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Social anxiety in autism spectrum disorder: A systematic review

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ABSTRACT

Purpose: Social anxiety (SA) commonly co-occurs with autism spectrum disorders (ASD). It is conceivable that inherent socio-communication impairments, or their impact on social experiences, contribute to the development of SA.
Method: We undertook a systematic review to summarise English-language research about relationships between core ASD symptoms and SA in individuals with ASD.
Results: We searched five databases for studies published up until 28 July 2017. Of 1481 publications retrieved, 24 cross-sectional studies (described in 25 papers) met the inclusion criteria. Given methodological and clinical heterogeneity, data were synthesised narratively. SA, in individuals with ASD, was associated with poorer social skills and functioning, and reduced social motivation. There were associations between self-report SA and ASD measures, but a trend towards non-significant relationships between parent-ratings of these symptoms. Tentative evidence indicated that SA symptoms were not associated with restricted, repetitive behaviours or sensory sensitivities.
Conclusion: These findings support the notion that there are links between core ASD characteristics and SA. Further studies, employing qualitative and quantitative designs are needed to enhance understanding of causal, maintaining and protective mechanisms for SA in ASD.

Autism spectrum disorders (ASD) are common lifelong neurodevelopmental conditions, characterised by qualitative impairments in social communication and interaction, engagement in rituals and routines, and hypo- or hyper-sensory sensitivities (APA, 2013). It is widely accepted that many young people and adults with ASD experience anxiety. In part due to the heterogeneous profile, there is debate about whether anxiety is best conceptualised as being derived of, or co-morbid to, ASD (see Kerns & Kendall, 2012). In either instance, data from a range of epidemiological and clinical samples, employing a range of data collection methods, consistently indicate that individuals with ASD have high rates of anxiety disorders (see van Steensel & Heeman, 2017).

Social anxiety (SA), also known as social phobia, is especially common, with prevalence estimates reported to be as high as 50% (Bellini, 2004; Maddox & White, 2015; Spain et al., 2016); substantially higher than estimates of 7–13% cited for the non-ASD population (NICE, 2013a). Disparities in prevalence estimates across studies may be attributable to a number of reasons, including differences in sampling and selection criteria (e.g. epidemiological vs. clinical samples), methods of assessment (e.g. self- vs. clinician-rated measures, or use of one vs. multiple measures), diagnostic overshadowing (whereby co-morbid symptoms are wrongly

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attributed to ASD alone), or impairments in cognitive functioning (e.g. in introspection) which render it difficult for individuals with ASD to describe their internal states.

Hallmark characteristics of SA include autonomic symptoms of anxiety manifesting in specific or general social situations, a fear of negative evaluation or judgement by others, and avoidance of or escape from cues that evoke anxiety (APA, 2013; WHO, 1992). In non-ASD individuals, SA symptoms often emerge during adolescence with wide-ranging and long-term consequences. Causal and maintaining mechanisms for SA in neurotypical individuals are considered to be multi-faceted. These primarily comprise psychosocial and environmental factors, potentially underpinned by a genetic or biological predisposition (see Clark, 2001; Clauss & Blackford, 2012; Fox & Kalin, 2014; Rapee & Heimberg, 1997). Psychological frameworks for SA indicate that this may develop and be maintained by some or all of the following factors: an inhibited temperament; adverse social experiences during formative years; overestimation of the threat associated with social situations; negative beliefs about the self, others or the world; biases in information, attention and emotion processing; negative imagery; and 'safety behaviours' such as avoidance, mental rehearsal and post-event processing, which indirectly reinforce anxiety over time (Clark, 2001; Rapee & Heimberg, 1997).

It is possible that additional risk factors, specifically those relating to and arising from core ASD characteristics, contribute to the development of SA in individuals with ASD. Inherent socio-communication impairments may affect interactions and relationships in several ways. Social motivation, behavioural inhibition and volition to initiate overtures can influence the number, frequency and range of social situations individuals engage in. Further, the nature of responses to others, and degree of cooperativeness and turn-taking may influence the extent to which these are sustained. Social skills deficits may derail interactions with others. Stereotyped and idiosyncratic speech or preferences for discussing circumscribed interests may affect the fluidity of conversation. Repetitive behaviours, such as hand mannerisms or stereotyped body movements, may appear odd. Together, these characteristics can increase susceptibility to social adversity, e.g. rejection, teasing or bullying (Schroeder, Cappadocia, Bebko, & Weiss, 2014), and thereby contribute to social withdrawal and isolation. Moreover, difficult social interactions can give rise to negative ways of thinking, including paranoia and rumination (Spain, Sin, & Freeman, 2016), negative thoughts (e.g. about being the 'odd one out' or different), and, ultimately, core beliefs (schema) pertaining to inadequacy and inferiority.

Sensory sensitivities to light, sound or sensations (e.g. heat) may prove distracting or anxiety-provoking in social settings. Similarly, aversions to very specific sensory stimuli (Lord, Rutter & Le Couteur, 1994), may give rise to anticipatory anxiety about meeting familiar or unfamiliar others. Both sensory sensitivities and aversions may lead to avoidance. While avoidance may initially manifest in relation to specific settings, such as one particular supermarket, we have found in our clinical experience that this can become generalised, e.g. to all shops. Finally, a tendency for adhering to rituals and routines may hamper engagement in some social opportunities, or be remarked upon negatively by others, further contributing to misunderstandings and avoidance.

Bi-directionally, SA can encourage individuals with ASD to withdraw further from social interaction, thereby resulting in fewer occasions to observe social norms and conventions. As a consequence, these individuals may be less able to augment their social knowledge and social skills *in vivo*. Importantly, data from intervention studies tentatively indicate that SA may in fact partly moderate the success of social skills interventions. That is, individuals with ASD and SA may attain less favourable outcomes from such interventions due to the impact of these co-occurring anxiety symptoms (see Maddox, Miyazaki, & White, 2016; Pellecchia et al., 2016, Spain, Blainey, & Vaillancourt, 2017).

The aim of the present review is to systematically gather together, for the first time, the empirical data regarding relationships between ASD symptomatology and SA in individuals with ASD across the lifespan. This may elucidate more fully causal and maintaining mechanisms for SA with implications for prevention, early intervention and the development of more targeted treatments. Our review sought to answer the following question: What relationships are there, if any, between ASD and SA symptoms?

1. Method

1.1. Search strategy

We searched five databases – the Cochrane Central Register of Controlled Trials (CENTRAL), PsycInfo, Medline, PubMed, and Web of Science – for studies published until 28 July 2017. Search terms were autis* – Asperger* – development* disorder* AND social* anx* – social* phobi*. *A priori* inclusion criteria were: 1) English-language articles, published in peer-reviewed journals describing empirical quantitative research; 2) about SA or social phobia, and associations with core ASD symptoms in any of the domains outlined by either the ICD-10 (1992) or DSM-4/5 (1994, 2013); and 3) in children, adolescents or adults diagnosed with any subtype of ASD, with or without a concurrent intellectual disability (ID), and irrespective as to whether participants had had or were receiving treatment at the time of research participation. We excluded studies reporting the prevalence of SA, but which did not measure relationships between this and ASD, and those examining associations between anxiety and other variables, but where no SA subscale data were provided.

