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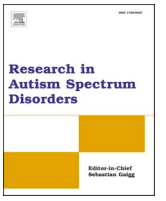

The relationships between restrictive/repetitive behaviours, intolerance of uncertainty, and anxiety in autism: A systematic review and meta-analysis

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ABSTRACT

Autistic people are more likely to experience anxiety than their non-autistic peers. Understanding mechanisms underpinning anxiety in autism is a vital aspect of developing effective interventions. Intolerance of uncertainty (IU) and restrictive/repetitive behaviours (RRBs) are proposed to contribute to anxiety for autistic people. This paper includes the first meta-analysis to investigate the associations between all three of these variables. A systematic search identified 33 papers that measured anxiety, IU and RRBs in 8347 autistic participants. Evidence was found for positive correlations between all three variables. Analysis of average participant age demonstrated that the relationship between anxiety and IU was stronger in younger participants. No significant differences were found between the associations in studies that included participants with intellectual disabilities and those that did not. A quality assessment framework identified methodological threats to validity. Most studies had good methods of recruitment; however, many anxiety and IU measurement tools were unvalidated in autistic populations. Results suggest that IU and RRBs should be considered when designing anxiety interventions for autistic people, however, the role of RRBs in particular needs to be investigated further to prevent interventions from taking away important coping strategies due to misunderstanding of causal relationships.

Autistic people are much more likely to experience anxiety than their non-autistic counterparts (Vasa & Mazurek, 2015). An enriched understanding of the contributors to and factors associated with anxiety in autistic people will allow us to provide better interventions for anxiety, particularly as the causes and symptoms of anxiety are often different in autistic people compared to non-autistic people (Kerns, Kendall, Berry et al., 2014). Autistic people report higher rates of Intolerance of Uncertainty (IU) (Jenkinson et al., 2020). Autistic people are also more likely to engage in restrictive and repetitive behaviours (RRBs) .[1] South and Rodgers (2017) presented a model that highlighted the importance of IU and RRBs for explaining anxiety in autistic people. South and Rodgers (2017) proposed that whilst factors such as atypical sensory function, rigidity of thought and emotional awareness are predictive of

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[1] We are conscious that the term 'restricted and repetitive behaviours' is not preferred by some of the autistic community. Due to the extensive use of this term in the literature, this term will be used in this review to describe the variable being investigated. When such terminology is used, it is to reflect conclusions drawn/analyses made by referenced studies, and not the view of the authors.

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increased anxiety, it is RRBs and IU that are supported as direct precipitants of anxiety. South and Rodgers's (2017) model proposes that IU can result in both anxiety and RRBs, which in turn influence each other bidirectionally. The current meta-analysis tests the evidence for this.

RRBs are patterns of stereotypic and/or ritualistic behaviours, often observed in autistic people (Lewis & Kim, 2009). Factor analysis of the Autism Diagnostic Interview-Revised (ADI-R) indicates two subtypes of RRBs: 'repetitive motor behaviours', such as hand flapping, and 'insistence on sameness' behaviours, such as specialised interests or rituals (Lidstone et al., 2014). Other measures of RRBs also refer to subtypes such as rigidity/adherence to routine, circumscribed interests, self-injurious behaviours, compulsive behaviours, and unusual sensory interests (Bodfish et al., 2000; Barrett et al., 2015; Leekam et al., 2007; Turner, 1996). A hypothesised link between RRBs and anxiety has been proposed since the earliest descriptions of autistic behaviour, with Kanner (1943) proposing that rigid and restricted behaviours in autism may be anxiety driven. Theoretical accounts often describe RRBs as a means of alleviating anxiety (Leekam et al., 2011) and there are multiple empirical studies that support this association in both autistic children and adults (e.g. Factor et al., 2017; Moore et al., 2021). Research into the physiological effects of RRBs also indicate that they may serve to calm the nervous system and therefore reduce anxiety (Joosten et al., 2008). A synthesis of such evidence is currently lacking, however. It is worth noting that qualitative evidence points towards RRBs having functions other than reducing anxiety - some autistic people may take pleasure in engagement with RRBs or use them as sensory-seeking behaviours (Collis et al., 2022).

IU is characterised by a tendency to react negatively towards ambiguous situations and an intense preference for predictability (Jenkinson et al., 2020). It is predominantly associated with generalized anxiety disorder (Dugas et al., 1998), but has also been associated with panic disorder, social anxiety disorder, obsessive compulsive disorder (OCD), depression, and eating disorders (McEvoy et al., 2019) in neurotypical samples. IU has been implicated in anxiety in non-autistic children (Osmanağaoğlu et al., 2018) and adults (Norr et al., 2013), as well as autistic children (Boulter et al., 2013) and adults (Maisel et al., 2016). Jenkinson et al. (2020) conducted a systematic review and meta-analysis, concluding that individual differences in anxiety and IU are positively associated in autism ($r = 0.62$). Following from this, and given that a number of evidence-based interventions exist that directly target IU (e.g., Rodgers et al., 2022), IU may be an ideal target for anxiety intervention in autism.

A recent thematic analysis by Goodwin et al. (2022) suggested that autistic children often engage in RRBs (both repetitive motor behaviours and insistence on sameness) in response to uncertainty. There is limited quantitative research to support this relationship, however several studies do show positive associations between RRBs and IU (e.g., Hwang et al., 2020), suggesting that RRBs may serve to help autistic people cope with change and uncertainty by enforcing predictability (Lau et al., 2020). An improved understanding of the functions and consequences of RRBs in autism may reinforce a shift in societal attitude towards them – RRBs have historically been viewed as disruptive and non-functional, but such research described in this review demonstrates that they may, when engaged with safely, be an effective coping mechanism for managing anxiety and IU.

The strength of the relationship between IU and anxiety was examined in a systematic review and meta-analysis by Jenkinson et al. in 2020. However, at the time of the search (March 2019), only a small number of studies ($k = 10$) had been published. The recent growth in this area means a substantial increase in the past four years. This review can now provide a more contemporary estimate of that effect. Furthermore, South and Rodgers (2017) proposed that RRBs also have a role to play in the IU/anxiety relationship. There have been suggestions that some RRBs may serve to limit uncertainty and therefore reduce anxiety (Moore et al., 2021), however some suggests that behavioural rigidity may increase uncertainty, and so anxiety, about whether exact expectations will be met (South & Rodgers, 2017). This research remains unsynthesised. Understanding how RRBs are associated with the relationship between IU and anxiety may be fundamental in reducing anxiety. The aim of this review is therefore to examine the literature that investigates the associations between these three constructs, to evidence the proposed model presented by South and Rodgers (2017), as well as to build on it. Three meta-analyses will be conducted to answer the questions:

- What is the strength of the association between anxiety and RRBs in autistic people?
- What is the strength of the association between IU and anxiety in autistic people?
- What is the strength of the association between RRBs and IU in autistic people?

Potential moderating variables will also be considered in this review. Approximately 60–70 % of autistic people also have an intellectual disability (Foundation for People with Learning Disabilities, n.d.). Some evidence suggests that IQ moderates anxiety levels in autistic people (van Steensel et al., 2011), though cf. Mingins et al. (2021), on whether this truly reflects lower anxiety levels. Therefore the effects of inclusion of participants with an intellectual disability on each of the above relationships will be considered. Average age of participants was also included as a potential moderator, due to the idea that the cognitive mechanisms needed for processing uncertainty develop with age (Osmanağaoğlu et al., 2018). Upon suggestions from reviewers, emergent moderators were also included. Some evidence suggests that autistic females experience higher levels of anxiety (Uljarevic et al., 2020), and the proportion of females in neurotypical samples has been significantly associated with stronger relationships between IU and anxiety symptoms (McEvoy et al., 2019) – therefore gender was added as a potential moderator. As the role of IU in the anxiety of autistic people has received accelerated attention in recent years, many of the included studies have been conducted by the same author-s/collaborators; the effect of research group was therefore also analysed. The effect sizes between studies that did and did not use autism-specific measures, as well as the impact of study quality on the analyses, were also added – as a means to understand the impact of methodological choices on estimates of the effect.

