested and rates of JTC bias. In both versions of the task, individuals with ASD made fewer requests for additional information: 50% of ASD participants displayed a JTC bias in version A, compared with 34% in version B. These rates are broadly comparable to those seen in studies of patients with delusions in the context of schizophrenia (Dudley et al, 2015).

3.7.5 Anxiety and depression

Self-ratings of anxiety and depression were obtained via the Hospital Anxiety and Depression Scale (HADS; Zigmond & Snaith, 1983) (Blackshaw, Kinderman, Hare & Hatton, 2001; North, Russell & Gudjusson, 2008), and the State-Trait Anxiety Inventory (STAI; Spielberger, Gorsuch, Lushene, Vagg, & Jacobs, 1983) (Maras & Bowler, 2012). In each of these studies, ASD participants endorsed higher levels of depression and anxiety, compared with non-clinical controls. Differences between groups however, were only found to be significant by North and colleagues (2008).

3.7.6 Fear of negative evaluation

In order to investigate social-evaluative concerns about others, The Brief Fear of Negative Evaluation scale (BFNE; Leary, 1983) was administered in two studies (Maras & Bowler, 2012; North, Russell & Gudjusson, 2008). The BFNE has good psychometric properties, albeit that the questionnaire involves reverse-item scoring, which feasibly can prove confusing (e.g. Weeks et al., 2005). In both studies, ASD participants endorsed more social evaluative concerns compared to non-clinical controls, albeit that differences between the groups were not significant. Potential associations between paranoia and FNE were not reported in detail.

3.7.7 Self-esteem

Self-esteem was measured by Maras and Bowler (2012), using the Rosenberg Self Esteem scale (RSE; Rosenberg, 1965); a measure commonly used with research and clinical populations, and which has good construct validity and a similar 'factor structure' across settings (e.g. Schmitt & Allik, 2005). At group level, there were no differences in mean scores: neither group scored within the low self-esteem range.

3.7.8 Suggestibility, vulnerability and compliance

Two studies investigated suggestibility and compliance in adults with ASD (Maras & Bowler, 2012; North, Russell & Gudjonsson, 2008), with the Gudjonsson Suggestibility Scale 2 (GSS-2; Gudjonsson, 1997), and the Gudjonsson Compliance Scale (GCS; Gudjonsson, 1989). There were no significant differences in scores, between groups, on the GSS-2 (Gudjonsson, 1997). Group differences were found on the GCS (Gudjonsson, 1989) in one study (North, Russell & Gudjonsson, 2008): ASD participants had higher scores compared to the non-clinical control group. Relationships between scores on the GCS, the GSS-2 and the PS were examined in one study (Maras and Bowler, 2012): significant associations were found between the GSS-2 and PS in the ASD group only (r = 0.52, p <

0.01), a finding which was accounted for by the fact that a proportion of the participants had scored particularly highly.

4 Discussion

4.1 Overview

Paranoia is increasingly recognised as a common experience in the general population, but relatively little is known about this phenomenon in ASD. To address this gap, we carried out the first systematic review on the topic. Seven cross-sectional studies met the pre-specified review inclusion criteria, highlighting the relative neglect of this topic in research. Across all studies, ASD participants had higher levels of paranoia compared with non-clinical controls. Furthermore, individuals with ASD had lower levels of paranoia compared with individuals with current psychotic experiences in the context of schizophrenia. It was also found that associations between attributional style and paranoia were not significant; theory of mind impairments were comparable for the ASD and schizophrenia groups, and more severe compared to non-clinical controls; ASD participants had higher levels of anxiety, depression, and social-evaluative concerns, compared to nonclinical samples (albeit that not all differences reached statistical significance); and there were indications that individuals with ASD had a jumping to conclusions reasoning style. The findings concerning correlates of paranoia are broadly similar to those found in the wider paranoia literature. That is, paranoia is associated with both negative affect and reasoning biases. Associations with externalising bias in patients with schizophrenia have been similarly hard to replicate (Garety & Freeman, 2013). What could be different in the results in the current review are the potential links with theory of mind impairments.