1.2. Study selection

Fig. 1 provides an overview of study selection. The database searches initially yielded 1481 reports. Duplicates (n = 166) were removed. Two authors (DS & JS) independently screened 1315 titles and abstracts. Of these, 81 articles were retrieved for full text review. Following discussion, 56 of these were excluded for the following reasons: not an ASD sample (n = 5), review paper (n = 3), treatment study (n = 3), study focused on general anxiety rather than SA specifically, and we could not extrapolate SA data (n = 24), and study examined aspects of SA in ASD, but did not focus on associations or relationships between these symptoms (n = 21). We

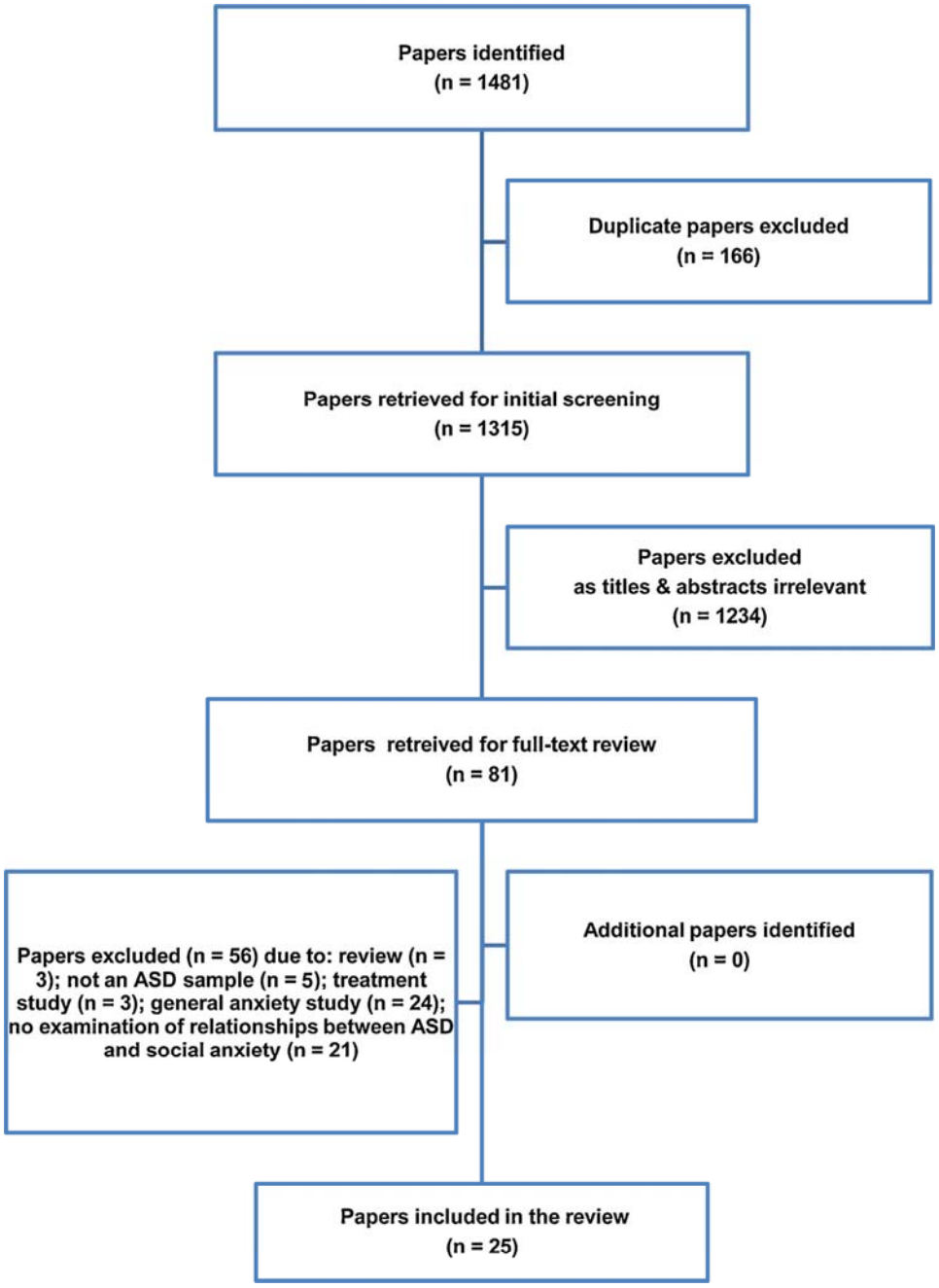

Fig. 1. PRISMA diagram.

also hand-searched the reference lists of the reviews and all papers included, and no additional papers were retrieved. Hence, the full sample was 25 papers. The list of excluded studies is available from the corresponding author.

1.3. Data extraction

We extracted and tabulated data about the study design; sampling frame; sample size; participant demographics in clinical and comparator groups; methods of ASD diagnosis; outcome measures employed; study results; and methodological considerations.

1.4. Analysis plan

While there was some overlap in outcome measures used, studies were methodologically heterogeneous (including different

designs and sample sizes) and clinically heterogeneous (including participants across the lifespan, with a range of core and co-morbid diagnoses). Data were therefore analysed using a narrative rather than meta-analytic approach.

1.5. Method of quality appraisal

We assessed study quality using the quality assessment tool for quantitative studies (Thomas, Ciliska, Dobbins, & Micucci, 2004). This method of quality assessment assesses nine aspects of empirical studies, as follows: 1) selection bias; 2) study design; 3) confounders; 4) blinding; 5) data collection methods; 6) withdrawals and drop-outs; 7) intervention integrity; 8) analyses; and 9) a global rating. Each aspect is assigned a rating of strong, moderate or weak. Following the suggestion by Thomas et al. (2004), we assigned a global rating of weak if two or more individual components were rated weak, moderate, if there was one weak and some moderate components, and strong, if there were no weak and at least two strong components. As per Butchart et al. (2017) we excluded the following study aspects: blinding, intervention integrity and analyses, as all studies included were cross-sectional, rather than interventional.

2. Results

In total, 24 studies (described in 25 papers) were included in this review (see Table 1) (Bejerot, Eriksson, & Mortberg, 2014; Bellini, 2004, 2006; Capriola, Maddox, & White, 2016; Cath, Ran, Smit, van Balkom, & Comijs, 2008; Chang, Quan, & Wood, 2012; Chen, Bundy, Cordier, Chien, & Einfeld, 2016; Corden, Chilvers, & Skuse 2008; Hallett et al., 2013; Kanai et al., 2011; Lever & Geurts, 2016; Maddox & White, 2015; Magiati et al., 2016; Meyer, Mundy, van Hecke, & Durosher, 2006; Orinstein et al., 2015; Perry, Levy-Gigi, Richter-Levin, & Shamay-Tsoory, 2015; Scharfstein, Beidel, Sims, & Rendon Finnell, 2011; Simonoff et al., 2008; South, Larson, White, Dana, & Crowley, 2011; Spain et al., 2016; Sukhodolsky et al., 2008; Swain, Scarpa, White, & Laugeson, 2015; Usher, Burrows, Schwarts, & Henderson, 2015; White & Roberson-Nay, 2009; White, Maddox, & Panneton, 2015).

2.1. Overview of included studies

Studies took place in the USA (n = 13), UK (n = 4), Netherlands (n = 2), Japan (n = 1), Australia and Taiwan (n = 1), Israel (n = 1), Sweden (n = 1) and Singapore (n = 1). All studies were cross-sectional. Ten studies compared two groups (ASD vs. clinical or non-clinical controls (NCC)), four compared three groups, and two included four groups. Thirteen studies recruited children and adolescents (aged 18 and under), six studies recruited adults, and five studies recruited across the age spectrum. A total of 1551 individuals with ASD took part, some of whom were recruited to more than one study. The majority of ASD participants were male. Where reported, most individuals were Caucasian.

2.2. Quality appraisal

See Table 2 for the quality assessment of included studies. Quality assessment was rated by two authors independently, and latterly discussed. Each study was assigned a rating of weak, moderate or strong for six aspects of the study design, as well as a global quality rating. We did not draw direct comparisons between studies and considered the merits of each separately.

In terms of potential selection bias, few studies described the total number of individuals in sampling frames, and the proportion of these who took part. Participants were recruited from a range of settings, including schools, higher education settings, inpatient and community clinical settings, previous research studies, or via adverts. Only two studies recruited epidemiological samples (Hallett et al., 2013; Simonoff et al., 2008).

In terms of study designs and confounding variables, it is noteworthy that all studies were cross-sectional. In studies which included two or more groups (n = 16), sample sizes were typically comparable. Several studies sought to match participants in terms of their baseline demographic characteristics, including sex and age. That said, other potentially influential factors, such as current or past treatment at the time of research participation, were not necessarily reported. Intelligence (IQ) was estimated in 14 studies (54%): four studies recruited participants with and without a concurrent ID (Hallett et al., 2013; Magiati et al., 2016; Simonoff et al., 2008; Sukhodolsky et al., 2008); participants in the remaining ten studies had an IQ in the average, or above average range.

Data collection methods varied. See Table 3 for an overview of ASD and SA measures utilised, general constructs assessed, the number of times each has been used and the method of rating. Diagnostic assessment of ASD was either undertaken during studies or a previous clinical assessment. Two studies (Bellini, 2004, 2006; Perry et al., 2015) used information obtained at clinical interviews. Seventeen studies confirmed diagnosis with ‘gold standard’ clinician-administered measures, specifically the Autism Diagnostic Interview (ADI-r; Lord et al., 1994) and/or the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2000). ASD screeners, including the Autism Quotient (AQ; Baron-Cohen et al., 2001), Social Responsiveness Scale (SRS; Constantino et al., 2003) and Social Communication Questionnaire (SCQ; Berument et al., 1999), were administered as standalone or adjunctive measures in 17 studies.

