#### **REVIEW PAPER**



## Anxiety in Children with Autism at School: a Systematic Review

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#### **Abstract**

Anxiety in autism is commonly reported by parents, but teacher reports of anxiety in their students with autism have received little attention. This paper presents the results from the first systematic review on anxiety in children with autism at school. Six intervention studies (five of which were based upon cognitive—behavioural therapy) and 26 descriptive/non-experimental studies met inclusion criteria. Sample populations from included studies were frequently drawn from psychiatric clinics, with females and children attending special schools underrepresented, making generalisability of results difficult. Few studies used anxiety-specific measures, with most reporting anxiety-related subscale means from broader emotional/behavioural questionnaires. While 19 studies included multiple informants, which is recommended practice, only seven studies combined parent, teacher and child reports of anxiety. Comparison between informants proved difficult, with varying sample sizes and few studies using the same measure across participant groups. To further our understanding of the presentation of anxiety in children with autism attending school, studies need to include multiple informants and, where possible, extend beyond reporting average scores from broad anxiety subscales to provide descriptions of presentation and symptomatology.

**Keywords** Autism · Anxiety · Mental health · School · Systematic review

Anxiety and worries were reported in a number of Leo Kanner's (1943) original case study descriptions of children with autism. However, it is only relatively recently that research has focussed upon this area, with a six-fold increase in papers published on the topic from 2005 to 2015 (Vasa et al. 2018). Anxiety disorders are now recognised as the most commonly co-occurring conditions within autism spectrum disorders, with meta-analyses suggesting prevalence rates of clinically significant anxiety of approximately 40% (van Steensel et al. 2011), compared to 13.4% of children worldwide without autism (Polanczyk et al. 2015). There is growing research to suggest that anxiety can exacerbate autism symptoms. Further, the relationship between anxiety and autism characteristics or profiles may be bidirectional (e.g. White et al. 2014), suggesting

difficulties, but also be a consequence of social and academic difficulties associated with autism characteristics. This suggestion is further supported by recent findings that anxiety symptomatology has a greater impact upon quality of life than autism characteristics in both child (Adams et al. 2019a) and adult (Smith et al. 2019) samples.

It is known that anxiety can impact on school performance

that the presence of anxiety may not only contribute to

for children without a diagnosis of autism, but research on the impact of anxiety on academic achievement for children with autism has been limited. It is evident, however, that children with autism are generally at risk of academic underachievement (see review by Keen et al. 2016) and participate in less school activities than expected for children their age (Simpson et al. 2018). It has been suggested that anxiety may be one of the barriers to successful school functioning in children with autism (Wood and Gadow 2010) and that anxiety symptomatology may be present in children with autism at the age they begin school (Adams et al. 2018a; Keen et al. 2017). The impact of autism and anxiety on the classroom has been noted by teachers, other professionals and parents, with anxiety being identified in the top 3 factors affecting school participation and the top 2 factors influencing classroom support needs (Saggers et al. 2016).



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Given the importance anxiety may play in the school context and on educational outcomes, research focussing upon anxiety within the school setting would seem particularly important. To advance our understanding of school anxiety, it is important to gain the teacher's perspective on anxiety, particularly given recent suggestions that the presentation of anxiety symptomatology may differ between home, school and community settings (Adams et al. 2018b). Input from teachers can be valuable as they spend a lot of time with children and are likely to see them in different, potentially anxiety-provoking situations compared to those observed by parents or primary caregivers (Lyneham et al. 2008). However, the majority of studies exploring anxiety in children with autism have to date relied solely on parent report (e.g. van Steensel et al. 2011). Parents may have only a limited knowledge of how anxiety may present for their child with autism at school, so gaining information from multiple informants across different contexts has the potential to provide a more comprehensive profile of anxiety and behaviour for students on the spectrum. For this and a number of other reasons, the use of multiple informants is recommended practice (Schniering et al. 2000).

Studies using multiple informants with typically developing children note frequent variability in informant agreement levels for internalising disorders such as anxiety (Achenbach et al. 1987; De Los Reyes et al. 2015), especially when ratings are provided across different settings with different demands and expectations (e.g. home and school). This variability has led to discussion around the possibility of differing presentations of anxiety symptomatology across settings. However, such multiinformant explorations are limited in autism, restricting the extent to which we are able to understand how anxiety may present at school compared with home and community contexts. Such information could be critical to help teachers identify anxiety in children with autism in their class, and therefore, instigate referral to evidence-based interventions to reduce anxiety within this setting.

So as to better understand how anxiety may present in children with autism in the school context, the aim of this study was to identify and review research that has focussed upon anxiety in children with autism in the school setting. The following research questions were posed:

- 1. What studies have reported upon anxiety in children with autism in the school setting?
- 2. Which measures have been used to report levels of anxiety in children with autism at school?
- 3. Is there concordance between the scores on teacher measures of anxiety with those completed by children on the autism spectrum and/or their parents?



The protocol for this systematic review was registered online with PROSPERO, an international register for systematic reviews with health-related outcomes (Registration number: CRD42017081739). Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) standards were followed for all stages of this systematic review.

## **Eligibility Criteria**

It is recommended (e.g. by COSMIN) to focus searches on a well-defined group and outcome. Therefore, the inclusion and exclusion criteria for this review required studies specifically to have at least a subscale of a measure or an intervention focussing upon anxiety (rather than broader emotional functioning) in children with autism in the school setting. Studies were included within this literature review if they met the following inclusion criteria: (a) study must report on anxiety in children within the school environment (including mainstream schools, special schools or specialist educational settings, either publicly or privately funded); (b) study focusses upon, measures, or describes the child's anxiety or anxiety symptomatology; (c) study must involve a sample of humans with autism with a mean age between 5 and 17 years; (d) autism diagnosis must be established through a reliable measure of autism (e.g. Autism Diagnostic Interview-Revised (ADI-R), Autism Diagnostic Observational Schedule (ADOS), and/or through educational or medical records); and (e) if there is a mixed-diagnosis or mixed-age sample, the anxiety data for the autism group of interest are reported separately. Reviews, opinions, dissertations, editorials, book chapters and descriptive pieces were not included in this systematic review. Case studies, single case designs and single subject design studies were also excluded because of the high risk of bias in this design and/or the possible lack of generalisability and representation of a case study to a broader population. This decision was taken in order to include as many studies as possible without jeopardising internal or external validity.