There are several factors which potentially limit the generalisability of study findings to the wider ASD population. A number of methodological limitations were noted, particularly in relation to small sample sizes, possible clinical heterogeneity of participants, and measurement bias. There was substantial reliance, across studies, on self-report measures. There were limited data available about the internal consistency of measures, which is an important aspect to consider, given that the validity and reliability of questionnaires is not known for samples with developmental disorders. Also, not all studies matched participant groups on baseline variables. Differences in IQ and mental health status, between groups, may have served to either over-inflate rates of paranoia, or may have affected the way in which clinical groups completed self-report questionnaires, e.g. leading to similarities in scores. Finally, non-parametric tests were used in some studies, due to small samples or skewed data; tests which may have limited power to detect significant differences, or associations between variables.

4.2 Conceptualising paranoia in ASD

If replicated in larger samples however, the high levels of paranoia in these ASD studies warrant explanation. A framework may aid such investigation. We hypothesise that causal and maintaining factors for paranoia in this clinical population share many similarities with typically-developing individuals, but that there is also likely to be an ASD-specificity which renders this group more vulnerable to developing mental health characteristics and paranoid features in particular (see Figure 3 for a schematic representation). First, we suggest that the combination of core ASD characteristics (in particular, difficulties with initiating and maintaining social interaction, deficits in the quality and quantity of conversational interchange, and preferences for routine), and aspects of neuropsychological functioning (specifically, weak central coherence, theory of mind impairments, cognitive inflexibility, and attentional problems), inform the means through which individuals with ASD think about themselves, others, and the world. We suggest that common cognitive styles include rumination (e.g. Crane, Goddard & Pring, 2011; Gotham, Bishop, Brunwasser & Lord, 2014), as well as a tendency for "all or nothing" thinking, over-generalising and catastrophising. We speculate that these factors shape the way in which individuals with ASD engage in, relate to, and make sense of the social world. For example, this group often have a literal interpretation of social norms and conventions, which can result in an inappropriately formal, incongruent, or awkward social style. Further, rather than being aware of social nuances and subtle variations between social situations - and hence modifying social behaviour accordingly individuals with ASD are more likely to adopt a formulaic approach, rather indiscriminately. In a bi-directional fashion, core ASD and associated characteristics affect the way in which others respond to them. For example, we note that social experiences for this clinical population can often seem, or be, aversive (e.g. being encouraged to engage in social situations such as group activities, needing to deal with unexpected social interactions, or being unable to undertake routinised behaviours), and/or adverse (e.g. being teased, victimised, or rejected). In either instance, social adversity is likely to have a bearing on beliefs, emotions, and behavioural responses. Over-generalisation of past experiences may encourage rumination, and enhance the propensity for catastrophising about future social situations. It is also plausible that these experiences contribute to negative core schema (for example, relating to themes of inferiority, worthlessness, a sense of difference, and hence vulnerability), which subsequently contribute to assumptions about others, and negative thoughts *in situ* as well as pre- and post- social interactions (for example, "why are they laughing at me?", "something might happen to me", and "people can't be trusted"). Additionally, we predict that these difficult experiences <u>situations</u> lead to, or exacerbate preexisting negative affect, such as anxiety and low mood, which are commonly experienced by individuals with ASD. Behavioural reactions potentially include an increase in social withdrawal, social isolation, avoidance, increased engagement in routines, and rumination. Although these are understandable short-term behavioural coping strategies, in the longer term they can prove to be unhelpful, as they reinforce anxiety, worry, and negative beliefs. We propose therefore that the combination of these predisposing factors, difficulties arising in the context of social interactions, and cognitive, behavioural, and affective responses, increase the potential for paranoid thoughts, general mistrust, and concerns about risk from others.

Figure 3 about here.