SA symptoms were primarily assessed with self- and/or parent-ratings on specific SA measures including the Liebowitz Social Anxiety Scale (LSAS; Liebowitz, 1987), Brief Fear of Negative Evaluation scale (BFNE; Leary, 1983), Social Anxiety Scale (SAS; La Greca and Stone, 2010) and the Multidimensional Anxiety Scale (MASC; March 1999). Relatively few studies (n = 8) included a standardised clinician-administered tool, such as the Anxiety Disorders Interview Schedule for DSM-IV (ADIS-IV; Silverman, Albano, & Barlow, 1996) or the Structured Clinical Interview for DSM disorders (SCID; First et al., 2002). Seven studies included one self- or informant-rated measure of SA, twelve studies included one clinician-rated assessment or multiple measures but no clinician-rated

Table 1
Summary of information for all studies included in the review.

Study: First author, Date, Location, Main focus, Theme addressed	Participants	Measures	Results
Bejerot et al. (2014) Sweden Investigation into prevalence and severity of SA Theme i	- *ASD* (n = 50) % male: 52 (n = 26); age: mean 30.0, sd 7.3, range 28-32; % higher ed.: 48 (n = 24); recruited via clinical services and a website - *SAD* (n = 100) % male: 37 (n = 37); age: mean 34.6, sd 9.1, range 33-36; % higher ed.: 43 (n = 43); recruited via adverts - *NCC* (n = 53) % male: 53 (n = 27); age: mean 32.3, sd 10.8, range 28-33; % higher ed.: 85 (n = 45); recruited via convenience sampling	- *ASD*:ADOS; HAGS; AQ - *SA*: SCID; LSAS	- Significant associations between the LSAS and AQ in the ASD group (LSAS total anxiety $r = 0.67$, $p < 0.001$; LSAS total avoidance $r = 0.56$, $p < 0.001$) - Significant differences in AQ scores of ASD participants: ASD + SAD > ASD-SAD ($p = 0.02$)
(2004) and Bellini (2006), USA Investigation into anxiety symptoms and associations between social skills and SA Theme iii, v	- *ASD* (n = 41) % male: 85 (n = 35); age: mean 14.2, range 12–18; FIQ: mean 100, sd 18.8; recruited via community ASD and education services	- *ASD*:No formal measure - *SA*: SSRS; SAS-A; MASC - *Behaviour*: BASC	- Significant negative associations between avoidance of, and distress about, specific or general social situations and social skills ($r > -0.031$, $p < 0.05$); and between performance worries and SA (all $r > -0.31$, all $p < 0.05$) - Associations between SA and social skills depended on skills under investigation: increased SA was associated with decreased assertiveness ($r = -0.31$, $p < 0.05$) - Curvilinear associations between empathy and SA: increased SA was associated with increased empathy scores (η from 0.43 to 0.63) - Non-significant associations between parent-ratings of social skills and self-reported SA - Predictor variables of SA were SSRS empathy, MASC physical symptoms, and SSRS assertion (all $B > -13.3$, all $p < 0.006$; model $R^2 = 0.34$, $p < 0.0005$)
Capriola et al. (2016), USA Examination of fear of negative evaluation Theme i, v	ASD (n = 44) - *ASD: teens* (n = 26) % male: 54 (n = 14); % ethnicity: Caucasian 89, African-American 4; age: mean 15.6, sd 1.6 - *ASD: adults* (n = 18) % male: 56 (n = 10); % ethnicity: Caucasian 89, Asian 6; age: mean 24.7, sd 7.3 NCC and CC (n = 69) - *NCC and CC: teens* (n = 20) % male: 55 (n = 11); % ethnicity: Caucasian 90, African-American 10; age: mean 14.6, sd 1.7 - *NCC and CC: adults* (n = 49) % male: 49 (n = 24); % ethnicity: Caucasian 80, Hispanic/Latino 6, African-American 4, Asian 8; age: mean 25.7, sd 7.1 Age range for all adolescents 12–17; age range for all adults 18–44; all recruited via research studies	- *ASD:* ADOS; SRS - *SA:* BFNE; MINI; ADIS	- Non-significant associations between, SRS and BFNE scores - Predictor variables for BFNE included social disability ($B = 0.55$, $p < 0.001$) and social motivation ($B = 0.56$, $p < 0.001$)
Cath et al. (2008), Netherlands Examination of phenomenology and symptoms of anxiety in clinical samples Theme i		- *ASD*: AQ - *SA*: LSAS; SCID	- Significant associations between the AQ total and subscale scores and the LSAS, excluding the attention to detail subscale ($p < 0.05$)

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Table 1 (*continued*)

Study: First author, Date, Location, Main focus, Theme addressed	Participants	Measures	Results
	- *ASD* (n = 12) % male: 83 (n = 10); age: mean 34.5. sd 10.5; % higher ed.: 100 (n = 12); recruited via clinical service - *SAD* (n = 12) % male: 83 (n = 10); age: mean 38.0, sd 11.0; % higher ed.: 100 (n = 12); recruited via clinical service - *OCD* (n = 12) % male: 83 (n = 10); age: mean 35.9, sd 11.9; % higher ed.: 100 (n = 12); recruited via clinical service - *NCC* (n = 12) % male: 83 (n = 10); age: mean 32.4, sd 11.3; % higher ed.: 100 (n = 12); recruited via a snowball method		
Chang et al. (2012), USA Examination of relationships between anxiety and social functioning Theme iii, iv, v	- *ASD* (n = 53) Age: mean 9.6, sd 1.7, range 7–11; recruited via clinical and education settings	- *ASD*: ADOS; ADI-R - *SA*: ADIS-C/P - *Social functioning*: SSRS	- Significantly poorer social functioning in participants with SA than those without ($p < 0.05$) - Social skills associated with SA severity included cooperation, assertiveness, responsibility, and self-control ($R^2 > 0.05$, all $p < 0.05$) - Significant associations between SA severity and social functioning ($r = -0.37$, $p < 0.01$): SA severity predicted poorer social functioning ($B = -0.39$, $p < 0.01$)
Chen et al. (2016), Australia and Taiwan Investigation into experiences and beliefs about everyday living Theme v	- *ASD Australia* (n = 14) % male: 29 (n = 4); % ethnicity: Caucasian 100; age: mean 24.8, sd 9, range 16–45; % higher ed.: 21% (n = 3); recruited via research adverts - *ASD Taiwan* (n = 16) % male: 75 (n = 12); age: mean 27.8, sd 6.3, range 16–45; % higher ed.: 75 (n = 13); recruited via clinical services	- *ASD*: SRS - *SA*: SIAS	- SA occurred more commonly when participants were with family or friends - Participants with less severe ASD were liable to feel more anxious in social situations; conversely, participants with more severe ASD seemed to experience greater interest and enjoyment in solitary or parallel activities
Corden et al. (2008), UK Examination of social-perceptual impairments, and relationships between SA, eye fixation, and emotion recognition Theme i	- *AS* (n = 21) % male: 76 (n = 16); age: mean 33.8, sd 13.6; FIQ: mean 118, sd 11.7; recruited via adverts and ASD support groups - *NCC* (n = 21) % male: 76 (n = 16); age: mean 32.1, sd 11.6; FIQ: mean 117, sd 8; recruited via adverts and ASD support groups	- *ASD*: ADOS; AQ - *SA*: SPAI; SDS	- Non-significant associations between ASD and SA
Hallett et al. (2013), UK Investigation into anxiety in clinical and non-clinical samples Theme ii	- *ASD* (n = 142) % male: 85 (n = 121); age: mean 13.5, sd 1.7; FIQ: mean 88, sd 22.3; epidemiological sample - *Co-twin* (n = 73) % male: 37 (n = 27); age: mean 13.5, sd 0.7; FIQ: mean 105, sd 13.2; epidemiological sample - *BAP* (n = 41) % male: 78 (n = 32); age: mean 13.4, sd 0.6; FIQ: mean 98, sd 17.2; epidemiological sample - *NCC* (n = 160) % male: 69 (n = 110); age: mean 12.8, sd 1.1; FIQ: mean 103, sd 15.2; epidemiological sample	- *ASD*: ADOS; ADI-R - *SA*: RCADF	- Non-significant relationships between self-rated SA and ADI-R scores - Significant negative associations between parent-rated SA and the social interaction domain of the ADI-R (ICC = -0.26, $p < 0.05$); and between parent-rated SA and the communication domain of the ADI-R (ICC = -0.22, $p < 0.05$)
Kanai et al. (2011), Japan Examination of anxiety, depression and personality Theme i	- *AS* (n = 64) % male: 78 (n = 50); age: median 32, range 19–50; JART: median 110, range 92–134; recruited via clinical setting - *NCC* (n = 65) % male: 80 (n = 52); age: median 32, range 19–57; JART: not reported; recruited via adverts	- *ASD*: AQ - *SA*: LSAS - *ASD*: ADOS; AQ - *SA*: MINI	- Significant associations between total AQ scores and anxiety, depression, SA, for AS participants (all $p < 0.042$)