### **Search Strategy**

The electronic databases ERIC, MEDLINE, PsycINFO and Web of Science were searched during October 2017. The Cochrane library and PROSPERO database were also searched to confirm that no other systematic reviews of the current study topic existed or were registered as being in progress. The search was restricted to articles in English and those published after 1994, when Asperger's disorder was entered into the DSM-IV. The search strategy included terms pertaining to autism, anxiety and school (see Table 1).



Table 1 Search strategy

Autism	Anxiety	School
Autis* OR Asperger* OR asd OR pdd OR pervasive developmental disorder	anxi* OR psycho* OR mental health OR emotion* OR internali* OR psychiatric	teacher* OR school* OR educat* OR academic OR informant

<sup>\*</sup>Search strings were connected by the Boolean operator "AND"

#### **Review Strategy**

Electronic searches resulted in 10,598 records being identified (see Fig. 1). Following removal of duplicates, the titles and abstracts of 8139 articles were assessed against inclusion

criteria by the primary rater, and a random sample of 20% was double reviewed by a second rater who was blind to the other researcher's ratings. This is accepted practice when a review is large and resources are limited (Pettigrew and Roberts 2005). The double reviewing resulted in a 99.3% agreement. The 11 articles where the ratings differed were discussed, and a consensus agreement was reached for two of the papers. A third rater was consulted for the remaining nine (0.6%) papers.

Where inclusion or exclusion could not be determined by the title and abstract alone, the full text was assessed by a primary rater and, with a random sample of 20%, double reviewed by a second rater blind to the other researcher's ratings. There were no disagreements at this stage. All papers which met inclusion criteria were double rated and data were extracted by two independent raters. In addition, once studies

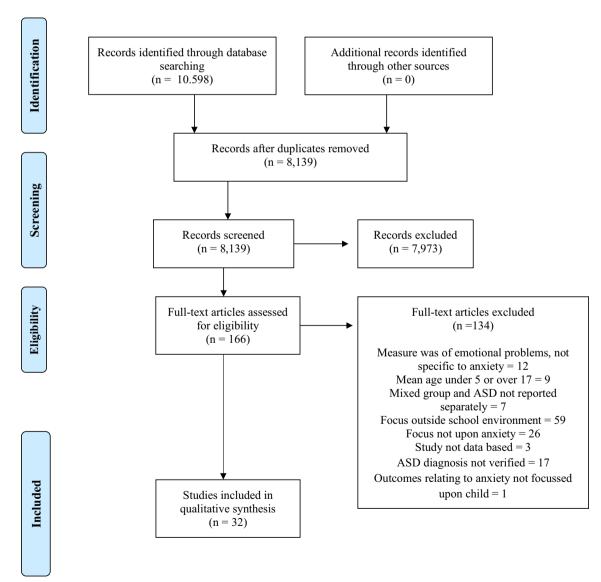


Fig. 1 PRISMA flow diagram

were identified, an ancestral search was conducted of reference lists to ensure no relevant research had been missed. No additional studies were found, leaving 32 studies that met the criteria for the current review.

## **Data Extraction and Synthesis**

Data were independently extracted by two authors and disagreements were resolved by both authors revisiting the article and discussing the information presented. There were no cases where a third reviewer was required to resolve disagreements. For each paper, information recorded included details of the research team (including geographical location of study), study design (intervention or non-experimental), sample characteristics (of both the autism sample and any control or comparison groups) and measure of anxiety (completed by teacher, parent and/or student). If the study described an intervention, the interventions used, location and numbers of sessions were also recorded.

To assess the extent to which the priori decision to exclude case studies and series may have impacted upon the results of the review, all case studies and series were identified and checked against the additional exclusion criteria. The systematic review had originally identified 171 case studies, all of which would be excluded for other reasons other than sample size (five were focussed outside of the school setting, four had participant ages outside 5–17 years, one was not a data-based study, 127 did not include a measure of anxiety or did not focus upon anxiety, 10 studies did not include participants with a diagnosis of autism, three studies were reporting on a participant who was not the child with autism and 21 were published before 1994). The systematic review also identified 15 case series, all of which would also have been excluded for reasons other than sample size (one had a focus outside of the school setting; five did not have a measure of, or focus upon, anxiety; three studies did not include participants with a diagnosis of autism; and six were not data-based studies). Therefore, the a priori decision to exclude case studies and series did not impact upon the studies meeting inclusion criteria for this review.

## **Assessment of Study Quality**

The studies were assessed to ensure they met a minimum quality standard using the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) checklist (von Elm et al. 2007) available at http://www.strobe-statement.org/fileadmin/Strobe/uploads/checklists/STROBE\_checklist\_v4\_cross-sectional.pdf). A global quality score was calculated for each article, giving 1 point for each STROBE item the article addressed. As per Fortin et al. (2012), articles had to have a quality score of at least 12 out of a possible 22 to be retained in the review. This cut-off was used as it indicates

that more than half of the STROBE quality markers were reported in the manuscript. None of the studies identified achieved a score below this cut-off.

#### Results

### **Included Papers**

Overall, 32 papers were included in the review. The included papers are summarised in Table 2. Of the 32 papers, 12 (37.5%) were from the same research group (Gadow et al. 2005, 2008a, b, 2009a, b, 2010, 2016; Guttmann-Steinmetz et al. 2010; Kaat et al. 2013; Lerner et al. 2017; Roohi et al. 2009; Weisbrot et al. 2005) with participants recruited through one university hospital developmental disabilities speciality clinic and assessed with similar standardised questionnaires. It is unknown to what extent the samples reported in these articles overlap, if at all. Therefore, where statistics described in this paper contain a large proportion of studies from the same research group, this will be highlighted.