Table 1
Search Terms.

Search 1:	(autis* **OR** asperger* **OR** ASD **OR** ASC **OR** "autis* spectrum disorder*" **OR** "auti* spectrum condition*") **AND** (anx*) **AND** (repetitive **OR** restrictive **OR** RRB)
Search 2:	(autis* **OR** asperger* **OR** ASD **OR** ASC **OR** "autis* spectrum disorder*" **OR** "auti* spectrum condition*") **AND** ("intolerance of uncertainty" **OR** IU)

Table 2
Inclusion/Exclusion Criteria.

Inclusion Criteria	Exclusion Criteria
a. Population: Reports data from a population of autistic people. b. Measures: Uses standardised measures for at least 2/3 required variables (anxiety/IU/RRBs). c. Data: Reports a correlation analysis between at least 2/3 required variables. d. Language: Paper available in English.	a. Reviews: No original data reported. b. Data: Only partial correlations reported.

1. Method

This review was registered on Prospero (registration number CRD42021295937). Methods and results are reported in line with Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) where appropriate. A full PRISMA checklist is included in the supplementary materials.

1.1. Search Strategy

A systematic search of the literature was conducted in January 2023 via Web of Science, PsychINFO, Medline, and ProQuest Dissertations and Theses databases. Two sets of search terms were adapted from Jenkinson et al. (2020) and McFayden et al. (2020), to maximise the return of relevant papers. Full search terms can be found in Table 1. Preliminary searches combining all three sets of search terms (for RRBs, anxiety, and IU) returned limited papers (particularly those looking at IU). To identify a broader range of papers, two separate searches were conducted. Search 1 identified papers that examined the relationship between anxiety and RRBs in

Table 3
Quality Assessment Framework.

Category	Item	Poor (0)	Satisfactory (1)	Good (2)	Excellent (3)
Sample	Sample Identification	Unspecified	Single restricted or non-random sample,(e.g., a specialist clinic or previous research study). Single regional sample, (e.g., a regional parent support group).	Multiple restricted or non-random samples, (e.g., multi-region specialist clinics, multiple schools). National non-random sampling, (e.g., national parent support groups).	Random sample
	Size of Sample	< 28	28–87	88–849	850 +
	Confirmation of Autism Diagnosis	Unspecified	Recruitment from group known to have a higher likelihood of autism (siblings or parents of autistic people). Self/parent/teacher report. Recruitment from specialist school or support group.High score on autism screening questionnaires such as Autism Spectrum Quotient (AQ).	Best-estimate diagnosis by a clinician.High score on one or more validated measures of autism, (e.g., ADOS, ADI-R). Previous diagnosis of autism by multidisciplinary team using multiple assessment methods unconfirmed inthe present study OR confirmed inthe present study using only ONE assessment method or measure.	Clinical diagnosis confirmed by a multidisciplinary team using DSM-IV, DSM-V or ICD-10 criteria and multiple, well-validated assessment tools (e.g., ADOS, ADI-R). Diagnosis must be confirmed in this study – not in previous studies or as part of their assessment through a clinic – and using two or more assessment methods and a multidisciplinary team (i.e., at least two professionals must have come to this verdict, can be speech and language therapist/OT/ psychiatrists/ psychologists etc.)
Measures	Measurement of Anxiety	Unspecified	Self/parent report using a well-validated measure, but not validated for autistic individuals.	Self/parent report using a well-validated measure with adaptations to make them somewhat appropriate for autistic individuals.	Formal measure validated in autistic individuals.
	Measurement of RRBs	Unspecified	Self/parent report using a well-validated measure, but not validated for autistic individuals.	Self/parent report using a well-validated measure with adaptations to make them somewhat appropriate for autistic individuals.	Formal measure validated in autistic individuals.
	Measurement of IU	Unspecified	Self/parent report using a well-validated measure, but not validated for autistic individuals.	Self/parent report using a well-validated measure with adaptations to make them somewhat appropriate for autistic individuals.	Formal measure validated in autistic individuals.

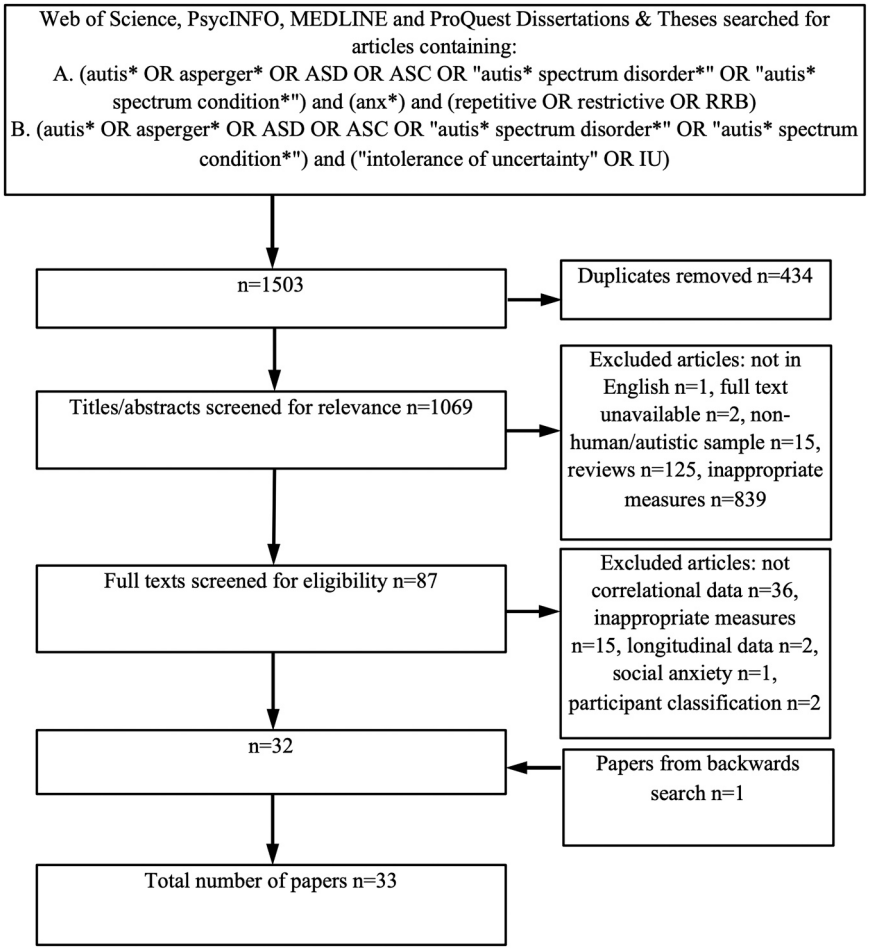

Fig. 1. Preferred Reporting Items for Systematic Reviews and Meta-Analyses Diagram Reflecting Full Search and Selection Strategy.

autistic people. Search 2 identified papers investigating IU in autistic people and thus was expected to deliver papers looking at the relationships between IU and RRBs, *and* between IU and anxiety. The reference sections of all selected papers, as well as two relevant review papers (Jenkinson et al., 2020; South and Rodgers, 2017), were also screened for any additional papers not found in the original search.

1.2. Study selection

Paper selection was completed by the first author, as per the PRISMA guidelines. After duplicates were removed, titles and abstracts were reviewed for all search results, with studies being excluded if any initial exclusion criteria were clearly met. Full papers were then screened and excluded if they met any of the exclusion criteria. Full inclusion/exclusion criteria are described in Table 2. Papers from the backwards literature search were screened in the same manner. A second researcher independently screened 20 % of the original search results at title/abstract and then full paper level, to check for validity of inclusion and exclusion of papers. Researchers agreed on inclusion in all cases.