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Table 1 (*continued*)

Study: First author, Date, Location, Main focus, Theme addressed	Participants	Measures	Results
Lever and Geurts (2016), Netherlands Investigation into psychiatric comorbidity in adults Theme i, ii	ASD (n = 172) - *ASD: Young* (n = 52) % male: 63 (n = 33); age: mean 29.3 - *ASD: Middle* (n = 72) % male: 63 (n = 45); age: mean 47.9 - *ASD: Older* (n = 48) % male: 79 (n = 38); age: mean 63.7 all recruited via clinical services and adverts NCC (n = 172) - *NCC: Young* (n = 60) % male: 62 (n = 37); age: mean 26.8 - *NCC: Middle* (n = 47) % male: 49 (n = 23); age: mean 47.0 - *NCC: Older* (n = 65) % male: 57 (n = 37); age: mean 63.0 all recruited via adverts at university and social media		- Significant associations between general anxiety and self-reported and clinician-rated ASD measures (all $B > 0.4$, all $p < 0.05$)
Maddox and White (2015), USA Investigation into SA in clinical and non-clinical samples Theme iii, v	- *ASD* (n = 28) % male: 54 (n = 15); % ethnicity: Caucasian 79, Hispanic/Latino 4, African-American 0, Asian-American 11; age: mean 23.9, sd 6.9, range 16–42; IQ: mean 107, sd 17; recruited via university and research databases, clinical and non-statutory community services - *SAD* (n = 26) % male: 50 (n = 13); % ethnicity: Caucasian 77, Hispanic/Latino 8, African-American 0, Asian-American 4; age: mean 26.0, sd 7.1, range 16–42; IQ: mean 109, sd 11; recruited via adverts - *NCC* (n = 25) % male: 48 (n = 12); % ethnicity: Caucasian 68, Hispanic/Latino 0, African-American 12, Asian-American 12; age: mean 24.8, sd 7.3, range 17–44; IQ: mean 114, sd 11; recruited via adverts at university, and clinical and community settings	- *ASD*: ADOS - *SA*: BFNE; SASPA; SIAS; SRS-2A; MINI	- Significant differences in SRS, social communication, social motivation and total scores in the ASD group: ASD + SA > ASD-SA ($d > 0.82$, $p < 0.05$) - Individuals with ASD + SA considered social skills impairment to be a contributory factor, much more so than the SA only group ($p = 0.004$)
Magiati et al. (2016), Singapore Investigation into ASD functioning, sex, age and anxiety in young people Theme ii, iv	- *ASD* (n = 241) % male: 82 (n = 197); % ethnicity: Chinese 77, Malay 10, Indian 7; age: mean 10.4, sd 3.0, range 6–18; recruited via special needs schools	- *ASD*: DBC screener - *SA*: SCAS - *Behaviour*: DBC	- Significant associations between SCAS total and DBC anxiety subscales ($r = 0.63$, $p < 0.001$) - Significant positive associations between adaptive functioning and SA ($r = 0.22$, $p < 0.001$) - Non-significant associations between repetitive behaviour and speech, and social communication symptoms, and SA - Predictor variables for SA included adaptive functioning (all $B > 0.13$, all $p < 0.05$), but not ASD symptoms as measured by the DBC
Meyer et al. (2006), USA Investigation into relationships between psychiatric symptoms and information processing and attribution style Theme iv, v	- *AS* (n = 31) % male: 84 (n = 26); age: mean 10.1, sd 1.9, range 8-14; V mental age: mean 11.2, sd 2.1; recruited via clinical database - *NCC* (n = 33) % male: 73 (n = 24); age: mean 10.2, sd 1.9, range 8–14; V mental age: 11.4, sd 2.1; recruited via research studies or education	- *ASD*: ASSQ; ASAS - *SA*: SAS-CR - *Behaviour*: BASC - *Social competence*: SCI	- Significant positive associations between FNE and BASC scores ($r = 0.4$, $p < 0.06$) - Significant associations between pro-social skills and sensitivity to rejection: increased sensitivity was correlated with poorer pro-social skills ($r = -0.38$, $p < 0.05$)
Orinstein et al. (2015), USA Investigation into psychiatric co-morbidity in clinical and non-clinical samples Theme ii		- *ASD*: ADOS - *SA*: K-SADS-PL	- Significant associations between ASD and psychiatric symptoms: higher ADOS scores were associated with higher K-SADS-PL, in particular for depression, SA, GAD and ADHD (all current $r > 0.29$, all $p < 0.004$; all past $r > 0.21$, all $p < 0.04$)

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Table 1 (*continued*)

Study: First author, Date, Location, Main focus, Theme addressed	Participants	Measures	Results
	- *ASD-OO* (n = 33) % male: 79 (n = 26); age: mean 12.8, sd 3.5, range 8–21; VIQ: mean 112, sd 13.3; PIQ: mean 110, sd 15.3; recruited via prior research study - *HFA* (n = 42) % male: 90 (n = 38); age: mean 13.9, sd 2.7, range 9–20; VIQ: mean 106, sd 14.7; PIQ: mean 111, sd 12.5; recruited via prior research study - *NCC* (n = 34) % male: 91 (n = 31); age: mean 13.9, sd 2.6, range 10–22; VIQ: mean 112, sd 11.2; PIQ: mean 113, sd 11.3; recruited via prior research study		
Perry et al. (2015), Israel Investigation into relationships between interpersonal distance and SA Theme vii	- *ASD* (n = 13) % male: 92 (n = 12); age: mean 25.0; recruitment source unclear - *NCC* (n = 13) % male: 100 (n = 13); age: mean 24.0; recruitment source unclear	- *ASD*: ADI-R or ADOS or no formal measure - *SA*: LSAS - *Interpersonal distance*: stop-distance paradigm, comfortable distance task	- Significant associations between SA and interpersonal distance for the ASD, but not NCC group ($r = 0.59$, $p < 0.05$)
Scharfstein et al. (2011), USA Investigation into social behaviours and verbal communication in clinical and non-clinical samples Theme iii, vi	- *AS* (n = 30) % male: 87 (n = 26); % ethnicity: Caucasian 90, Latino 3; age: mean 10.6, sd 1.6, range 7–13; FIQ: mean 114, sd 14.1; recruited via research studies - *SA* (n = 30) % male: 77 (n = 23); % ethnicity: Caucasian 60, African-American 23, Latino 3, Asian 10; age: mean 10.0, sd 1.8, range 7–13; recruited via research studies - *NCC* (n = 30) % male: 73 (n = 22); % ethnicity: Caucasian 37, African American 30, Latino 20; age: mean 10.6, sd 2.0, range 7–13; recruitment via research studies	- *ASD*: ADI-R - *SA*: ADIS-C/P; SPAI-C; SAM - *Behaviour*: SRPA: brief scenarios of interaction with peers of a similar age	- Non-significant differences in observer-ratings of social skills in AS participants scoring above and below the SA threshold
Simonoff et al. (2008), UK Investigation into rate of psychiatric comorbidity and associations between these and demographic characteristics Theme ii	- *ASD* (n = 112) - % male: 88 (n = 98); % ethnicity: Caucasian 95; age: mean 11.5, range 10–14; FIQ: mean 73, sd 21.6, range 19–174; epidemiological sample	- *ASD*: ADOS, ADI-R, SCQ - *SA*: CAPA	- Non-significant associations between ASD and SA
South et al. (2011), USA Examination of relationships between IQ, social functioning, anxiety, and psychophysiological responses Theme i	- *ASD* (n = 30) % male: 90 (n = 27); age: mean 12.4, sd 2.7, range 8–18; FIQ: mean 106, sd 11.9; recruited via clinical settings, schools and adverts - *NCC* (n = 30) % male: 87 (n = 26); age: mean 13.2, sd 3.1, range 8–18; FIQ: mean 109, sd 9.0; recruitment source unclear	- *ASD*: ADOS; SCQ - *SA*: SCARED	- Significant positive associations between skin conductance response, social functioning and social anxiety in the ASD group ($r = -0.45$, $p < 0.05$)
Spain et al. (2016), UK Investigation into SA, ASD and socio-emotional processing Theme i, ii	- *ASD* (n = 51) % male: 100 (n = 51); age: mean 26.3, sd 5.8, range 19–42; VIQ: mean 108, sd 14.9; PIQ: mean 105, sd 15.8; recruited via previous research study	- *ASD:* ADOS; ADI-R; AQ - *SA:* LSAS; BFNE; SPS; SIAS	- Non-significant associations between SA, the ADOS or ADI; significant associations between self-rated ASD on the AQ and SA (all $r > 0.38$, $p < 0.04$)
		- *ASD*: ADI-R - *SA*: CASI - *Behaviour*: VABS; ABC	- Significant associations between anxiety (total scores), functional language and stereotyped behaviour: increased anxiety was correlated with increased impairment (all $B > 0.1$, all $p < 0.05$)