## **Research Team and Study Design**

The research was conducted in seven countries, with the majority being conducted in the USA (62.5%; more than half of which were from the same research group), Singapore (12.5%) and the UK (9.4%). The most common disciplines of the lead author were psychiatry (42.4%) and psychology (33.3%), with only four (12.5%) studies describing school anxiety in children with autism being led by researchers within education departments.

The majority (81.2%) of studies were descriptive, non-experimental studies, with the remaining six being intervention studies. Three of the intervention studies were school-based cognitive behavioural therapy (Clarke et al. 2017; Drmic et al. 2017; Luxford et al. 2017), one was cognitive—behavioural therapy with some school-based sessions (Fujii et al. 2013), one was a clinic-based cognitive—behavioural therapy study (Ooi et al. 2014) and one explored an intervention to reduce listening-related stress and anxiety in the class-room (Rance et al. 2017).

All studies reported a quantitative component. The most frequent method of collecting anxiety data was through surveys (96.9%). Three studies used interviews and two of them reported the qualitative data in addition to questionnaires; Clarke et al. (2017) used interviews after the intervention to explore emotions and behaviours with parents and children on the autism spectrum; and Drmic et al. (2017) used a semi-structured interview to obtain feedback on the intervention. Fujii et al. (2013) used a standalone standardised diagnostic interview assessment to report categorical data on diagnostic status in relation to anxiety, pre- and post-intervention. No



Studies included in the review

Table 2

I, Q: SCAS-C Q: SCARED Q: BASC-C BASC-SRS Q: YSI Child Q: BASC-TRS Q: CCBRS ASEBA-TRF Q: CASI-4R Q: BASC-T Q: DBC-T Q: CSI-5 Q: CSI-5 Q: CSI-4 Q: CSI-4 Q: CSI-4 Q: CSI4 Q: CSI-4 **Feacher** DBC-T Q: ASI I, Q: SCARED Q: BASC-PRS I, Q: SCAS-P Q: CASI-4R Q: BASC-P Q: DBC-P Q: CSI-4 Q: CSI-5 Q: CSI-4 Q: CSI-4 Q: CSI-5 measure I: ADI-S Q: CSI-4 Q: CSI-4 Anxiety Q: ASI Parent mainstream) School (% Check 001 100 001 20 9 77 20 17 26 26 16 16 Previous study Population study Sampling schools technique Schools Schools Clinic No additional diag. No additional diag. Anxiety 100% Med: Psy 29% Med: Psy 39% Med: Psy 24% **ADHD** 100% Gifted 100% medication Additional diagnoses, Med 41% Med 45% Med 24% Med 38% Med 24% ID 0% %0 CI V: GARS, GADS verification (V) Asp 36% AD 29% PDD-NOS 7% DK 29% PDD-NOS 20% PDD-NOS 43% PDD-NOS 45% PDD-NOS 39% PDD-NOS 37% PDD-NOS 37% Diagnoses and V: ADOS, ADI V: ADOS, ADI V: ADI, ADOS PDD-NOS 9% AD 34%, Asp 27% Asp 38% V: Prac-M AD 40% Asp 23% V: Prac-M Asp 23% AD 37% Asp 43% AD 91% AD 19% AD 40% AD 17% Asp 38% V: SCQ V: MDA Asp V: Prac ASD ASD ASD ASD ASD ASD male design Study О Ω О Д О О О О О Ω Ω Ω Ω 100 100 86 95 82 98 85 71 86 85 84 87 87 87 93 81 ASD 8.7 (1.9) ASD + (mean, SD, range) ADHD 8.6 (1.9) 6–18 G1 6.8 (2.6) 4–14 G1 6.8 (2.6) 4–14 10.4 (3.3) 10.4 (3.0) 5–17 13.9 (2.2) 12–19 12.8 (0.8) 6.9 (2.6) 4–14 8.6 (1.9) 6–12 8.3 (1.8) 6–12 8.5 (1.9) 6–12 8.7 (1.8) 7-11 5-16.86 (1.1) 4–8 13-15 11 - 146 - 1010.7 Age  $N^1$  with ASD 28 238 88 67 2 67 14 277 4 54 221 204 301 Gadow et al. (2008a) Gadow et al. (2009a) Gadow et al. (2008b) Gadow et al. (2009b) Guttmann-Steinmetz Barnhill et al. (2000) Gadow et al. (2005) Gadow et al. (2016) Gadow et al. (2010) Clarke et al. (2017) Drmic et al. (2017) Foley Nicpon et al. Hoffman (2014) Fujii et al. (2013) Ashburner et al. et al. (2010) Hammond and Chandler et al. (2010) (2016)Study