1.3. Data extraction

Data were extracted from the included papers about sample sizes and participant demographics, as well as the measurement tools used in the study and results of uncontrolled correlational analyses (i.e. those that did not control for additional variables). Where both parent- and child-report measures were used for child participants, only child-report values were taken for measures of IU, and only parent-report values were taken for anxiety, as research suggests that autistic children with co-occurring anxiety tend to underreport overall levels of anxiety (Kalvin et al., 2020). To our knowledge, there is no research to suggest this occurs for IU, so child-report values were taken. No studies included both parent- and child-report values for RRBs. Where values for RRBs were split into sub-categories (e.g. insistence on sameness and repetitive motor behaviours), values were combined to generate a total RRB value. A second researcher independently repeated the data extraction process for all papers to check for data quality, with no differences in data identified.

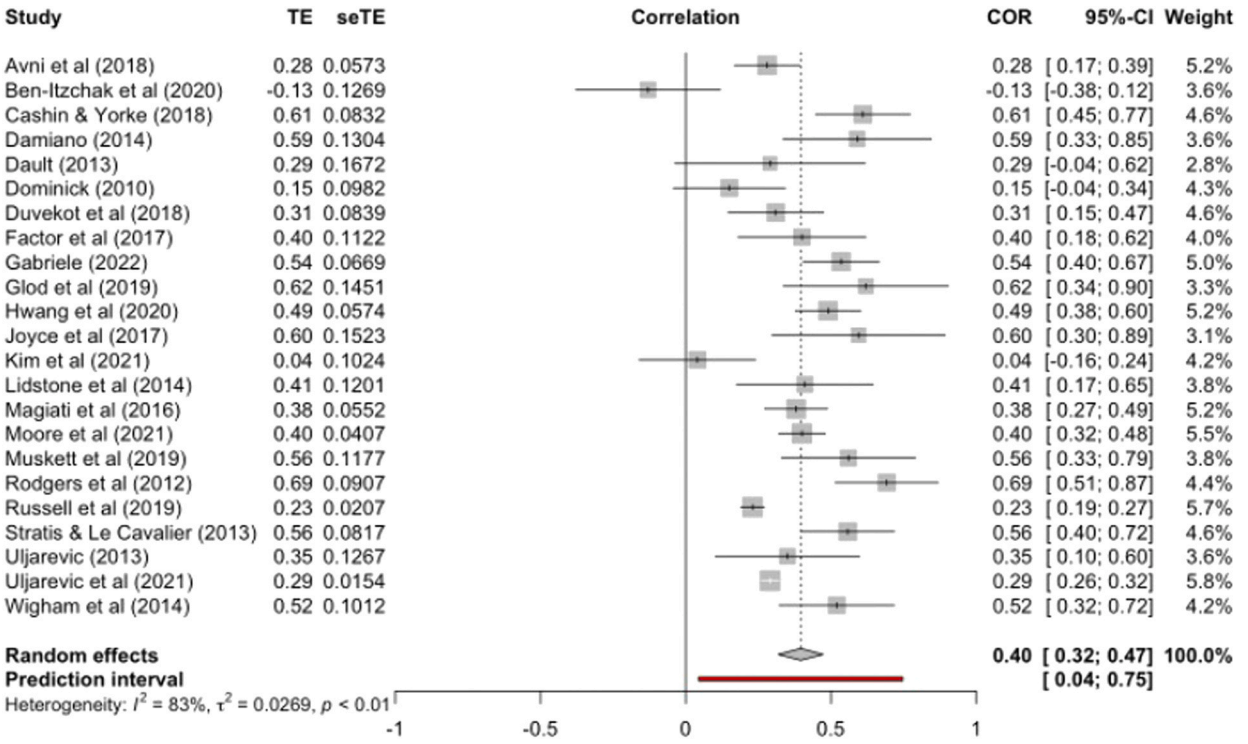

Fig. 2. Forest Plot for the Random Effects Model of the Meta-Analysis Between Anxiety and Restrictive/Repetitive Behaviours (Meta-Analysis 1).

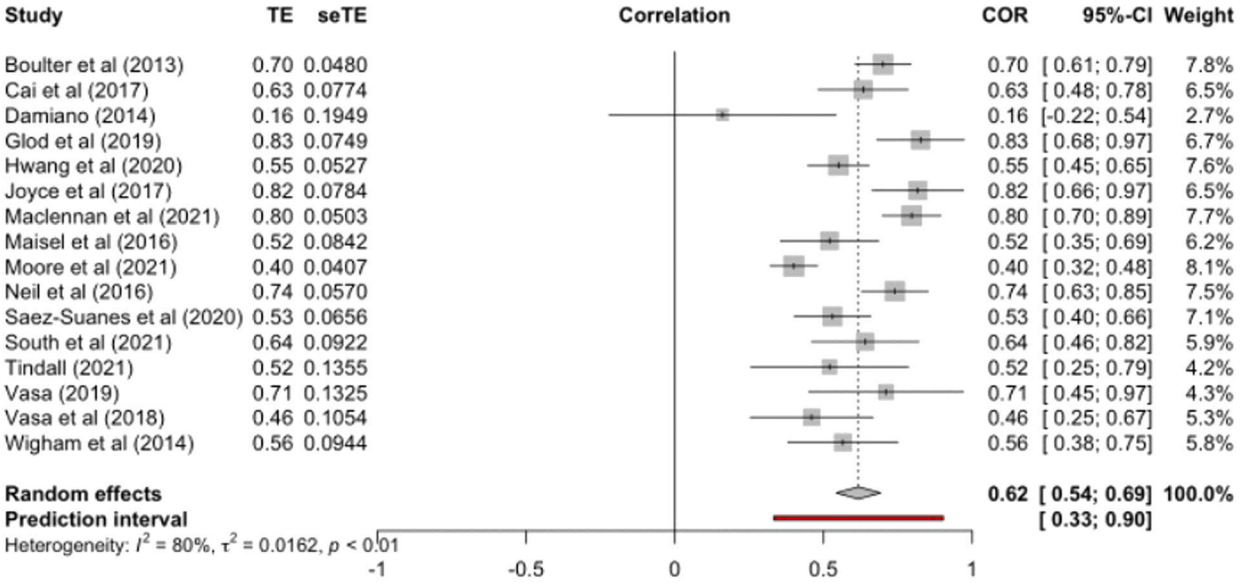

Fig. 3. Forest Plot for the Random Effects Model of the Meta-Analysis Between Intolerance of Uncertainty and Anxiety (Meta-Analysis 2).

1.4. Quality review

A quality assessment framework (Table 3) was developed, based on Mingins et al. (2021). The framework was created to review aspects of each study related to this specific meta-analysis and its research questions, therefore does not necessarily reflect the quality of the paper in meeting the authors' own objectives. Each paper was given a score (0−3) based on six factors, with the mean score being taken as the overall quality score. Developing a novel framework was chosen over using an established one, to reflect the particular demands and limitations of the literature better.

Table 4
Full Data Extraction.