(*continued on next page*)

Table 1 (*continued*)

Study: First author, Date, Location, Main focus, Theme addressed	Participants	Measures	Results
Sukhodolsky et al. (2008), USA Examination of rates and correlates of anxiety Theme ii	- *PDD* (n = 171) % male: 84 (n = 144); % ethnicity: Caucasian 70, African-American 12, Latino 6, Asian 8; age: mean 8.2, sd 2.6, range 5–17; FIQ: range profound disability to no intellectual disability; recruited via research studies		
Swain et al. (2015), USA Examination of relationships between social motivation, emotion dysregulation, and SA Theme v	- *ASD* (n = 69) % male: 71 (n = 49); % ethnicity: Caucasian 60, African-American 3, Latino 12, Asian 17; age: mean 20.5, sd 2.0, range 17–27; recruited from clinical settings or research programs	- *ASD*: SRS - *SA*: SAS	- Significant negative associations between SA, and social motivation and emotion dysregulation (all $\beta > 0.22$, all $p < 0.05$) - Significant predictors of informant-ratings of SA included goal-directed behaviour for negative emotions, impaired awareness of emotions, and social motivation (all $\beta > 0.24$, all $p < 0.05$)
Usher et al. (2015), USA Investigation into interactions between people with and without ASD, and relationships between social competence, theory of mind, and SA Theme iv	- *ASD* (n = 39) % male: 87 (n = 34); age: mean 13.9, sd 2.8, range 10–18; VIQ: mean 103, sd 15.4; recruited from an existing research study - *NCC* (n = 39) % male: 87 (n = 34); age: mean 14.1, sd 2.4, range 10–18; VIQ: mean 108, sd 11.6; recruited via schools - % ethnicity across groups: Caucasian 40,African-American 3, Latino 53, Asian 1	- *ASD*: ADOS; SCQ; ASSQ - *SA*: SAS-CR - *Social competence*: get to know you, teaching, and teamwork tasks	- Significant associations between social initiation and theory of mind in the ASD group ($\beta = 0.58$, $p = 0.01$)
White and Roberson-Nay (2009), USA Examination of relationships between anxiety, loneliness, and social skills deficits Theme ii, iii, iv, v	- *ASD* (n = 20) % male: 90 (n = 18); age: mean 12.1, sd 1.8, range 7–14; IQ: mean 92, sd 14.4; recruited via outpatient clinical setting	- *ASD*: ADOS; SCQ; SRS - *SA*: MASC - *Social competence*: SCI	- Significant associations between affect and initiation of social interaction: increased general anxiety and depression was associated with reduced propensity to initiate social interaction ($r = -0.59$, $p < 0.05$) - Non-significant associations between social skills and anxiety - Non-significant associations between anxiety, and ASD symptoms or social competence
White et al. (2015), USA Investigation into relationships between SA and eye fixation to facial expressions Theme ii	- *ASD* (n = 15) % male: 53 (n = 8); % ethnicity: Caucasian 80, African-American 7; age: mean 14.9, sd 1.6, range 12–17; recruited via clinical setting, research database and adverts - *NCC* (n = 18) % male: 56 (n = 10); % ethnicity: Caucasian 94, African-American 6; age: mean 4.3, sd 1.5, range 12–17; recruited via adverts and research databases	- *ASD*: ADOS; ADI-R; SRS; SCQ - *SA*: BFNE; SWQ	- Non-significant associations between ASD characteristics and SA in the ASD group; significant associations between ASD characteristics and parent-reported SA in the NCC participants ($p < 0.01$)

ASD measures: ADOS – autism diagnostic observation schedule; ADI-R – autism diagnostic interview–revised; AQ – autism quotient; HAGS – high-functioning autism/Asperger syndrome global scale; SCQ – social communication questionnaire; SRS – social responsiveness scale (adult); ASSQ – autism spectrum screening questionnaire; ASAS – Australian scale for Asperger's syndrome; *Measures of psychiatric symptoms*: SCID – structured clinical interview for DSM-IV; LSAS – Liebowitz social anxiety scale; MINI – mini international neuropsychiatric interview; SADS – social avoidance and distress scale; BFNE – brief fear of negative evaluation scale; SASPA – social anxiety scale for people with ASD; SPS – social phobia scale; SIAS – social interaction anxiety scale; SSRS – social skills rating scale – ADIS-IV – anxiety disorders interview schedule for DSM-IV; SAS – social anxiety scale (c – children, a – adolescence); MASC – multi-dimensional anxiety scale for children; SPAI – social phobia and anxiety inventory; SWQ – social worries questionnaire; SCAS – Spence children's anxiety scale; CASI – child and adolescent symptom inventory (4R); SCARED- screen for child anxiety related emotional disorder; K-SADS-PL – schedule for affective disorders and schizophrenia for school age children, present and lifetime version; SDS – social desirability scale; SAM – self-assessment manikin; CAPA – child and adolescent psychiatric assessment;. *Behavioural measures*: CSBQ – children's social behavioural questionnaire; SCI – social competence inventory; SRPA – structured role-play assessment; BASC – behaviour assessment system for children; ABC – aberrant behaviour checklist; VABS – Vineland adaptive behaviour scale.

Table 2
Quality assessment of included studies.

Study	Selection bias	Study design	Confounders	Data collection	Withdrawals/drop outs	Global ratings
Bejerot et al. (2014)	W	M	M	S	W	W
Bellini (2004)	M	W	W	W	M	W
Bellini (2006)	M	W	W	W	M	W
Capriola et al. (2016)	M	M	M	S	W	M
Cath et al. (2008)	M	M	M	M	W	M
Chang et al. (2012)	M	W	W	M	W	W
Chen et al. (2016)	M	M	W	W	W	W
Corden et al. (2008)	M	M	M	M	W	M
Hallett et al. (2013)	S	S	S	M	M	S
Kanai et al. (2011)	M	M	W	W	W	W
Lever and Geurts (2016)	M	M	M	M	W	M
Maddox and White (2015)	M	M	M	S	M	M
Magiati et al. (2016)	M	W	W	W	W	W
Meyer et al. (2006)	M	M	M	W	W	W
Orinstein et al. (2015)	M	M	M	M	M	M
Perry et al. (2015)	W	M	W	W	W	W
Scharfstein et al. (2011)	M	M	M	S	W	M
Simonoff et al. (2008)	S	S	S	M	M	S
South et al. (2011)	M	M	M	W	W	W
Spain et al. (2016)	W	M	M	M	W	W
Sukhodolsky et al. (2008)	M	W	M	M	W	W
Swain et al. (2015)	M	W	W	W	W	W
Usher et al. (2015)	M	M	M	W	W	W
White and Roberson-Nay (2009)	M	W	M	M	W	W
White et al. (2015)	M	M	M	W	W	W

Ratings: W – weak; M – moderate; S – strong.

instrument, and four studies included multiple measures including a clinician-administered assessment. Psychometric properties of psychopathology measures, e.g. internal consistency, were largely unreported. Further, studies typically relied on normative cut-off scores (indicating clinical caseness) using those thresholds cited for the non-ASD population, although whether these normative values also apply to individuals with ASD is uncertain.