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Table 2 (continued)											
Study	$N^1$ with ASD	Age (mean, SD, range)	% male	Study design	Diagnoses and verification (V)	Additional diagnoses,	Sampling technique	School (% mainstream)	Anxiety measure		
						IIICUICAUOII			Parent	Teacher	Child
					V: $n = 10$ Com., n = 4 ASDS						
Hass et al. (2012)	09	ı	87	О	ASD V: Ed	ID 0%	Schools	ı	ı	Q: BASC-TRS	
Hurtig et al. (2009)	43	13 (1.5) 11–17	74	D	Asp 51% HFA 49%	Receive psychiatric treatment 23%	Previous study	I	Q: ASEBA-CBCL	Q: ASEBA-TRF	Q: ASEBA-SR
Kaat et al. (2013)	115	8.5 (1.8) 6–12	98	О	AD 31% ASP 19% PDD-NOS 50%	ID 23% Med 26%	Clinic	25	Q: CASI-4R	Q: CASI-4R	ı
Kanne et al. (2009)	177	7.3 (3.9) 3–18	82	О	V: Frac-IM Asp 73% AD 27% V: SRS, ADOS	I	Clinic	ı	Q: ASEBA-CBCL	Q: ASEBA-TRF	ı
Lemer et al. (2017)	283	10.5 (3.3) 6-18	82	D	(87.76) ASD V: ADOS	Med: Psy 42%	Clinic	31	Q: CASI-4R	Q: CASI-4R	I
Luxford et al. (2017)	35	13.2 (1.1) 11–15	68	I	ASD V: Prac, SCQ	High anxiety 100% ID 0%	Schools	100	Q: SCAS-P	Q: SAS-TR Q: SWQ	Q: SCAS-C Q: SWQ
Matsushima et al. (2008)	36	PDD: 10.9 (3.1) PDD + ADHD 9.6 (2.4)	100	О	AD 89%, PDD-NOS 11% V: Prac-M	ID 0%	Clinic	I	Q: ASEBA-CBCL	Q: ASEBA-TRF	I
McDonald et al.	118	8.74 (1.32) 6–11	06	D	HFA V: ADI	Med: Psy 33%	Previous	18	Q: BASC	Q: BASC-	ı
Ooi et al. (2008)	9	11.5 (0.8) 9-13	DK	Ι	ASD or Asp	Anxiety 100%	Schools	0	Q: SCAS-P	Q: ACAS	Q: SCAS-C
Ooi et al. (2014)	98	9.8 (3.6) 4-18	62	D	ASD V: Prac	Med: Psy 47%	Clinic	ı	Q: ASEBA-CBCL	Q: ASEBA-TRF	ı
Pearson et al. (2012)	98	9.3 (1.9) 6–13	77	О	AD 58% PDD-NOS 23% Asp 19%	I	Schools Parent group	I	Q: CCBRS	Q: CCBRS	ı
Poon (2012)	322	9.5 (3.0)	71	D	V: ADI, ADOS ASD	ı	Schools	0	ı	Q: DBC-T	1
Rance et al. (2017)	10	Study B 14.9 (1.2)	100	I	v: Frac ASD V: Prac	Auditory dysfunction 100%	ı	100	ı	Q: ASEBA-TRF	
Roohi et al. (2009)	43	7.85 (2.4) 5-14	100	Ω	AD 37% Asp 30% PDD-NOS 40%	Med: Psy 30%	Clinic	35	Q: CSI-4	Q: CSI-4	ı
Ung et al. (2017)	32	7.5 (1.4) 6–10	81.3	D	V: Frac-M ASD V: ADOS, CARS,	ID 35%	Clinic	I	Q: ASEBA-CBCL	Q: ASEBA-TRF	ı
	301	8.3 (1.9)	84	D	AD 34%	Med 38%	Clinic	17	O: CSI-4	0: CSI-4	



<b>Table 2</b> (continued)	
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Study	$N^1$ with Age	% Study	Diagnoses and	Additional	Sampling	School (%	Anxiety		
	ASD (mean, SD, range)	male design		diagnoses, medication	technique	mainstream)	measure		
							Parent	Teacher	Child
Weisbrot et al.	6–12		Asp 27%						
(2002)			V: Prac-M						

Where multiple samples or subgroups are described, only the sample and results relevant to this review (e.g. age, diagnosis) are reported on here

<sup>2</sup> Dash (–) means this information was not stated in pape

intervention. Measures (Q, questionnaire; I, interview)—ACAS, Asian Children Anxiety Score—Caretaker version; ADI-S, Anxiety Disorder Interview ASEA-CBCL, Achenbach System of Empirically Based Assessment Child Behaviour Checklist (Parent) (Ch., Chinese version); ASEBA-TRF, Achenbach System of Empirically Based Child and Adolescent Symptom Inventory; CCBRS, Conners Comprehensive Behaviour Rating Scales; CSI, Child Symptom Inventory; SCAS, Spence Child Auxiety Inventory (P, parent; S, self-report); SAICA, Social Diagnoses—ASD, autism spectrum disorder; AD, autistic disorder; Asp, Asperger's; Com, community diagnostic reports; Ed., educational diagnosis; PDD-NOS, pervasive developmental disorder—not Verification—ASDS, Asperger Syndrome Diagnostic Scale; ADOS, Autism Diagnostic Observation Schedule; ADI, Autism Diagnostic Interview/Autism Diagnostic Interview-Revised; MDA, multidisciplinary assessment; Prac, practitioner or clinician diagnosis (sources of information may not be stated), Prac-M, multidisciplinary assessment; Prac, practitioner or clinician diagnosis (sources of information may not be stated). Social Responsiveness Scale. Medication—Med, prescribed medication (not specified); Psy, psychotropic medication **Feacher** Screen for Child Anxiety Related Disorders; SWO: Social Worries Questionnaire; Adolescent Symptom Inventory-Fourth Edition; BASC, Behavior Assessment System for Children (TRS, otherwise specified; HFASD, high functioning autism (spectrum disorder); DK, do not know. practitioner or clinician diagnosis with a range of sources of information; SRS, Adjustment Inventory for Children and Adolescents; SCARED, Assessment Teacher Report Form; ASI, Study design-

studies used qualitative approaches to explore or describe anxiety symptomatology in children with autism in school settings.

## **Samples**

Sample sizes ranged from 14 to 301 for descriptive and from 6 to 89 for intervention studies. The sample sizes were largest for the population-based study (N = 277, Chandler et al. 2016) and many of the clinic studies (e.g. N = 221 Gadow et al. 2016; N = 115 Kaat et al. 2013). Studies recruiting through schools tended to have sample sizes of 60 or fewer. The most common method of recruitment was via psychiatric clinic (56.2%; of which 37.5% was the same psychiatric clinic). Eight (25%) studies, half of which were intervention studies, recruited at least some of their participants directly through schools.