Author (year)	N	Mean Age (years)	Anxiety Measure	RRB Measure	IU Measure	Anx/RRB Correlation	RRB/IU Correlation	IU/Anx Correlation	ID Included?	% Male Ppts	Autism-Specific Measure - Anxiety	Autism-Specific Measure - RRBs	NE England Researcher Group
Papers investigating all three relationships													
Damiano (2014)	26	14.08	SCARED	RBS-R	IUS-C	0.59 * *	0.16	0.51 * *	No	92.3	No	Yes	No
Glod et al. (2019)	19	7.06	SCAS-P/ PAS	RBQ	ASC-ASD	0.62^{+}	0.826 * *	0.50^{+}	Unclear	84.21	No	No	Yes
Hwang et al. (2020)	176	41.7	SMGAD	RBQ-2A	IUS-12	0.49 * *	0.55 * *	0.50 * *	Yes	39.9	No	No	No
Joyce et al. (2017)	19	16.81	SCAS-P	RBQ-2A	IUS-C	0.595 *	0.817 * *	0.45^{+}	No	84.21	No	No	Yes
Moore et al. (2021)	426	42.78	HADS	RBQ-2A	IUS-12	0.40^{+}	0.50 * *	0.40^{+}	Unclear	44/83	No	No	Yes
Wigham et al. (2014)	53	12.49	SCAS-P	RBQ	IUS-P	0.52^{+}	0.565 * *	0.43^{+}	No	88.7	No	No	Yes
Papers investigating Anxiety and RRB													
Avni et al. (2018)	260	7.58	CPRS-R:L	ADI-R	-	0.28 * *	-	-	Unclear	87.7	No	Yes	No
Ben-Itzchak et al. (2020)	61	13.73	SCARED	ADOS	-	-0.13	-	-	Yes	92.3	No	Yes	No
Cashin and Yorke (2018)	58	11.66	SCAS-P	RBQ	-	0.61 * *	-	-	Yes	86.0	No	No	No
Dault (2013)	31	Not Reported	SCAS	RBS-R	-	0.29	-	-	Unclear	Not Reported	No	Yes	No
Dominick (2010)	100	12.4	BAI	RBS-R	-	0.15	-	-	Yes	86.0	No	Yes	No
Duvekot et al. (2018)	117	6.7	CBCL	SRS	-	0.31 * *	-	-	Yes	80.5	No	Yes	No
Factor et al. (2017)	57	7.25	CBCL	SRS-2	-	0.40 * *	-	-	Unclear	82.46	No	Yes	No
Gabriele (2022)	115	10.1	PRAS-ASD	RBS-R	-	0.535 * *	-	-	Yes	80.0	Yes	Yes	No
Kim et al. (2021)	96	14.3	CBCL	ADI-R	-	0.04	-	-	Yes	94.8	No	Yes	No
Lidstone et al. (2014)	49	10.58	SCAS-P/ PAS	RBQ-2	-	0.41^{+}	-	-	Unclear	91.9	No	No	Yes
Magiati et al. (2021)	241	10.33	SCAS-P	DBC-P	-	0.38 * *	-	-	Unclear	81.7	No	No	No
Muskett et al. (2019)	35	7.6	CBCL	RBS-R	-	0.56 * *	-	-	Yes	82.9	No	Yes	No
Rodgers et al. (2012)	34	12.17	SCAS-P	RBQ	-	0.692 * *	-	-	Unclear	85.3	No	No	Yes
Russell et al. (2019)	2093	10.29	CBCL	RBS-R	-	0.23^{+}	-	-	Yes	86.6	No	Yes	No

(*continued on next page*)

Table 4 (*continued*)

Author (year)	N	Mean Age (years)	Anxiety Measure	RRB Measure	IU Measure	Anx/RRB Correlation	RRB/IU Correlation	IU/Anx Correlation	ID Included?	% Male Ppts	Autism-Specific Measure - Anxiety	Autism-Specific Measure - RRBs	NE England Researcher Group
Stratis and Lecavalier (2013)	72	11.0	CSI-4	RBS-R	-	0.558 *	-	-	Yes	88.0	No	Yes	No
Uljarevic (2013)	49	10.58	SCAS-P/ PAS	RBQ-2	-	0.35^{+}	-	-	Unclear	91.9	No	Yes	No
Uljarevic et al. (2021)	3532	Not Reported	CBCL	SRS-2	-	0.29^{+}	-	-	Unclear	Not Reported	No	Yes	No
Papers investigating IU and Anxiety													
Boulter et al. (2013)	114	12.7	SCAS-C	-	IUS-C	-	0.70 * *	-	No	87.7	No	-	Yes
Cai et al. (2017)	61	18.18	DSMV-DAS	-	IUS-12	-	0.633 * *	-	Unclear	70.49	No	-	No
MacLennan et al. (2021)	54	4.0	PAS	-	RULES	-	0.796 * *	-	Yes		No	-	No
Maisel et al. (2016)	76	33.82	STAI-T	-	IUS-12	-	0.52 * *	-	No		No	-	Yes
Neil et al. (2016)	64	10.36	SCAS-P	-	IUS-12-P	-	0.74 * *	-	No				No
Sáez-Suanes et al. (2020)	121	35.46	ASA-T	-	IUS-12	-	0.53 * *	-	Yes		Yes	-	No
South et al. (2021)	42	11.08	SCAS-P	-	IUS-12	-	0.64 * *	-	Unclear		No	-	Yes
Tindall (2021)	30	9.13	SCARED	-	IUS-12	-	0.52 * *	-	Unclear		No	-	No
Vasa (2019)	15	4.5	ADIS-ASA	-	Not Reported	-	0.71 * *	-	Unclear		Yes	-	No
Papers investigating IU/RRB and IU/Anxiety													
Vasa et al. (2018)	57	10.94	SCARED-P	RBS-R	IUS-C	-	0.46 * *	0.43 * *	Unclear	82.5	No	No	No

CPRS-R:L: Connor's Parent Rating Scales Revised Long; SCARED: Screen for Child Anxiety Related Disorders; SCAS-P/C: Spence Children's Anxiety Scale-Parent/Child; DSMV-DAS: Diagnostic and Statistical Manual of Mental Disorders-5 Dimensional Anxiety Scale; BAI: Beck Anxiety Inventory; CBCL: Child Behaviour Checklist; PAS: Preschool Anxiety Scale; SMGAD: Severity Measure of Generalised Anxiety Disorder; STAI-T: State-Trait Anxiety Inventory-Trait; HADS: Hospital Anxiety and Depression Scale; ASA-T: Anxiety Scale for Adult with ASD-Informant; CSI-4: Child Symptom Inventory-4; ADIS-ASA: Anxiety Disorders Interview Schedule-Autism Addendum; ADI-R: Autism Diagnostic Interview-Revised; ADOS: Autism Diagnostic Observation Schedule; RBQ: Repetitive Behaviour Questionnaire; RBS-R: Repetitive Behaviour Scale-Revised; SRS: Social Responsiveness Scale; DBC-P: Developmental Behaviour Checklist-Parent; IUS-C/P/12: Intolerance of Uncertainty Scale-Child/Parent/Short Form; ASC-ASD: Anxiety Scale for Children with Autism Spectrum Disorder; RULES: Response to Uncertainty and Low Environmental Structure Questionnaire; PRAS-ASD: Parent-Rated Anxiety Scale for Autism Spectrum Disorder.

*significant at $p < 0.05$ level; * *significant at $p < 0.01$; $^{+}$combined correlation

Table 5
Analysis of Study Quality.