4.3 Limitations

This review has several limitations. First, we excluded non-English language publications. Second, because the remit of this review was symptom-focused rather than diagnosis-led, we excluded studies that investigated schizophreniform disorders (including schizophrenia, psychosis or bipolar affective disorder), or delusional beliefs in ASD, but where paranoia data were not reported, i.e. whereby total symptom scale information rather than sub-scale scores were provided. Third, we did not contact authors of studies that included typically-developing participants recruited due to high levels of paranoia, some of whom may conceivably have also met ASD diagnostic criteria.

4.4 Research implications

Five key areas warrant research. First, study findings suggest that levels of paranoia may well be higher in ASD compared with non-clinical controls. This, however, remains to be definitively demonstrated. Such future studies could aim to establish whether levels of paranoia differ according to ASD diagnosis or symptom profiles, and other characteristics such as IQ, sex, and age, and co-morbid mental health characteristics (e.g. depression, anxiety, or worry). Ideally, studies should incorporate objective measures of ASD – in order to facilitate investigation of childhood and current symptom profiles – and it would prove

useful for samples to comprise individuals who have comparable ASD presentations, i.e. representing a more homogenous group. Second, we suggest that a more comprehensive assessment of neuropsychological functioning processes, including theory of mind, memory, and attention, are needed, in order to better understand the extent to which drivers for paranoia in ASD are similar or distinct from typically-developing samples. Third, the ecological validity of self-report paranoia measures in ASD is uncertain: it may be that some social situations described do not apply or statements may be interpreted too literally (Maras & Bowler, 2012). Hence, future studies should include several methods of assessment, such as an informant-rating or clinician-administered scale, as well as different self-report measures, so as to be able to establish the reliability and validity of these instruments, and to enhance understanding of the paranoia symptom profile in ASD. Alternatively, virtual reality techniques have been used to assess paranoia in typically-developing populations in order to accurately assess unfounded ideation (Freeman, 2008b); such methods may also be suitable for individuals with ASD (Parsons & Mitchell, 2002). Also, given that similarities have been noted in neural substrates of individuals with ASD and schizophrenia (see Pinkham, Hopfinger, Pelphrey, Piven & Penn, 2008), (f)MRI studies may facilitate further understanding of the biological basis of disorders characterised by impairments in social cognition, and associated with paranoia. Fourth, causal and maintaining mechanisms for paranoia in ASD clearly require sustained research. Finally, given that paranoia may exacerbate psychopathology symptoms, such as affect and anxiety, and vice versa, future studies should investigate the potential dynamic interactions.

4.5 Clinical implications

Several implications for clinical practice are pertinent. It is easy to assume that social avoidance, and concerns about others' intentions and actions, are attributable to core ASD characteristics. We suggest, however, that clinicians should be mindful that individuals with ASD may well have paranoia i.e. excessive mistrust, potentially manifesting in the absence of psychosis or schizophrenia. It is therefore pragmatic to establish whether this is the case, so as to disentangle contributory factors that account for presenting difficulties. This is likely to involve a considered approach on behalf of clinicians, and is contingent on the development of rapport and therapeutic trust. Also, it may be that operationalisation of terminology, e.g. 'anxiety' or 'paranoia', would prove useful (Attwood, 2004). In terms of treatment, CBT is reported to be effective for reducing paranoid features and secondary symptoms (e.g. Freeman et al., 2015), although no studies have sought to specifically target paranoia in