The average percentage of males across all of the included studies was 86.5%, which is an approximate ratio of 6:1 males:females. The samples were all male in five of the included studies (Clarke et al. 2017; Guttmann-Steinmetz et al. 2010; Matsushima et al. 2008, Rance et al. 2017; Roohi et al. 2009), and five studies (Fujii et al. 2013; Hurtig et al. 2009; Ooi et al. 2014; Pearson et al. 2012; Poon 2012) reported a ratio reflective of the current reported ratio of males: females in autism (3:1; see Loomes et al. 2017). Of the 29 studies that reported the participant age range, six (20.7%) focussed upon primary-school-aged children, six (20.7%) upon secondaryschool-aged children and 17 (58.6%) reported on children who were in both secondary and primary schools. The majority of studies included children from both mainstream and special educational settings. Three (9.1%) studies focussed solely upon children in special educational settings and five (18.2%) solely recruited children in mainstream settings.

Studies used a range of methods to confirm autism diagnoses. The most common method used to confirm diagnosis was multi-practitioner assessment which was used in 37.5% of studies (however, it must be acknowledged that this was the method used in 11 out of the 12 studies from Gadow's research team). Eight studies (25%) used the ADOS and/or ADI-R. Fifteen studies provided the proportion of autism diagnostic subtypes for the participants (eight of which were from the same clinic, so there is potential for participant overlap). Within these 15 studies, the average proportion of children with autistic disorder was 44.1%, with Asperger's 32.7% and with PDD-NOS 29.6%. Two studies reported a diagnosis of "high functioning ASD".

There was variability across papers in reporting additional or co-morbid diagnoses in the samples. Thirteen studies (40.6%) reported on specific additional diagnoses within the sample, with nine of these stating the level of intellectual disability in the sample and two stating that there were no additional diagnoses. Only three (9.4%) studies, all of which



were intervention studies (Fujii et al. 2013; Luxford et al. 2017; Ooi et al. 2008), provided details on whether any children in the samples had anxiety diagnoses. From the 32 included studies, 15 (45.4%) reported prescription medication use, 10 of which were from the same research team. However, little detail was given as to the types of medication. Two (Fujii et al. 2013; Luxford et al. 2017) of the six intervention studies reported on medication use in their sample.

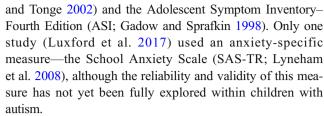
## Respondents

Twenty-nine (90.6%) of the 32 studies reported parent data and eight (25%) included a self-report measure of anxiety for the child with autism. Unsurprisingly, given that this review focusses upon anxiety in the school context, 30 studies (93.8%) reported on an anxiety measure completed by teachers. The two studies that did not report upon teacher data were intervention studies, one of which was based at school (Clarke et al. 2017) and one of which integrated sessions at school and collaborated with school providers (Fujii et al. 2013). Clarke et al. (2017) note that they decided not to include teacher ratings of anxiety in their study due to the inconsistencies between schools as to which teacher could fill in the questionnaire in a secondary school environment.

Twenty-seven (84.4%) studies reported on anxiety measures from multiple informants; 20 studies (62.5%, of which 12 were from the same research clinic) reported data from parents and teachers. None of the studies reported data from anxiety measures from teachers and children (without parents). Seven studies reported on anxiety measures completed by parents, teachers and children, although in two of these studies the measures used were not comparable between informants (e.g. in Drmic et al. [2017] parent and teachers completed the DBC but the child completed the SCARED, and in Luxford et al. (2017) parents and children completed the Spence Child Anxiety Scales and the teacher and child completed the Social Worries Questionnaire).

## Methods of Collecting Child Anxiety Data from Teachers

Eight different measures were used to document teacherreported anxiety within the school setting. All but one of the studies (97%) used an anxiety subscale within a broader measure of emotional/behavioural characteristics. Measures used were the Achenbach System of Empirically Based Assessment (ASEBA; Achenbach 2009), Behaviour Assessment System for Children (BASC; Reynolds and Kamphaus 1992), Child Symptom Inventory (CSI; Gadow and Sprafkin 2002), Child and Adolescent Symptom Inventory (CASI; Gadow and Sprafkin 2005), Conners Comprehensive Behaviour Rating Scales (CCBRS; Conners 2008), Developmental Behaviour Checklist (DBC; Einfeld



All papers except two (Guttmann-Steinmetz et al. 2010; Weisbrot et al. 2005) reported subscale mean scores or diagnostic status and did not provide detail on the profile of items that contributed to those scores. The 15 papers using the ASEBA, BASC, Conners and DBC only reported overall "anxiety" subscale scores (including the anxious/depressed subscale in the ASEBA and the anxious/shy subscale of the CCBRS). The teacher questionnaires of the CSI, CASI and ASI have generalised anxiety, social phobia, OCD and specific phobia subscales, although reporting of subscales varied across the 13 studies that used these measures.

Two papers reported item-level data. Guttmann-Steinmetz et al. (2010) compared ratings from mothers and teachers for children with autism and attention deficit hyperactivity disorder (ADHD) (n = 74) to children with autism (n = 130) and non-clinical controls (n = 170). Teacher ratings on the five generalised anxiety items were higher for the children with autism and ADHD and autism compared to controls. For three items (acts restless, irritable, tense), children with autism and ADHD scored significantly higher than those with autism alone. For three items (difficulty controlling worries, acts restless and tense), the mean scores for both the autism and autism with ADHD groups were between 1 and 2, suggesting that teachers identified these symptoms between sometimes (score of 1) and often (score of 2). In contrast, the mean scores for the controls ranged from 0.25 to 0.31 for these three items. The second paper to describe item-level data (Weisbrot et al. 2005) contrasted teacher ratings for children with a PDD diagnosis (n = 268) and those without (n = 167). They noted that children with PDD scored significantly higher on all items, with the mean scores for children with PDD on items in the social phobia subscale (avoids contact with strangers, excessively shy with peers and withdraws or freezes in uncomfortable situations) being approximately twice that reported for the controls. When comparing scores for children with Asperger's (n = 71), autistic disorder (n = 85) and PDD-NOS (n = 105), they noted significantly higher scores in children with Asperger's than children with autistic disorder on four items (overly concerned about abilities, difficulty controlling worries, tense or unable to relax and "distressing thoughts").