Study Name	Sample Identification	Size of Sample	Confirmation of Autism Diagnosis	Measurement of Anxiety	Measurement of RRBs	Measurement of IU	Quality Score
Avni et al. (2018)	Good	Good	Excellent	Satisfactory	Excellent	N/a	2.2
Ben-Itzchak et al. (2020)	Satisfactory	Satisfactory	Excellent	Excellent	Excellent	N/a	2.2
Boulter et al. (2013)	Good	Good	Good	Satisfactory	N/a	Satisfactory	1.6
Cai et al. (2018)	Good	Satisfactory	Satisfactory	Satisfactory	N/a	Satisfactory	1.2
Cashin and Yorke (2018)	Good	Satisfactory	Good	Satisfactory	Excellent	N/a	1.8
Damiano et al. (2015)	Satisfactory	Poor	Good	Excellent	Excellent	Satisfactory	1.7
Dault (2013)	Good	Satisfactory	Satisfactory	Satisfactory	Excellent	N/a	1.6
Dominick (2010)	Good	Good	Excellent	Satisfactory	Excellent	N/a	2.2
Duvekot et al. (2018)	Good	Good	Good	Satisfactory	Excellent	N/a	2
Factor et al. (2017)	Good	Satisfactory	Excellent	Satisfactory	Excellent	N/a	2
Gabriele (2022)	Good	Good	Satisfactory	Excellent	Excellent	N/a	2.2
Glod et al. (2019)	Good	Poor	Good	Satisfactory	Excellent	Excellent	1.8
Hwang et al. (2020)	Satisfactory	Good	Satisfactory	Satisfactory	Excellent	Satisfactory	1.5
Joyce et al. (2017)	Good	Poor	Good	Satisfactory	Excellent	Satisfactory	1.5
Kim et al. (2021)	Good	Good	Excellent	Satisfactory	Excellent	N/a	2.2
Lidstone et al. (2014)	Good	Satisfactory	Good	Satisfactory	Excellent	N/a	1.8
MacLennan et al. (2021)	Good	Satisfactory	Good	Satisfactory	N/a	Satisfactory	1.4
Magiati et al. (2016)	Good	Good	Good	Satisfactory	Good	N/a	1.8
Maisel et al. (2016)	Good	Satisfactory	Good	Satisfactory	N/a	Satisfactory	1.4
Moore et al. (2021)	Satisfactory	Good	Satisfactory	Excellent	Excellent	Satisfactory	1.8
Muskett et al. (2019)	Good	Satisfactory	Excellent	Satisfactory	Excellent	N/a	2
Neil et al. (2016)	Good	Satisfactory	Good	Satisfactory	N/a	Satisfactory	1.4
Rodgers et al. (2012)	Good	Satisfactory	Good	Satisfactory	Excellent	N/a	1.8
Russell et al. (2019)	Good	Excellent	Good	Satisfactory	Excellent	N/a	2.2
Sáez-Suanes et al. (2020)	Good	Good	Good	Excellent	N/a	Good	2.2
South et al. (2021)	Good	Satisfactory	Satisfactory	Satisfactory	N/a	Satisfactory	1.2
Stratis and Lecavalier (2013)	Good	Satisfactory	Good	Excellent	Excellent	N/a	2.2
Tindall (2021)	Good	Satisfactory	Good	Excellent	N/a	Satisfactory	1.8
Uljarevic (2013)	Good	Satisfactory	Good	Satisfactory	Excellent	N/a	1.8
Uljarevic et al. (2021)	Good	Excellent	Good	Satisfactory	Excellent	N/a	2.2
Vasa (2019)	Poor	Poor	Poor	Excellent	N/a	Poor	0.6
Vasa et al. (2018)	Good	Satisfactory	Good	Excellent	Excellent	Good	2.2
Wigham et al. (2015)	Good	Satisfactory	Satisfactory	Satisfactory	Excellent	Satisfactory	1.5
Mean Score	1.8	1.3	1.9	1.5	3.0	1.3	1.8

The first three factors in the framework rated papers on the quality of their sample. The first assessed the recruitment process: random samples were classified as excellent, multiple/national restricted or non-random samples were classified as good and single restricted or non-random samples were classified as satisfactory. The second factor rated the sample sizes of the papers. A G*power analysis revealed that to detect an effect size of.01 (at $p < 0.05$ with 90 % power), 850 participants would be required. This number was therefore classified as excellent in the quality framework. To detect a medium effect size, with the same parameters, 88 participants would be needed (classified as good), and for a large effect size, 28 participants (satisfactory). The third factor assessed studies on their method of confirming autism diagnosis in their samples, with an excellent classification being given to studies that confirmed clinical diagnoses via a multi-disciplinary team (using validated diagnostic assessment tools), a good classification to those whose participants had a best-estimate diagnosis/previous unconfirmed diagnosis/high score on multiple autism measures, and a satisfactory classification given to those who recruited from a group with higher likelihood of autism diagnosis, or had confirmation by parent/teacher/self-report or a high score on a screening questionnaire.

The other three factors assessed the measurement tools used in the studies (the measures of IU, RRBs and anxiety). Measures were considered excellent if they had been formally validated in autistic samples, good if they included adaptations to attempt to make them

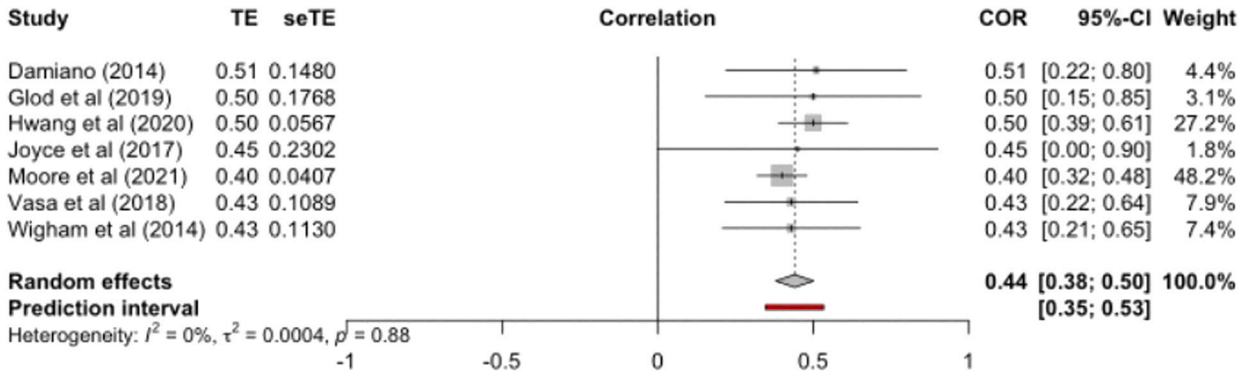

Fig. 4. Forest Plot for the Random Effects Model of the Meta-Analysis Between Restrictive/Repetitive Behaviours and Intolerance of Uncertainty (Meta-Analysis 3).

appropriate for autistic samples, and satisfactory if they were well-validated but not in autistic samples.

The framework criteria were used by two researchers who independently rated all the papers. A good degree of inter-rater reliability was observed, $k = 0.89$. Final scores were agreed upon after any discrepancies had been discussed.

1.5. Analysis strategy

Data were analysed using R (R Core Team, 2021). Three separate meta-analyses were conducted: one for studies looking at the correlation between anxiety and RRBs, one for the correlation between IU and anxiety, and one for the correlation between RRBs and IU. A random effects model (REM) was calculated using the generic inverse variance method and the restricted maximum likelihood estimator (REML). The REML is a more robust estimator in non-normal distributions of effects than the default DerSimonian-Laird estimator, as it minimises bias and controls for underestimation (Cheung, 2013). The REM was chosen over a fixed effects model after reviewing the quantile-quantile (Q-Q) plots (see Supplementary Materials, Figs. 1–3). Heterogeneity statistics were calculated as Higgins I^2. I^2 values above 80 % were considered as high (in line with Higgins et al., 2003). In the case of high heterogeneity, studies deemed discrepant and influential (via review of Baujat plots and leave one out analyses) were reviewed, to check for any significant risk of bias or standout methodological differences. Papers were only removed from further analysis if they evidenced statistical contribution to heterogeneity and the effect estimate *and* either high overall risk of bias or notable methodological deviation from the field. Confidence intervals and Prediction intervals are also reported and were considered during this process, in line with Borenstein et al. (2017). Quality effects models (QEMs) were also calculated for each meta-analysis. Whereas a REM assumes that any variation between studies is random, a QEM models variance as a function of the methodological quality of the studies.