It is noteworthy that there are overlaps in some of the constructs assessed by the ASD and SA measures, but also differences. As outlined in Table 3, domains such as general social skills, social competence, affect and physical sensations, empathy and attention were potentially assessed by both ASD or anxiety measures. Domains such as concerns about negative evaluation or performance, quality and quantity of communication, and general or specific ways of coping, were assessed as a facet of either type of measure, but not generally both.

Although all studies were cross-sectional, we assessed the degree to which information was provided about response rates and withdrawal. Limited data were provided about possible differences between non-responders and responders, e.g. in terms of demographic characteristics or clinical symptoms. Further, most studies provided limited information about the number of participants, if any, who consented to take part, but subsequently withdrew from the study, or who took part and then withdrew consent for their data to be used.

Overall, the most common methodological limitations across studies concerned: 1) the reliance on inclusion of participants from clinical and research contexts, rather than epidemiological or non-treatment seeking samples; 2) measurement issues, whereby core and or co-morbid symptoms were not assessed using robust measures, the validity and reliability of some measures was not established, and also, that there was duplication or overlaps in constructs assessed; and 3) that studies were insufficiently powered to detect potential differences between groups, or samples were too small to be able to establish if findings were mediated by variables such as sex and age. Table 2 lists global ratings for each study. In summary, two studies were considered strong, ten studies were considered moderate, and fifteen were considered weak.

2.3. Summary of results

Study results are clustered into themes, as follows: relationships between SA and i) self-reported ASD; ii) clinician-rated ASD; iii) social skills; iv) social competence; v) social motivation; vi) speech latency; and vii) interpersonal distance.

Sixteen studies (Bejerot et al., 2014; Capriola et al., 2016; Cath et al., 2008; Corden et al., 2008; Hallett et al., 2013; Kanai et al., 2011; Lever & Geurts, 2016; Maddox & White 2015; Magiati et al., 2016; Orinstein et al., 2015; Simonoff et al., 2008; South et al., 2011; Spain et al., 2016; Sukhodolsky et al., 2008; White & Roberson-Nay 2009; White et al., 2015) explored relationships between ASD symptoms and SA in young people or adults with ASD, compared to NCC (n = 6 studies), clinically anxious and NCC groups (n = 5), or in single samples (n = 5). Findings were mixed, which may be partly attributable to differences in recruitment sources (epidemiological vs. clinical sampling frames) as well as the type of measure used to assess core or co-occurring symptoms, as well as who completed these (e.g. participants themselves, informants or clinicians).

Table 3
Measures used.

Measure	Number of times used	Method of rating	Domain assessed						
			Social motivation	General social skills	Quality of communication	Reciprocity	Objective rating of social competence	Worries about social competence	Fear of negative evaluation
ASD									
ADI	8	IR	X	X	X	X	X		
ADOS	16	CR	X	X	X	X	X		
AQ	6	SR	X	X	X	X			
ASAS	1	IR	X	X	X	X	X		
ASSQ	2	IR	X	X	X	X	X		
HAGS	1	CR	X	X	X	X	X		
SCQ	5	IR	X	X	X	X	X		
SRS	5	SR, IR	X	X	X	X	X		
Social anxiety									
ADIS-IV	3	CR					X	X	X
BFNE	4	SR						X	X
CAPA	1	IR			X	X	X	X	X
CASI	1	CR	X					X	
K-SADS-PL	1	CR		X			X	X	X
LSAS	5	SR							
MASC	2	SR, IR						X	X
MINI	3	CR					X	X	X
SAS	4	SR							X
SASPA	1	SR						X	X
SCARED	1	SR, IR						X	X
SCAS	1	SR, IR						X	X
SCID	2	CR						X	X
SDS	1	SR	X						
SIAS	1	SR		X				X	X
RCADS	1	SR, IR						X	X
SPAI	2	SR						X	X
SPS	1	SR						X	X
SSRS	2	IR, SR		X		X			
SWQ	1	SR, IR						X	
Behaviour									
ABC	1	IR, SR	X	X	X				
BASC	2	IR	X	X		X			
SCI	2	IR	X	X			X		
VABS	1	IR, CR		X	X	X	X		

Measure	Domain assessed								
	Coping: avoidance	Coping: general strategies	Repetitive behaviours	Emotions and feelings	Empathy	Attention	Imagination	Adaptive functioning	Interests
ASD									
ADI	X	X	X	X	X		X	X	X
ADOS			X	X	X		X		X
AQ						X	X		
ASAS	X		X	X	X		X		X
ASSQ			X		X				X
HAGS					X			X	X
SCQ			X		X		X		X
SRS			X	X	X		X		
Social anxiety									
ADIS-IV	X	X						X	
BFNE									X
CAPA	X	X		X				X	
CASI			X	X				X	
K-SADS-PL		X		X		X		X	
LSAS	X			X					
MASC		X		X					
MINI	X							X	
SAS	X			X					
SASPA	X	X		X					

(*continued on next page*)

Table 3 (*continued*)

Measure	Domain assessed								
	Coping: avoidance	Coping: general strategies	Repetitive behaviours	Emotions and feelings	Empathy	Attention	Imagination	Adaptive functioning	Interests
SCARED				X					
SCAS		X	X	X					
SCID	X	X		X					X
SDS	X		X	X	X				
SIAS				X					
RCADS			X	X					
SPAI	X			X					
SPS				X					
SSRS				X	X				
SWQ	X			X					
Behaviour									
ABC		X	X	X				X	
BASC			X	X		X		X	
SCI					X				
VABS						X		X	

SR – self-report; IR – informant-report; CR – clinician-rated.
ASD measures: ADOS – autism diagnostic observation schedule; ADI-R – autism diagnostic interview–revised; AQ – autism quotient; HAGS – high-functioning autism/Asperger syndrome global scale; SCQ – social communication questionnaire; SRS – social responsiveness scale (adult); ASSQ – autism spectrum screening questionnaire; ASAS – Australian scale for Asperger's syndrome; *Measures of psychiatric symptoms*: SCID – structured clinical interview for DSM-IV; LSAS – Liebowitz social anxiety scale; MINI – mini international neuropsychiatric interview; SADS – social avoidance and distress scale; BFNE – brief fear of negative evaluation scale; SASPA – social anxiety scale for people with ASD; SPS – social phobia scale; SIAS – social interaction anxiety scale; SSRS – social skills rating scale – ADIS-IV – anxiety disorders interview schedule for DSM-IV; SAS – social anxiety scale (c – children, a – adolescence); MASC – multi-dimensional anxiety scale for children; SPAI – social phobia and anxiety inventory; SWQ – social worries questionnaire; SCAS – Spence children's anxiety scale; CASI – child and adolescent symptom inventory (4R); SCARED – screen for child anxiety related emotional disorder; K-SADS-PL – schedule for affective disorders and schizophrenia for school age children, present and lifetime version; SDS –social desirability scale; SAM – self-assessment manikin; CAPA – child and adolescent psychiatric assessment;. *Behavioural measures*: SRPA – structured role-play assessment; BASC – behaviour assessment system for children; ABC – aberrant behaviour checklist; VABS – Vineland adaptive behaviour scale.

Five of these studies (Bejerot et al., 2014; Cath et al., 2008; Kanai et al., 2011; Lever & Geurts 2016; Spain et al., 2016) investigated associations between *self-reported* SA on the LSAS (a measure of anxiety about and avoidance of specific social situations) and self-reported ASD on the AQ (a measure of traits associated with ASD, including communication, social skills, imagination, attention to detail and attention switching). All studies reported significant positive relationships between these measures: higher ASD traits were associated with increased SA symptoms. Of note, one study (Bejerot et al., 2014), which compared adults with ASD to SA and NCC participants, found that these associations only held true for the ASD group. Another study (Corden et al., 2008) administered the AQ and examined the relationships between this and two self-rated SA measures, the Social Phobia Anxiety Inventory (a measure of thoughts, feelings and behaviours associated with social anxiety; SPAI, Turner et al., 1999) and Social Desirability Scale (a measure of personality traits and attitudes indicative of socially desirable behaviour and adherence to social norms and conventions; SDS, Crowne & Marlowe, 1960), reporting non-significant associations. Relationships between the BFNE (a self-report scale relating to thoughts and beliefs characteristic of social evaluative concerns; Leary, 1983) and AQ were assessed in two studies (Capriola et al., 2016; Spain et al., 2016), only one of which reported significant positive associations (Spain et al., 2016). Correlations between the Social Interaction Anxiety and Social Phobia Scales (which together, assess thoughts, feelings and avoidance behaviours associated with social anxiety) (SIAS and SPS; Mattick & Clarke, 1998) and the AQ were positively correlated in the one study to investigate this (Spain et al., 2016).