## Method of Describing and Analysing Child Anxiety Data from Teachers

Four studies (Ashburner et al. 2010; Hass et al. 2012; Poon 2012; Rance et al. 2017) only reported teacher data. Three of



these studies compared teacher ratings for autism with either typically developing children or children with multiple disabilities. Ashburner et al. (2010) reported significantly higher scores for children with autism on the CCBRS anxious/shy subscale (p < 0.001) and the ASRBA-TRF anxious—depressed subscale compared to typically developing children. Hass et al. (2012) reported that the mean anxiety scores were larger for both the child and adolescent autism sample than typically developing controls, with the mean score for the children with autism (but not adolescents) falling within the clinically significant range. Poon (2012) reports significantly higher scores on the DBC-T anxiety subscale in children with autism than in those with multiple disabilities (p < 0.001). The other study (Rance et al. 2017) reported teacher scores pre-/post-intervention.

Fifteen studies report both parent and teacher data, but do so independently of each other. Two of these were intervention studies focussing upon reducing anxiety; Drmic et al. (2017) who reported reductions post-intervention on the parent but not the teacher measure and Ooi et al. (2008) who reported no pre-post significant reductions on parent or teacher measures of anxiety. The majority of studies that reported parent and teacher separately had a focus on comparing profiles between subgroups, either comparing children with autism to another diagnostic group (Gadow et al. 2009a; Guttmann-Steinmetz et al. 2010; Ooi et al. 2014) mixed clinical controls (Weisbrot et al. 2005) or neurotypical control data (Hammond and Hoffman 2014), or comparing subgroups within autism based upon age (Foley Nicpon et al. 2010) or autism genetic markers (Roohi et al. 2009; Gadow et al. 2008b, 2009b, 2010). Two studies (Gadow et al. 2008a, 2016) explored the clinical correlates of psychiatric ratings in autism but only one (Gadow et al. 2016) goes on to discuss the unique and overlapping correlates between parents and teachers in detail in the discussion. Although not explored statistically, all except three (Gadow et al. 2010; Ooi et al. 2008, 2014) of these studies discuss the similarities and/or differences between parents and teacher ratings in the discussion, although to a varying degree.

Eight papers explored parent and teacher ratings for anxiety (some using multiple methods); six used correlations, three used tests of comparison and one compared agreement in clinical cut-offs. The intervention study by Luxford et al. (2017) correlated parent, teacher and self-report measures of anxiety at baseline, with only the significant correlation being between parent and self-report ratings on the SCAS (r=0.49, p<0.001). Their pre-/post-intervention comparisons were conducted separately for each respondent type. Correlations between parent and teacher ratings of anxiety were also conducted by Hurtig et al. (2009; CBCL: r=0.34, p= ns), Kanne et al. (2009; CBCL: r=0.14, p= ns), McDonald et al. (2016; BASC-2, r=0.51, p<0.001), Pearson et al. (2012; CCBRS: r=0.001, p<0.005) and Ung et al. (2017; CBCL: ICC = 0.41,

p= ns). Three papers (Barnhill et al. 2000; Gadow et al. 2005; Pearson et al. 2012) compared mean scores on the anxiety subscales between parents and teachers, both reporting either no significant differences between informants or only reporting effect sizes (which were below 0.2). One paper (Kaat et al. 2013) used kappa to report parent—teacher agreement on scores above the symptom and impairment clinical cut-offs for subscale level. Agreement for the generalised anxiety disorder subscale of the CASI-4R was 64–68% with kappa scores of 0.05–0.14 (considered "poor").

# **Teacher, Parent and Child Scores on Measures of Anxiety**

Table 3 summarises the 19 studies where data were collected and mean scores reported on comparable measures of anxiety across multiple respondents, for example, where both the ASEBA parent and teacher forms were used. Twelve of these studies had the same number of respondents in each respondent group (e.g. 14 parents, 14 teachers and 14 self-report), and of these, eight had equal numbers of parents and teachers. The remaining seven studies had a different number of respondents in each respondent group, for example, 277 parents and 228 teachers in Chandler et al. (2016) and 46 parents and 23 teachers in Hurtig et al. (2009), which has important implications when comparing similarities or differences between teacher and parent ratings of anxiety.

Table 3 summarises the mean (and standard deviation) of the teacher and parent scores on corresponding measures. Fifteen studies reported on comparable parent and teacher measures, although only eight of these had an equal number of parent and teacher respondents. On average, across all subscales reported in the 15 studies with parent and teacher data, teacher scores were within 10.2% of the parent scores. If this is restricted to the studies with equal numbers of parents and teachers, teacher scores were, on average, within 12.8% of parent scores. Studies varied as to whether the teacher or parent reported higher scores (representing more symptoms of anxiety).

A mixed profile of scores is noted across the four studies that report on comparable child, parent and teacher data. Barnhill et al. (2000) reported data from an equal number (N=20) of children, parents and teachers on the BASC, noting relatively equal scores between parents and teachers (59.6 and 60.1 respectively) and lower scores on the self-report completed by the child with autism (47.2). A similar pattern was noted by Hammond and Hoffman (2014) using the ASI, but a contrasting pattern (with the highest score being reported by the individual with autism) was reported by Hurtig et al. (2009). Foley Nicpon et al. (2010) reported relatively equal scores across parents (56.4, N=52), teachers (56, N=55) and children with autism (56, N=45).