A subgroup analysis was performed to assess differences in relationships between studies that included participants with intellectual disabilities and those that did not. Studies were classified as those that explicitly reported inclusion participants with ID, or a range of IQs with a minimum < 70 (labelled 'yes'), those that explicitly reported exclusion of participants with ID, or a range of IQs with a minimum $\geqslant 70$ (labelled 'no') and those that did not report on their inclusion/exclusion of participants with ID or their range of IQs (labelled 'unclear'). Subgroup analysis was also conducted on the research group who conducted the study. There are many ways in which this can be defined. To maximise power, and provide a coherent strategy, this was determined after retrieval and review. Papers were split into two groups: papers that included at least one author who has been based in the North-East of England (United Kingdom) and papers that did not (a list of which papers were classified in which group is included in Table 4). This allowed for separate consideration of a group of researchers who have conducted a substantial amount of work in the field with multiple collaborations. Only one author (Mirko Uljarevic) featured on different papers classified as in vs. outside of this group. Subgroup analysis was conducted where possible on whether measures included were specific to the autistic population or not. This was only conducted in cases where there were multiple studies in both subgroups.

A meta-regression was conducted with average age of participants in the study as a continuous variable. Similarly meta-regression was conducted on the proportion of male participants in each group; there was a lack of clarity across studies as to whether this referred to the sex or gender of participants (or both). Meta-regression was conducted on the impact of quality rating on the effect; it is worth noting here that whilst the QEM is an important determiner as to whether the findings are reliable in the light of methodological variation/Risk of Bias, the meta-regression based on quality supports consideration of whether quality explains heterogeneity.

2. Results

Extracted data and analysis files are available at https://osf.io/kd36p/?view_only= 65764c74b14644078767eacefaba3ed0".#.

2.1. Study selection

Search 1 returned 1234 results, and Search 2 returned 457 results (Fig. 1). Eighty-seven papers were reviewed at full text, with 33 papers meeting criteria to be included in the review.

2.2. Study characteristics

Full data extraction is detailed in Table 4. Overall, there were 8347 participants across the 33 papers. All studies were correlational in nature, and all were included in the meta-analysis. Twenty-two studies measured anxiety and RRBs, 16 studies measured IU and anxiety, and seven studies measured RRBs and IU. Fourteen studies reported Pearson correlation coefficients, six reported Spearman's coefficients, and 12 did not specify. Participant ages ranged from 2–78 years, with the majority of studies comprising child and adolescent participants. Most samples had a higher proportion of male participants (39.9–94.8 %).

2.3. Study quality

The mean overall quality of papers was 1.8/3 (range of 0.6–2.2) based on the quality framework (Table 5). The highest scoring factor was measurement of RRBs, due to the majority of RRB measures being designed for autistic people, and therefore validated in autistic populations. Following this were the methods of confirmation of autism diagnosis and sample identification. Most studies involved participants with confirmed previous diagnoses by a multi-disciplinary team, however few confirmed these diagnoses in the study itself. No studies recruited from truly random samples, but almost all recruited from multiple regional or national sources. As for the lowest scoring factors: only limited studies used anxiety measures that were validated in autistic populations, and sample sizes were widely varied (ranging from 15–3532), but mostly fell in the 'satisfactory' range. The vast majority of studies measuring IU used the short version of the Intolerance of Uncertainty Scale (IUS), which was developed by Carleton et al. (2007), and adapted by Rodgers et al. (2012) for autistic populations. Studies only scored 'good' if they explicitly cited Rodgers' version.

3. Meta-analysis

3.1. Meta-analysis 1 (Anxiety/RRBs)

Twenty-three studies measured the relationship between Anxiety and RRBs, with a total of 7719 participants. The REM indicated a medium strength positive correlation of $r = 0.40$, 95 % CI: [0.32–0.47], Prediction Interval: [.04–.75]. The forest plot for this analysis is presented in Fig. 2. A high level of heterogeneity was observed, $I^2 = 83$ %. One study (Ben-Itzchak et al., 2020) was noted (via leave one out analysis; see Supplementary Materials, Fig. 4) as contributing substantially to heterogeneity, however, due to a good quality score and methods broadly in line with the other studies it was retained in the analysis. The QEM returned a marginally smaller estimate of the effect, $r = 0.38$, 95 % CI: [0.31–0.46].

Moderators: There was a significant subgroup effect based on whether the study included an autism-specific measure of Restricted and Repetitive Behaviours, $\chi^2(1) = 6.61$, $p = .01$, with those studies that used an autism-specific measure returning lower estimates of the effect, 0.32, 95 % CI: [0.21–0.43], than those studies that did not, 0.49, 95 % CI: [0.42–0.56]. There was also evidence for differences across research groups, $\chi^2(1) = 5.14$, $p = .02$, with studies conducted by the group in the North-East of England finding larger effects, 0.52, 95 % CI: [0.41–0.63], than those conducted by other groups, 0.35, 95 % CI: [0.26–0.44]. Further, there was a significant impact of Study Quality, $Q(1) = 8.12$, $p < .01$, $R^2 = 33$ %, with higher quality studies predicting lower effect sizes. The effect was not significantly moderated by whether studies included people with intellectual disabilities or not, $\chi^2(2) = 5.09$, $p = 0.08$. There was neither a significant impact of average participant age on the effect, $Q(1) = 0.037$, $p = 0.85$, $R^2 = 0$ %, nor a significant impact of proportion of male participants on the effect, $Q(1) = 0.013$, $p = 0.79$, $R^2 = 0$ %,

3.2. Meta-analysis 2 (IU/Anxiety)

Sixteen studies measured the relationship between IU and Anxiety, with a total of 1353 participants. A REM was once again calculated, indicating a large positive correlation of $r = 0.62$, 95 % CI: [0.54–0.69], Prediction Interval: [.33–.90]. The forest plot for this analysis is presented in Fig. 3. A high level of heterogeneity was observed, $I^2 = 80$ %. Once again, one study (Moore et al., 2021) was noted as contributing substantially to heterogeneity, however similarly to above, it was retained in the analysis after review (see Supplementary Materials, Figures 6–7 for Baujat plot/leave one out analysis). The QEM returned a marginally smaller estimate of the effect, $r = 0.61$, 95 % CI: [0.53–0.68].

Moderators**:** Participant age was observed as a significant moderator in the IU/ Anxiety relationship, indicating that the younger the autistic person, the stronger the relationship was ($Q(1) = 14.92$, $p < 0.001$, $R^2 = 73.32$ %). There was no significant subgroup effect based on whether the study included an autism-specific measure of Anxiety, $\chi^2(1) = .15$, $p = .70$. The effect was not significantly moderated by whether studies included people with intellectual disabilities or not, $\chi^2(2) = 0.22$, $p = 0.89$. There was no evidence for differences across research groups, $\chi^2(1) = .15$, $p = .70$. There was no significant impact of Study Quality, $Q(1) = 1.48$, $p = .22$, $R^2 = 4.2$ %. There was not a significant impact of proportion of male participants on the effect, $Q(1) = 2.47$, $p = 0.12$, $R^2 = 25$ %,

3.3. Meta-analysis 3 (RRBs/IU)

Seven studies measured the relationship between RRBs and IU, with a total of 770 participants. The calculated REM indicated a medium positive strength correlation of $r = 0.45$, 95 % CI: [0.37–0.52], Prediction Interval: [.35–.53]. The forest plot for this analysis is presented in Fig. 4. No heterogeneity was observed, $I^2 = 0$ %. The QEM returned a marginally smaller estimate of the effect, $r = 0.44$, 95 % CI: [0.38–0.50].