When looking at links between domain scores on *clinician*-rated ASD assessments, most commonly the ADOS (Lord et al., 2000) and ADI-R (Lord et al., 1994), and SA, six studies (Hallett et al., 2013; Magiati et al., 2016; Simonoff et al., 2008; Spain et al., 2016; White & Roberson-Nay, 2009; White et al., 2015) found non-significant associations. Conversely, two studies (Hallett et al., 2013; Orinstein et al., 2015) showed significant relationships between parent-rated (as opposed to self-rated) anxiety and ASD severity, and one study (Lever & Geurts, 2016) described significant associations between general anxiety and clinician-rated measures: in each of these studies, increased ASD characteristics and associated impairment was associated with elevated SA ratings. Only one study (Sukhodolsky et al., 2008) using these ASD assessments reported significant relationships between higher total anxiety scores and increased stereotyped behaviours.

Five studies, described in six articles (Bellini, 2004, 2006; Chang et al., 2012; Maddox & White 2015; Scharfstein et al., 2011; White & Roberson-Nay, 2009), examined associations between social skills and SA, primarily using observational behavioural rating scales. Several studies (Bellini, 2004, 2006; Chang et al., 2012; Maddox & White 2015) described significant negative associations

between SA and social skills, including assertiveness, self-control, co-operation and responsibility. In contrast, one study (White & Roberson-Nay, 2009) found no significant relationships. Compared to young people with Asperger syndrome and NCC (Scharfstein et al., 2011), individuals with SA had marginally poorer social skills during structured role-play assessments (SRPA). When assessed by blinded observers, social skills did not differ significantly between young people with Asperger syndrome and SA vs. those with Asperger syndrome alone. While self-reported poorer social skills were significantly correlated with increased SA, this was not the case for parent-ratings (Bellini, 2004).

Four studies (Chang et al., 2012; Meyer et al., 2006; Usher et al., 2015; White & Roberson-Nay, 2009) examined associations between SA and social competence or functioning in children and adolescents. While one study (White & Roberson-Nay, 2009) found no significant relationships, the remaining studies found that these were linked whereby poorer functioning correlated with elevated SA scores. In two studies (Chang et al., 2012; Magiati et al., 2016), social and adaptive functioning was significantly poorer in young people with ASD. Two studies (Meyer et al., 2006; Usher et al., 2015) found that relative to a NCC group, participants with ASD or Asperger syndrome appeared less socially competent, and were significantly less likely to initiate social overtures, pro-social behaviour, or display reciprocity.

Seven studies (Bellini, 2004; Capriola et al., 2016; Chang et al., 2012; Maddox & White, 2015; Meyer et al., 2006; Swain et al., 2015; White & Roberson-Nay, 2009) examined relationships between SA and either social motivation or propensity to initiate overtures, measured using self- or informant- rated questionnaires, including the Social Competence Inventory (a measure of social skills and responses; SCI, Rydell et al., 1998). Findings across studies were consistent, irrespective of participants' ages and measures administered. Significant positive associations were found between SA and increased interpersonal sensitivity, reduced social motivation, decreased assertiveness, reduced propensity to initiate social interactions, and general pro-social behaviour (Bellini, 2004; Chang et al., 2012; Maddox & White, 2015; Meyer et al., 2006; Swain et al., 2015; White & Roberson-Nay, 2009).

One study (Scharfstein et al., 2011) measured speech quality and response latency of children with ASD, compared to SA and NCC during SRPA. SA participants displayed significantly longer speech latency, relative to the other groups. SA participants also had significantly less range in vocal pitch, intensity and variability.

Finally, one study (Perry et al., 2015) explored relationships between SA and preferred physical interpersonal distance. Comparing adult ASD and NCC groups, differences in SA and mean preferred distance were not significant, although the variance of preferred distance did differ. Further, there were significant positive associations between SA and interpersonal distance for the ASD group only.

3. Discussion

Individuals with ASD commonly experience SA, with rates far exceeding non-ASD population norms. It is conceivable that risk and maintaining mechanisms for SA in ASD partially reflect core socio-communication impairments and/or a tendency towards engaging in restricted interests and repetitive behaviours. We undertook a systematic search for empirical data examining potential associations between ASD and SA symptoms, and included 24 studies described in 25 papers in the resulting narrative analysis. Studies were methodologically and clinically heterogeneous. A wide range of ASD and SA self- and informant-rated measures were used in diverse child, adolescent and adult samples, all of which precluded formal meta-analysis.

The main aim of the review was to establish whether there is empirical data to support the hypothesis that ASD and SA symptoms are associated. A relatively consistent trend in the data indicated that correlations are significant when assessed via self-ratings (of both ASD and SA) (Bejerot et al., 2014; Cath et al., 2008; Kanai et al., 2011), but not necessarily when measured via parent-ratings (Hallett et al., 2013; Simonoff et al., 2008). This may reflect common methods variance, whereby correlations between measures from the same informant may be inflated. Negative self-image, or depression, might lead to more severe self-ratings for both ASD and SA. It may also be the case that individuals with ASD and parents report higher levels of SA when in fact they are describing ASD characteristics (e.g. social difficulties). More generally, how self-report questionnaires operate for individuals with ASD is yet to be definitively established. For example, it may prove more difficult for informants to accurately endorse cognitive and affective characteristics, compared to behaviours, indicative of SA, because these are less overtly evident. Studies employing multiple methods of assessment, such as self-rated questionnaires, and clinician-administered interviews and biological measures (e.g. of anxiety) may aid with understanding discrepancies between these ratings.

Narrative synthesis of the data also indicated that there were significant relationships between elevated SA scores and poorer social skills and social competence. This included general skills as well as specific skills e.g. relating to the quality and quantity of verbal and non-verbal communication and degree of reciprocity. It is unclear whether these impairments are solely attributable to ASD, or if in fact these represent features of early onset SA, given that social skills impairments may contribute to SA (or exacerbate SA symptoms) (Beidel et al., 2010; Halls et al., 2015). It is surprising that in one study, SA controls seemed to have poorer social skills compared with ASD participants (Scharfstein et al., 2011). Similar findings have been reported in a comparable study of social skills in ASD and SA cohorts (Wong et al., 2012), albeit that the social skills of individuals with SA (and no diagnosed ASD) are not necessarily significantly different to non-SA (and non-ASD) samples; rather, it is a self-perception that social competence is poorer (Clark, 2001; NICE, 2013a). Perhaps in this instance, the testing appointment evoked heightened anxiety, and thus, anxious controls appeared less reciprocal and quieter in demeanour. Alternatively, SA controls may have had ASD traits (undiagnosed), compounding these impairments. Further studies comparing SA and ASD (with and without SA) groups on socio-cognitive tasks or measures of the quality and quantity of social skills, are needed to better understand these findings.

A further tentative theme emerged, namely, that poorer social motivation, assessed via self- and informant-rating scales was associated with increased levels of SA. Risk and causal mechanisms for (diminished) social motivation, in studies reviewed, were not

explicitly or fully investigated. At least five explanations seem possible: 1) this represents a core ASD characteristic; 2) this is attributable, at least in part, to an innate proneness for behavioural inhibition – a temperament associated with SA in non-ASD samples, and also observed in individuals with ASD (Stein, Chavira, & Jang, 2001); 3) this manifests as a consequence of negative social experiences, perhaps due to the impact of ASD characteristics, whereby individuals become less motivated to engage socially; 4) this is a consequence of SA; and/or 5) a combination of these factors. While this is beyond the scope of the findings described in this review, we would speculate that social motivation is comprised of cognitive, affective and behavioural elements. For example, positive and negative thoughts and beliefs about social situations and the utility and importance of these; emotional or physiological responses occurring during social situations (or indeed, before or afterwards); and varied behavioural responses which are helpful and encourage individuals to engage socially, or indirectly unhelpful and encourage avoidance (and thus, perpetuate negative thoughts). Further studies using longitudinal and/or intervention designs are needed to disentangle causal and maintaining factors for social motivation, both in individuals with ASD and individuals with ASD and SA.