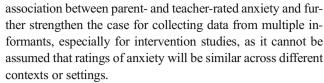


#### Discussion

The purpose of this review was to identify and review the research that has focussed upon anxiety in children with autism within the school setting. The findings indicate that, while there is a growing research base on anxiety in children with autism, the research relating to anxiety in autism within an academic setting remains very limited, with only 32 published studies meeting inclusion criteria (12 of which were conducted by the same research group). This lack of research is concerning given the combination of the elevated prevalence of anxiety in autism and the levels of academic underachievement reported in students on the spectrum.

Despite the limited number of studies, several important findings emerged through conducting this review. Firstly, that the research to date has documented teacher-reported levels of anxiety in children with autism through mean scores on a range of measures, with the consistent finding that teachers report higher mean scores on anxiety subscales for children with autism than children without autism. However, the research has not yet described the phenomenology of the symptomatology within the classroom setting. It cannot be assumed that the symptomatology will present the same as that in children without autism (e.g. see Kerns et al. 2015) nor that it will be the same as that seen in by children with autism within the home setting given the difference in both cognitive and social demands of home and school settings (Allen and Lerman 2018) and the recent work suggesting that parents of children with autism report different anxiety symptomatology between home and community settings (Adams et al. 2018b). There may also be issues of diagnostic-overshadowing influencing teacher's interpretation and responses to anxiety-related behaviours in students with autism (Adams et al. 2019b). There is therefore a critical need for more research to be undertaken to further understand anxiety symptomatology in children with autism in the school setting so as to allow educators to identify the presence of anxiety in children with autism, assess its impact on learning and decide whether to implement appropriate educational support strategies and/or refer on for intervention.

Secondly, the use of teacher data across the studies has not provided a consistent outcome in terms of agreement or differences from parent-reported levels of anxiety. Of the six papers that correlated parent and teacher data, half reported a positive correlation and half reported no correlation. Of the three that used a test of difference, none reported any significant difference between parent and teacher scores but the one study exploring agreement between clinical cut-offs reports "poor" agreement. Changes in parent and teacher scores from pre- to post-intervention were also inconsistent, with one study reporting no changes in either informant and one study reporting changes in parent scores but not teacher scores. Together these results highlight the possible complex



Finally, despite aiming to conduct research relating to anxiety, researchers have tended to report on scores collected on anxiety subscales of broad emotional and/or behavioural measures, some of which have combined anxiety with depression (CBCL) or shyness (CCBRS). Of the 30 included studies that gathered information from teachers on student anxiety, only one study (Luxford et al. 2017) used an anxiety-specific questionnaire for teachers, the School Anxiety Scale (SAS-TR; Lyneham et al. 2008). This measure has had limited assessment of its validity in typically developing children and has only been used in one other study of children with autism (which did not formally assess its validity; Adams et al. 2018a). Differentiating anxiety symptoms from autism characteristics can be challenging, and as a result, there are concerns that relying on measures standardised on typically developing children may not accurately represent the way anxiety manifests in children with autism (Ozsivadjian et al. 2012) due to possible typical and atypical signs of anxiety in children with autism (Kerns et al. 2015). An international working group of autism experts has identified the issue of anxiety measurement (including validation of existing measures to address the overlap between autism and anxiety symptoms and the further development of objective measures of anxiety) as a top research priority (Vasa et al. 2018). Such measures (e.g. the Anxiety Scale for Children with Autism Spectrum Disorder; Rodgers et al. 2016) are showing good reliability and validity in school-aged children with autism (e.g. den Houting et al. 2018a, b). The findings of this review support the conclusions of the working group, highlighting a clear need for either further validation of the SAS-TR for children with autism, or the development of an autismspecific measure of anxiety in the academic setting which aims to capture the full range of anxiety presentations seen in autism.

### **Methodological Issues of Available Research**

There were a number of methodological issues across a number of the studies identified which should be carefully considered when future research into anxiety and autism in the school setting is planned. Few studies reported on equal sample sizes between teacher and parent questionnaires, limiting the extent to which the scores (e.g. in Table 3) can be truly compared, as the samples are not "like for like". There was notable variability in the depth of the cohort descriptions and in the representativeness of the cohort to the broader autism population. Given the included studies were reporting on anxiety (even as part of a broader emotional or behavioural



Table 3 Studies reporting means and standard deviations on comparable multi-informant measures of anxiety