Moderators: There were no significant moderators of this effect. There was no significant subgroup effect based on whether the study included an autism-specific measure of Restricted and Repetitive Behaviours, $\chi^2(1) = .004$, $p = .85$. The effect was not significantly moderated by whether studies included people with intellectual disabilities or not, $\chi^2(2) = 1.90$, $p = 0.39$. There was no evidence for differences across research groups, $\chi^2(1) = 1.7$, $p = .19$. There was no significant impact of Study Quality, $Q(1) = .01$, $p = .92$, $R^2 = 0$ %. There was neither a significant effect of age, $Q(1) = 0.073$, $p = 0.79$, $R^2 = 0$ %, nor a significant impact of proportion of male participants on the effect, $Q(1) = .01$, $p = .91$, $R^2 = 0$ %.

4. Discussion

Thirty-two studies adopting questionnaires to measure anxiety, RRBs and IU in autistic people were analysed in this review. This is the first time that studies correlating anxiety and RRBs, as well as RRBs and IU, have been collated and meta-analysed. All three relationships of interest returned significant correlations. The strongest effect was observed between IU and anxiety, followed by RRBs and IU, and anxiety and RRBs. The role of IU and RRBs in explaining individual differences in autistic experiences of anxiety is substantial. Models of anxiety in autistic people need to provide a clear explanation of this.

South and Rodgers's (2017) model proposed particularly crucial roles for IU and RRBs in understanding autistic experiences of anxiety; IU mediates the influence of all sensory, emotional and cognitive factors predicting autistic anxiety, other than RRBs. This review is highly consistent with South and Rodgers's (2017) model, in providing evidence for associations between anxiety and RRBs, IU and anxiety, and RRBs and IU. It also updates the review by Jenkinson et al. (2020), consolidating evidence by producing an equal estimate of the relationship between anxiety and IU ($r = 0.62$), and very similar confidence intervals, with a 60 % increase in number of included studies and 1253 % increase in number of participants. Individual variation in IU explains a substantial proportion of individual variation in anxiety in autistic people. It is often theorised that anxiety reduces the experience of uncertainty and increases perception of control over future events (Greco & Roger, 2003). As the probability of occurrence of the anxiety-inducing potential events is actually low, the person often perceives that the occurrence is prevented by the experienced anxiety; as a result, avoidance of anxiety-provoking situations is reinforced, and anxiety is strengthened due to lack of disconfirmation (Boswell et al., 2013). Interventions focussing on a reduction of IU have been demonstrated to reduce anxiety in neurotypical people (Boswell et al., 2013), and notably also show promise for autistic people, including the Coping with Uncertainty in Everyday Situations (CUES) programme (Rodgers et al., 2022). Our findings provide further support for the potential of such interventions.

Each of the three meta-analyses presented here would be firmly predicted by South and Rodgers's (2017) model. The strength of association between IU and anxiety in particular makes IU a good candidate to play a mediating role between anxiety and other factors in autistic people. Notably, though, the model goes further than to predict association; South and Rodgers (2017) propose a bidirectional relationship between IU and anxiety, but unidirectional relationships indicative of IU *causing* anxiety and RRB. As the data analysed in this review were correlational, causation cannot be inferred. However, research into the theoretical mechanisms behind these relationships allows us to generate hypotheses. There is substantial literature suggesting that RRBs may be performed because of anxiety or IU. Both Goodwin et al. (2022) and Joyce et al. (2017) reported qualitative interview data in which young autistic people described engaging with RRBs as a result of anxiety or IU (sometimes as a self-soothing strategies). There appears to be a tentative trend relating IU more specifically to the insistence on sameness subtype of RRBs. For example, participants in Joyce et al. (2017) described engaging in routines to combat feelings of uncertainty. Theoretically, this follows logically, as behaviours that promote sameness serve to reduce unpredictability/uncertainty. Glod et al. (2019) found a significant relationship between IU and insistence on sameness, but not IU and repetitive motor behaviours. Moore et al. (2021) found a stronger correlation between IU and insistence on sameness than IU and repetitive motor behaviours, as did Wigham et al. (2015), Hwang et al. (2020) and Glod et al. (2019). Associations between certain subtypes of RRBs and anxiety have also been found, such as self-injurious behaviour (Kerns et al., 2015; Russell et al., 2019). Therefore, a rational direction for future research would be to analyse the relationship between IU, anxiety and the different subtypes of RRBs separately, leading to more in-depth revelations about the mechanisms relating the two constructs and how we may best support autistic people to cope with uncertainty.[2]

5. Heterogeneity and moderation

As is often the case in meta-analyses of complex real-world relationships with heterogeneous populations, methodological disagreement and relatively small sample sizes, heterogeneity was high for correlations reported between anxiety and RRBs and, anxiety and IU. The confidence and prediction intervals reported for these relationships are therefore wide, suggesting that over time, if study quality/consistency increases, we may see some change in the pooled estimate. Notably, though, we would strongly expect

[2] This was not possible in the present study due to the lack of studies that split RRBs into subtypes, with those that did split RRBs lacking in consistency as to how these subtypes were measured/defined (e.g. between the RBS-R and RBQ).

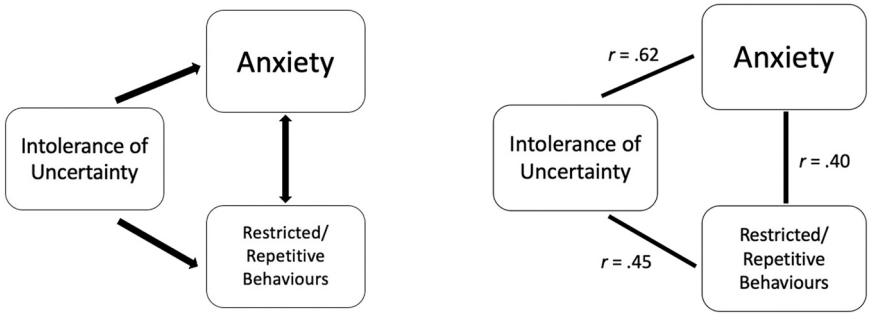

Fig. 5. Diagrams Comparing South and Rodgers' (2017) Model (Left) and Associations Found in Present Review (Right).

these relationships to remain significant. Perhaps more surprising was that heterogeneity was low for the relationship between IU and RRBs, suggesting a high level of consistency across findings. One obvious suggestion here is that IU and RRBs may be related in a more direct manner, with RRBs as a direct response to IU; conversely, their individual relationships to anxiety may allow for a more complex network of interrelations, and mediations/moderations by additional factors, which may promote a higher degree of heterogeneity.

Across the three meta-analyses, a number of significant moderators were identified. There was a significant effect of participant age found on the relationship between IU and anxiety, such that 73 % of heterogeneity was explained by participant age; the relationship was stronger for younger children. McEvoy et al. (2019) found that participant age was not a significant moderator between IU and symptoms of anxiety disorders in neurotypical samples, suggesting that this relationship may be specific to autistic people. Further research is warranted to investigate the difference in continuity of the relationship between IU and anxiety across the lifespan between autistic and non-autistic samples. Age did not significantly moderate either of the other correlations. Proportion of male participants also had no significant moderating effects on any of the relationships. This was perhaps surprising for IU and anxiety, given McEvoy et al. (2019) had shown the relationship to be stronger for studies with a higher proportion of female participants. We remain cautious in interpreting the lack of effect here. McEvoy et al. (2019)'s moderation analysis was conducted on effects drawn from substantially more studies ($k = 181$), giving greater power to identify moderators. Equally, though, relationships between sex/gender and broader experiences in autistic people are complex (Lai et al., 2015), meaning it is not necessarily clear we should expect equivalent moderations across autistic and neurotypical groups. No relationship was moderated by whether the study used an autism-specific measure of anxiety. Only three studies employed such a measure: Gabriele (2022), Sáez-Suanes et al. (2020), and Vasa (2019). The lack of autism-specific measures of anxiety has been recognised, leading to the development of the Anxiety Scale for Children - Autism Spectrum Disorder (ASC-ASD; Rodgers et al., 2016), the Parent-Rated Anxiety Scale for Youth with Autism Spectrum Disorder (PRAS-ASD; Scahill et al., 2019), and the Anxiety Scale for Autism – Adults (ASA-A; Rodgers et al., 2020). We hope that future research will use these tools in order to provide more valid estimates of anxiety in autistic people.