individuals with ASD. We perceive that CBT interventions may well have clinical utility for concomitant paranoia in ASD, but that several adaptations may be needed, as is also the case when treating other psychiatric comorbidities (Lang et al., 2010; Spain et al., 2015). We suggest that the formulation used to guide treatment (at least in the short term) focuses on maintaining mechanisms rather than causal factors. Also, due to difficulties with introspection, and alexithymia (Bird and Cook, 2013), it may be easier to support individuals with ASD to start noticing links between situations, behavioural responses, and beliefs rather than focusing on the role of affect. That said, we would advocate that treating clinicians should consider the extent to which affect may precipitate or perpetuate paranoia. --While cognitive interventions are likely to be crucial, e.g. in order to address assumptions and beliefs about the self and others, it may be that an appropriate starting point is to offer psycho-education (e.g., about social relationships), social skills interventions (e.g., to enhance verbal and non-verbal communication skills), and behavioural techniques (e.g., to increase assertiveness skills, and to reduce social withdrawal). Further, it is quite possible may be that targeting other symptoms, e.g. anxiety or low mood may could also help to reduce paranoid features. Finally, as in treating all paranoia, suitably individualised direct behavioural tests are needed to help individuals relearn that they are safe.

4.6 Conclusion

Paranoia in ASD is an under-researched area, but potentially clinically important. The early indications are that levels of paranoia are raised in ASD. There are plausible grounds for expecting interactions between ASD core and associated characteristics, and paranoia, with each exacerbating the other. However, the mechanisms underlying these relationships are yet to receive systematic scrutiny. In this paper we have outlined the findings from the extant literature and proposed a framework which may help guide future investigations.

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Conceptualising paranoia in ASD

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Study	Aims & Design	Sample Characteristics	Outcome Measurements	Results
Blackshaw et al. (2001) UK	Aim To investigate aspects of the attributional model of paranoia in individuals with AS, compared to NCC Cross-sectional design	Recruitment SourceASD: charitable organisations, community settings, or postal surveyNCC: informal methodsASD 20 males & 5 femalesAge: mean 23 (s.d. 6.8) IQ: mean 93 (s.d. 11.5)NCC 7 males & 11 females Age: mean 31 (s.d. 7.9) IQ: mean 110 (s.d. 10.7)	Paranoia PS Theory of Mind PIT Attributional Style IPSAQ Self-representation SDQ SCS Anxiety/Depression HADS	 ASD participants had higher paranoia scores, compared to the NCC participants ToMdeficits were more evident in the ASD, compared to the NCC group There were no significant differences, between groups, in causal attributions: both groups used personal and situational external attributions; and they were equally as likely to attribute negative and positive events to themselves There were no significant differences, in self-context, between groups ASD participants had significantly higher levels of private self-consciousness; a variable found to be associated with paranoia.
Craig et al. (2004) UK	Aim To investigate relationships between theory of mind, attributional style, and paranoia, in individuals with ASD compared to SCZ and NCC Cross sectional design	Recruitment Source ASD: support groups SCZ: inpatient clinical setting NCC: not reported ASD 15 males & 2 female Age: mean 24 (s.d. 6.7) IQ: mean 105 (s.d. 7.1) SCZ 11 males & 5 females Age: mean 32 (s.d. 9.9) IQ: 105 (s.d. 8.4)	Paranoia PS Theory of Mind RMET Hints task Attributional Style ASSI	 Paranoia scores differed between groups: the SCZ participants had the highest scores; the ASD group had higher scores than the NCC group, but lower scores than participants with SCZ ToM deficits were comparable between ASD and SCZ groups, and greater than the NCC group ToM and paranoia were negatively correlated in the ASD group There were no significant differences, between groups, in attributional style