In the wider literature, it has been proposed that anxiety in individuals with ASD may be partly related to restricted and repetitive behaviours, and sensory aversions. On the whole, study findings reported here do not suggest that there are strong links between these core ASD characteristics and SA, either when measured using self-report questionnaires or informant-ratings. It is possible that the methods of assessment, primarily focusing on ASD domain scores (e.g. on the ADOS) rather than particular sensory experiences or repetitive behaviours, lacked specificity, i.e. measuring general rather than unique experiences. Alternatively, it may be that the drivers for social anxiety in ASD are more related to socio-communication impairments, or their impact, than sensory characteristics. This perhaps highlights the importance of multi-informant ratings of core and co-morbid symptoms in future research.

3.1. Generalisability of study findings

Several factors affect the generalisability of study findings. Sampling methods varied between studies: the proportion of participants recruited from or involved with clinical services is unknown. There maybe differences in the demographic characteristics or other clinical outcomes of individuals who are treatment-seeking, compared with those people recruited from community or epidemiological sources. It is possible that individuals who considered that they have either minimal or severe SA were deterred from participating, thereby skewing the sample and data obtained. Overall, study samples were small. Also, methods used to assess ASD and co-morbidity varied somewhat according to age: informant-ratings were, on average, more likely to be obtained for younger rather than older participants, with little investigation of age-related effects. In some cases, the number of participants in ASD and comparison groups was unequal, which may have meant that there was insufficient power to detect possible differences (or the magnitude of these) between groups. Ethnicity data were not consistently reported, but there does appear to have been an over-representation of Caucasian individuals. As there may be cultural differences in the presentation of SA, and the psychometric properties of psychopathology measures (e.g. Asnaani et al., 2015; Hsu et al., 2012), it is not clear whether findings are valid for non-Caucasian samples. Also, most participants were male; we cannot be sure that drivers for SA in females with ASD are precisely the same as for males, given hypothesised sex differences in core symptoms and use of camouflaging strategies. Most participants had an IQ in the average range. It may be that the range and or levels of SA symptoms in individuals with a concurrent ID differ from those without. SA symptoms were measured using instruments which have not yet been validated for ASD samples (Kreiser & White, 2014). This suggests a degree of caution may be needed when interpreting study findings, as normative thresholds may differ between clinical and non-clinical samples. Finally, as noted above, all studies were cross-sectional, thereby limiting causal interpretations of data.

3.2. Limitations and considerations

We note several limitations to this review. We omitted non-English language publications due to resource constraints. Findings may therefore not reflect those of studies published in other languages, or in non-Western settings. We excluded studies in which SA scores were amalgamated with other data (e.g. summed anxiety totals), meaning that we may have inadvertently omitted relevant, but inaccessible, data. Finally, we did not have resources to contact researchers working in the ASD field to establish if any unpublished data were available.

Although not a limitation as such, it is important to consider issues pertaining to assessment of core and co-morbid symptoms and the potential impact this has for study findings and synthesis of data described here. As is commonplace, researchers utilised a broad range of measures. In samples of young people, informant-based ratings were often incorporated; in adult samples, self-report questionnaires were more frequently used. In studies where the same informant rated both ASD and SA, correlations may be inflated due to common methods variance; indeed associations reported were generally lower when different informants (e.g. parent, clinician, self) provided ratings of the two constructs. Informants may also affect ratings for ASD groups differently from those for other groups; Hallett et al. (2013), for example, found lower self- than parent- ratings of general anxiety in teenagers with ASD, and the opposite pattern in typically developing teenagers, Different informants clearly have access to different perspectives, and multiple sources are clearly preferable in order to take into consideration potential factors such as insight, bias, and ability to judge against wider or age-relevant norms. While there was a degree of overlap (see Table 3), it is also evident that different studies assessed distinct aspects of social anxiety, ranging from affect and avoidance specifically (e.g. via the LSAS), to the degree of negative evaluation (e.g. with the BFNE), Which assessment tools are best suited to assess social anxiety in ASD is an interesting question which the current review cannot address.

3.3. Clinical implications

Building on the findings here, it seems important that clinicians are proactive in asking about behaviours and beliefs that may be indicative of SA in individuals with ASD. We cannot assume that individuals with ASD will seek advice or help for these anxiety symptoms, either because of core ASD traits, e.g. lack of social overtures, or the social evaluative concerns characteristic of SA. Assessment may be particularly important before, during or following times of transition (e.g. from school to college), as these periods involve multiple new social situations in new settings. The clinical assessment is likely to take longer – both in terms of session duration and number of appointments – so as to mitigate the potential impact of core socio-communication impairments, co-morbid difficulties (e.g., alexithymia; difficulty reflecting on and reporting own feelings), and socio-evaluative concerns. While brief face-to-face and telephone triage assessments for psychological therapy are offered routinely in UK NHS primary and secondary care settings, this is unlikely to be suitable for most individuals with ASD. Conceivably, self-report measures may be of use; the review findings indicate that the LSAS, BFNE and SAS have been most commonly used in empirical studies, although other measures may well have clinical utility. Given the range of SA measures described here, discussion with the clinical team or supervisors is a pragmatic step in decision-making about which measures are most appropriate. When consent permits and when appropriate, information from carers or teachers may enhance the assessment, particularly as more familiar adults may notice subtle changes in behaviour, e.g. avoidance of specific vs. general situations, or antecedents to anxiety. While cut-off thresholds delineating SA symptoms from the full-blown disorder are useful, it is noteworthy that sub-threshold symptoms can nevertheless be highly debilitating and cause substantial impairment.

Cognitive and cognitive behavioural interventions are a recommended treatment for social anxiety in non-ASD populations (NICE, 2013a). Preliminary evidence suggests that these may also be effective for reducing SA in ASD, albeit that there are very few intervention studies published (Spain, Sin, Harwood, Mendez, & Happé, 2017). Decisions about which interventions to offer first or concurrently, are best made on a case-by-case basis, ideally following discussion with patients and their significant others (NICE, 2013b). In light of the findings of this review, and the wider literature, individuals may benefit from skills-based interventions, such as those designed to enhance social skills or emotional literacy, before undertaking targeted SA work; or a combined approach (Spain, Blainey et al., 2017; White et al., 2013).

Assessment of change is an important aspect of treatment. Many UK NHS services are expected to utilise standard generic and disorder-specific self-report scales (NICE, 2012). Their utility for individuals with ASD, however, remains ambiguous. Perhaps the parsimonious approach is to use outcome measures that are standardised, but also potentially, those that are personalised and co-produced with patients, e.g. measuring subjective units of distress (commonly referred to as SUDS ratings). Moreover, the utility of outcome measures is likely to be enhanced if treating clinicians consider carefully how, when, where and by whom outcome measures are best completed.

3.4. Research implications

We suggest that future studies should incorporate multiple measures of SA, as well as two or more measures of the full range of core ASD symptoms. This may facilitate a more in depth understanding of cognitive, affective and behavioural facets of SA in ASD, and allow for examination of the psychometric properties of self- and informant-rated measures. Choice of specific outcome measures should be considered carefully, in order to avoid overlaps in constructs measured via ASD and SA, and also to facilitate comparisons between studies. Table 3 outlines measures used to date and this may inform decisions about replicability of self-and informant-instruments for future studies. Inclusion of a combination of biological, neuropsychological and standardised self-report and clinician-administered measures may help to illuminate the extent to which core ASD characteristics may be related to SA. Recruitment of clinical as well as NCC groups may help to shed light on whether there are unique and/or overlapping drivers for SA in ASD samples. Addition of an alexithymia measure would help to quantify the validity and reliability of self-report psychopathology measures. Also, studies should seek to establish similarities or differences in the SA symptom profile (and potentially, risk factors) in females as well as males across the lifespan, and in individuals with and without a concurrent ID. Finally, use of prospective longitudinal designs could help to identify causal mechanisms and ultimately, effective treatments for these commonly co-occurring symptoms.

4. Conclusion

It is unsurprising that individuals with ASD experience anxiety and worry about social interactions. A review of English-language publications has revealed that SA may be associated with socio-communication impairments, specific social skills and diminished social motivation. Links between restricted and repetitive interests and behaviours, and SA, are less well supported in the findings to date. The literature indicates that some of these symptoms may cause and/or maintain SA. Further studies – using qualitative and quantitative designs – are needed to extend the evidence base, so that prevention, early detection, and targeted interventions for SA can be put in place.

Authors' contributions

DS proposed and designed the review, and drafted the manuscript. JS and DS conducted the searches, reviewed the findings, and discussed and agreed studies to be included. JS, KL, JM and FH contributed to the manuscript. All authors have read, commented on and approved the final manuscript.

Conflict of interests

All authors declare that they have no conflict of interests.

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