Study	Anxiety subscale score				
	Measure	Parent	Teacher	Child	
Barnhill et al. (2000)	BASC	N=20	N = 20	N=20	
Chandler et al. (2016)	DBC	59.6 (13.4) <i>N</i> = 277 7.8 (4.2)	60.1 (7.91) $N = 228$ $3.4 (2.6)$	47.2 (10.2) -	
Clarke et al. (2017)	SCAS	N=14 Pre 34.04 (12.2) Post 27.9 (9.6)	-	N= 14 Pre 33.9 (11.4) Post 33.9 (6.4)	
Drmic et al. (2017)	SCARED	N=35 Pre 23.1 (11.8)	-	N=35 Pre 26.7 (13.3)	
Foley Nicpon et al. (2010)	BASC	Post 19.5 (9.6) N = 52 56.4 (13.3)	<i>N</i> = 55 56.0 (15.0)	Post 19.6 (11.3) N=45 56.0 (15.0)	
Gadow et al. (2008a)	CSI	N = 238 GAD 5.1 (3.8)	N = 238 GAD 4.5 (3.3)	-	
Gadow et al. (2005)	CSI	SpPhob 1.0 (1.1) N = 284 GAD 4.7 (3.7)	SpPhob 0.5 (0.8) N = 284 GAD 4.1 (3.2)	-	
Gadow et al. (2009a)	CSI	SpPhob 1.0 (0.9) N = 87 GAD 5.2 (3.7) SePhob 0.0 (0.0)	SpPhob 0.5 (0.8) N = 79 GAD 5.3 (3.5) SpPhob 0.5 (0.8)	_	
Gadow et al. (2009b)	CSI	SpPhob 0.9 (0.9) N = 67 GAD 2.8 (2.7)	SpPhob 0.5 (0.8) N = 67 GAD 2.7 (2.3)	_	
Gadow et al. (2010)	CSI	N=67 GAD 2.8 (2.7)	N = 67 GAD 2.7 (2.3)	_	
Hammond and Hoffman (2014)	ASI/YSI	N=14 GAD 73.9 (6.9) Sep/panic 67.1 (9.2)	N = 7 GAD 75.1 (3.2)	N=14 GAD 55.1 (6.4) Sep/panic 65.4 (12.6)	
Hurtig et al. (2009)	ASEBA	N = 46 Anx/dep 5.2 (4.3)	N = 23 Anx/dep 5.8 (5.2)	N = 46 Anx/dep 6.5 (6.3)	
Luxford et al. (2017)	SCAS; SWQ	N = 18 Pre N = 15 Post SCAS: Pre 47.6 (16.3) Post (2), 31.9 (14.9)	N=18 SWQ: Pre 11.3 (3.6) Post (2), 8.0 (4.4)	N=18 SCAS: Pre 40.5 (16.9) Post (2), 27.5 (14.7) SWQ: Pre 12.3 (4.7) Post (2), 8.8 (4.4)	
Matsushima et al. (2008)	ASEBA T score	N = 36 Anx/dep 65.0 (9.2)	N = 34 Anx/dep 63.5 (8.7)	_	
McDonald et al. (2016)	BASC	N=118 55.43 (12.59)	N=118 57.57 (12.49)	_	
Ooi et al. (2008)	SCAS	Intervention $N = 6$ Pre 20.1 (6.9) Post 22.0 (9.1)	-	Intervention $N = 6$ Pre 40.6 (20.8) Post 33.3 (20.3)	
Ooi et al. (2014)	ASEBA	N = 86 Anx/dep 5.3 (4.4)	N = 39 Anx/dep 3.9 (3.7)	_	
Pearson et al. (2012)	CCBRS	N = 86 Anxious/shy $62.5 (14.8)$	N = 86 Anxious/shy $65.7 (13.8)$	_	
Ung et al. (2017)	ASEBA	N = 32 Anx/dep 6.8 (5.2)	N = 32 Anx/dep 7.3 (5.7)	_	

Measures—ASEBA, Achenbach System of Empirically Based Assessment; ASI/YSI, Adolescent/Youth Symptom Inventory; BASC, Behavior Assessment System for Children; CCBRS, Conners Comprehensive Behaviour Rating Scales; CSI, Child Symptom Inventory; DBC, Developmental Behaviour Checklist; SCAS, Spence Child Anxiety Inventory (P, parent; S, self-report); SWQ, Social Worries Questionnaire. Subscales—Anx/dep, anxious/depressed; GAD, general anxiety; Sep/panic, separation panic; SpPhob, specific phobia

profile), it was surprising that only 15 included data on medication and 11 on additional diagnoses (including those reporting on the presence of intellectual disability). It was also surprising that the educational setting (mainstream or special

school) was not described, given that the studies had collected data from teachers and/or that the interventions had taken place within the school setting. There are differences in anxiety levels for children with autism with and without an



intellectual disability (van Steensel and Heeman 2017), so combining populations from these settings may mean that specific patterns are being missed. Females and children attending special schools were generally underrepresented in the available literature as were participants recruited outside of psychiatric clinics. Future studies should provide more comprehensive descriptions of such important factors when describing their samples and consider the profile of participants compared to the broader autism population in schools.

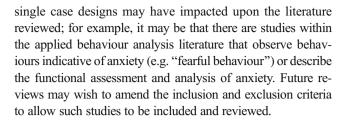
# Barriers to Autism and Anxiety Research in Academic Settings

There may be many possible reasons for the lack of research in this area. The absence of anxiety measures that can be completed by teachers, who are arguably the best placed to observe and report on external indicators of anxiety in their students, may mean that researchers are not able to report on specific symptomatology in the school setting. Another possible reason is the barriers that can be encountered when undertaking research in schools (Kasari and Smith 2013). For example, Clarke et al. (2017) noted that they intended to gain teacher anxiety ratings for participants but decided against this due to difficulties finding consistency between schools in terms of which teachers would complete the questionnaires. Other studies (e.g. Adams et al. 2018a) report teacher questionnaire completion rates of less than 50%, even with a fulltime project manager focussing upon school liaison. To further our understanding of anxiety at school and to develop effective interventions, it will require researchers to address some of these challenges.

#### **Areas for Further Research**

Further research is needed that incorporates well-validated anxiety-specific measures that can be completed by multiple informants and are suitable for use with children on the autism spectrum. Due to the possibility of differing presentations and the increased reliance on proxy report (Adams and Oliver 2011), there is also a recognised need to develop specific measures of anxiety (alongside other aspects of mental health) in individuals with co-morbid intellectual disabilities (see reviews by Flynn et al. 2017). In addition, future work needs to go beyond reporting total and subscale scores if our understanding of anxiety in autism is to be enriched, using qualitative approaches and/or item-level analyses to provide rich information about the variability in anxiety presentation in autism across home and school settings (Magiati et al. 2017).

Future reviews may wish to include search terms for specific disorders including "social phobia" and "obsessive compulsive disorder" or specific treatments (e.g. "exposure therapy") to explore the extant research on specific disorders and treatments within the school setting. The exclusion of studies based upon



#### **Conclusions**

There is limited research describing anxiety in autism in the school context and the research that is available tends to report on non-anxiety-specific measures and to report a numerical score rather than any detail on presentation or symptomatology. More research must be conducted into the way that anxiety in children with autism presents at school and what, if any, impact it has on their academic engagement and achievement. Further development of teacher- and self-report methods is also an important focus for future research with work to establish the methodological quality of such measures.

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#### **Compliance with Ethical Standards**

**Conflict of Interest** The authors declare that they have no conflict of interest.

**Ethical Approval** This article does not contain any studies with human participants performed by any of the authors.

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