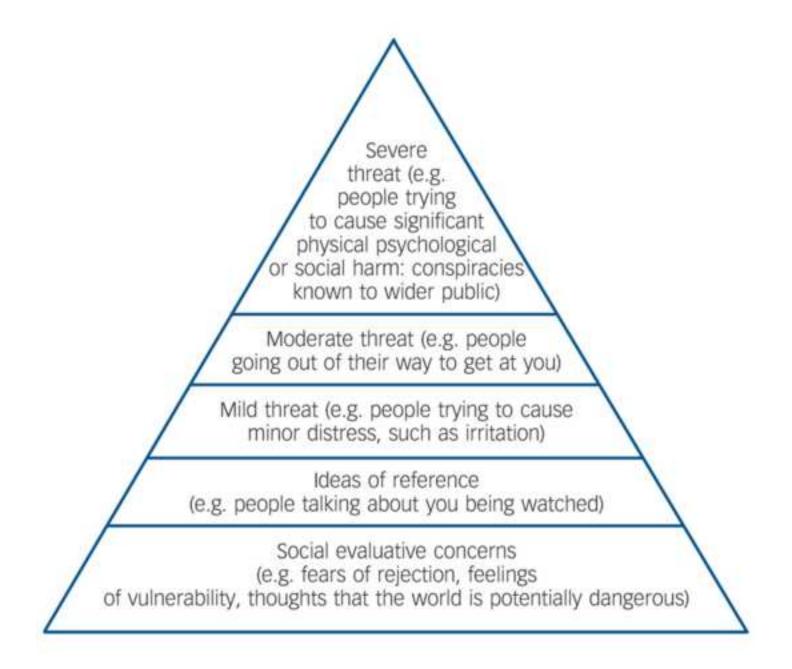
Table 1 Overview of included studies

(2008) UK	To investigate interrogative suggestibility and compliance in adults with ASD,compared to NCC Cross-sectional design	ASD: clinical & community sample NCC: existing research database ASD 21 males & 5 females Age: mean 35 (s.d. 11.1) IQ: not reported NCC 21 males & 6 females Age: mean 34 (s.d.11.9) IQ: not reported	PS Anxiety HADS FNE Suggestibility & Compliance GCS GCS- informant ratings GSS-2	 scores on the GCS compared to the control group; there were no significant differences, between groups, on the GSS-2 Levels of paranoia were higher in the ASD group There were no significant associations between paranoia, suggestibility and compliance, for either group Significant differences were found in self-reported anxiety and depression; ASD participants had higher levels of anxiety and depression Scores on the FNE differed significantly between groups: ASD participants endorsed higher levels of concern about negative evaluation
Pinkham et al. (2012) USA	Aim To investigatesimilarities and differences in paranoid ideation between ASD and clinical comparator groups (SCZ and SCZ-NP), and NCC Cross sectional design	Recruitment Source ASD: clinical settings & ongoing research projects SCZ: community settings & research projects NCC: not reported ASD 17 males & 1 females Age: mean 25 (s.d. 6.0) IQ: not reported SCZ 21 male & 3 females Age: mean 27 (s.d. 6.0) IQ: not reported SCZ-NP	Paranoia PS	 PS scores differed between the four groups: participants with ASD had higher paranoia scores than NCC and individuals with SCZ-NP, and lower scores than the SCZ participants Discriminant correspondence analysis suggested that the PS could be divided into three factors: 1 – paranoia; 2 – social cynicism; 3 – insightful acknowledgment Responses on the PS differed somewhat between the four groups. Participants with ASD had significantly higher levels of social cynicism compared to the clinical and NCC groups. Conversely, participants with schizophrenia (with and without paranoia) had significantly higher levels of insightful acknowledgmentthan the ASD group

Taylor et al.(2015) UK	<i>Aim</i> To investigate relationships between ASD traits, in typically-developing and ASD samples, and psychotic experiences occurring during adolescence Cross-sectional design	25 males & 5 females Age: mean 30 (s.d. 7.2) IQ: not reported <i>NCC</i> 22 males & 7 females Age: mean 29 (s.d. 5.4) IQ: not reported <i>Recruitment Source</i> ASD: epidemiological sample NCC: epidemiological sample ASD 29 males & 3 females Age: mean 16 (s.d. 0.68) IQ: not reported <i>NCC</i> 5,074participants Age: mean 16 (s.d. 0.68) IQ: not reported	Paranoia SPEQ Autistic traits CAST AQ	 Autistic traits were measured at four time points; psychotic experiences were assessed at one time point Correlations between autistic traits and paranoia were not significant, however associations approached significance for the ASD sample Some associations were found between negative symptoms and autistic traits
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ASD – Autism Spectrum Disorder; NCC –non-clinical control; AS – Asperger Syndrome; IQ – Intelligence Quotient; PS –Paranoia Scale; PIT – projected Imagination task; IPSAQ – Internal, Personal, and Situational Attributions Questionnaire; SDQ – Self DiscrepanciesQuestionnaire; SCS – Self Consciousness Scale; HAD – Hospital Anxiety Depression Scale; SCZ – schizophrenia; RMET – Reading the Mind in the Eyes Task; ASSI – Attributional Style Structured Interview; JTC – Jumping to Conclusions; GPTS – Green Paranoid Thought Scale; STAI – State-Trait Anxiety Inventory; FNE – Brief Fear of Negative Evaluation scale; RSE – Rosenberg Self Esteem scale; GCS – Gudjonsson Compliance Scale; GSS-2 Gudjonsson Suggestibility Scale; SPEQ – Specific Psychotic Experiences Questionnaire; CAST – Childhood Autism Spectrum Test







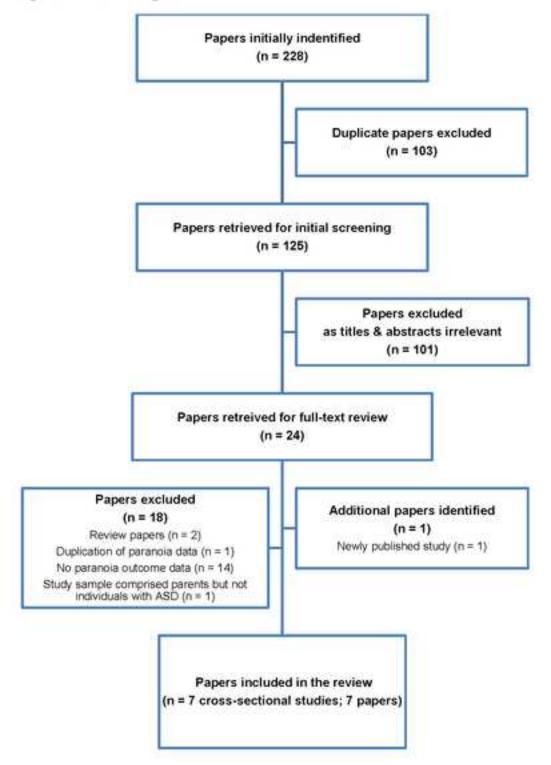
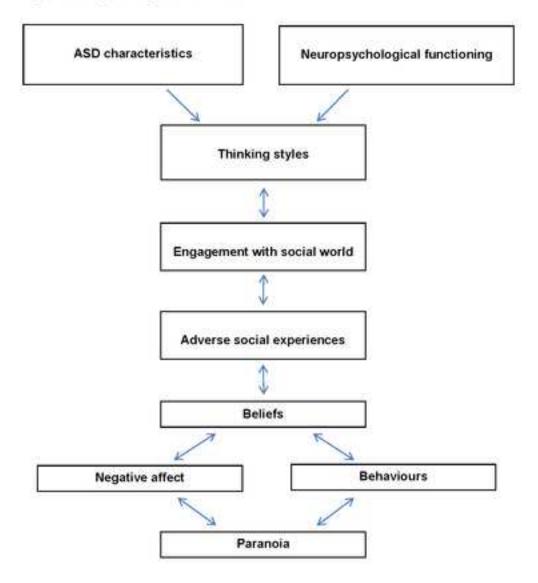


Figure 3 Conceptualising Paranoia in ASD



Highlights

- Psychosocial factors **may** render individuals with ASD vulnerable to developing paranoia
- Seven cross-sectional studies have examined paranoia in ASD
- Levels of paranoia are higher in ASD, compared to typically-developing samples
- ASD traits and neuropsychological functioning may precipitate and perpetuate paranoia
- A framework outlining causal and maintaining factors for paranoia in ASD is proposed