The relationship between anxiety and RRBs was significantly moderated by three factors: whether the study used an autism-specific measure of RRBs, the research group that conducted the study, and the quality index score gained by the study. Studies with larger effects were identified as less likely to use an autism-specific measure of RRBs, more likely to be conducted by the research group based in the North-East of England, and more likely to be scored as lower quality. These findings afford several interpretations. Our view is that they are likely best understood in concert with one another. Research group and methodological choice are clearly not independent, and this is evident here (see Table 4). We would particularly caution against a view that higher effects for non-autism-specific measures of RRBs were somehow evidence of poor methodological choices. Most studies that used a more generally applicable measure of RRBs employed the Repetitive Behaviour Questionnaire (RBQ; Turner, 1996) in one of its variants. Though appropriate for use in broader groups, the RBQ is well-validated for use with autistic people and widely respected in the field. RRBs obviously reflect a range of different behaviours, with a range of different potential functions; our working assumption is that the RBQ measures RRBs that are more strongly linked to anxiety than, for instance the Repetitive Behaviour Scale - Revised (RBS-R, Bodfish et al., 2000). We interpret the moderating influence of research group to be associated with the effect of RRB measure choice. The RBQ is widely used by the group based in the North-East of England (e.g. Glod et al., 2019; Rodgers et al., 2012; Wigham et al., 2014). That the RBQ appears to be strongly associated with anxiety affords potential for future study in closer testing of why this is the case. That lower methodological quality predicted higher effect sizes is harder to interpret. It is worth noting that though lower methodological quality was associated with higher effect sizes, the Quality Effects Model did not produce a substantially different estimate of the effect- suggesting that the moderating effect of study quality did not substantially affect the pooled estimate.

No difference in any relationship was found between studies that included participants with ID and those that did not. This is reassuring in one sense: the anxiety of autistic people with intellectual disabilities remains understudied (Tarver et al., 2020), and lack of moderation suggests that similar models (regarding IU and RRBs at least) may be a good starting point regardless of intellectual ability. Conversely, however, reporting of inclusion of ID groups was inconsistent, measurement of anxiety in autistic people with ID has a smaller evidence base, and those studies that did include people with ID likely focussed on the mild-moderate ID range.

6. Limitations

Across multiple studies, use of measures unvalidated in autistic populations was common, particularly in the measurement of anxiety. Measures that have not been validated in autistic populations may overlook aspects of the autistic experience of anxiety that differ in nature to those of non-autistic people, resulting in inaccurate estimates of anxiety levels in autistic people. For example some RRBs, and autistic traits in general, bear considerable resemblances to anxiety symptoms (Hikmiah, 2019; Kerns & Kendall, 2014), potentially resulting in an artificial inflation of anxiety scores, if not accounted for in measurement tools. This may have added random variability to the data in this review, and experimental power may therefore be reduced. One possibility, therefore, is that the estimates here are underestimates of the true relationships between IU, RRB and anxiety. Another possibility is that confidence intervals are artificially high. Further of note, is that all studies employed questionnaire measures of all variables. One broad risk of validity this presents is that measurement invariance may increase the strength of correlations. The aforementioned differences in the assessment and conceptualisation of RRBs and their subtypes in particular may have contributed to significant variance. A more specific threat is that anxiety, IU and RRB are not mutually exclusive constructs – questionnaire items measuring IU and insistence on sameness/rigidity/adherence to routine in particular regularly overlap (for example, items in the IUS such as 'I should be able to organise everything in advance', or in the ASC-ASD such as 'I am afraid of new things, new people or new places). Direct measures of IU and RRBs may be beneficial in furthering our understanding of these relationships.

Despite demonstrating that anxiety, IU, and RRBs are associated, this review lacks analysis into potential mediating interactions between the variables that may provide insight into directional causal relationships. South and Rodgers (2017) imply causality in these relationships (see Fig. 5). They suggest that one of the reasons that IU related to anxiety is due to its association with RRBs (i.e., RRBs mediate between anxiety and IU). However, quantitative data are lacking to support this proposition; we can only conclude that the three variables co-occur (see Fig. 5). Future research should aim to provide evidence for causality by performing mediation analyses between the three variables, as well as investigating the role of other factors that might additionally co-occur with each of the three.

Given the strength of cross-sectional relationships identified here, it is vital that causality is examined. Recent studies have suggested that RRBs, particularly insistence on sameness behaviours, may predict future anxiety levels in autistic children (Baribeau et al., 2020, 2023), however it is unclear whether this effect continues into early adulthood (Masjedi et al., 2022). Further longitudinal testing of IU, RRBs and anxiety is a key avenue for future research. Also required are more studies that carefully manipulate one of these factors and measure the impact on the others. One obvious way to do this is through intervention to increase tolerance of uncertainty or reduce RRBs. We do, however, urge caution with the second of these approaches, for reasons that highlight the importance of determining directionality in these relationships. If we assume that RRBs *cause* anxiety, we design interventions to reduce RRBs, with the aim of reducing anxiety. Importantly, though, anxiety may *cause* an increase in RRBs. If this is the case, intervening to reduce RRBs may instead be taking away a key coping mechanism used by autistic people and hence be increasing anxiety. Again, emphasis must be placed on qualitative evidence, such as Goodwin et al. (2022) and Joyce et al. (2017), who demonstrate that RRBs can appear to be markers of IU and anxiety rather than causes of them. Such research can direct theory as to how the relationships between variables shown in this analysis play out in the lived experience of autistic people, and point us towards potential methods of intervention. We must be wary, however, until further evidence is collected. Until then, individualized formulation is likely a key approach, working collaborative with anxious autistic people to understand the role of RRBs for individuals. The distinction between RRBs acting as coping mechanisms (reducing the impact of anxiety on everyday life) vs. safety behaviours (perpetuating anxiety) likely varies across individuals and within individuals over time.

7. Implications

This meta-analysis is the first to bring together research into the associations between anxiety, IU and RRBs in autistic people, to calculate pooled estimates of correlations between these variables. Crucially, it is also the first to collate evidence for how RRBs may fit in to the anxiety/ IU relationship. The analysis demonstrates that there are strong positive relationships between anxiety, IU, and RRBs in autistic people. Based on this, an emphasis should be placed on addressing IU when designing anxiety interventions for autistic people. However, if we fail to comprehend the role of RRBs, we risk modelling this relationship insufficiently, which may have harmful consequences. Without understanding the details of the relationship between RRBs and IU/ anxiety, as well as RRBs in general, we cannot know what the cost of implementing such interventions could be on autistic people, and on the neurodiverse community as a whole.

CRediT authorship contribution statement

Sydney Bird: Writing – review & editing, Writing – original draft, Visualization, Investigation, Formal analysis, Conceptualization. **Christopher A. Jones:** Software, Methodology. **Lucy Anne Elizabeth Moid:** Validation. **Andrew D. R. Surtees:** Writing – review & editing, Writing – original draft, Supervision, Project administration, Formal analysis, Conceptualization.

Declaration of Competing Interest

None.

Data availability

Data available online (link provided in manuscript).

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Appendix A. Supporting information

Supplementary data associated with this article can be found in the online version at doi:10.1016/j.rasd.2024.102428.

References

Avni, E., Ben-Itzchak, E., & Zachor, D. A. (2018). The presence of comorbid ADHD and anxiety symptoms in autism spectrum disorder: Clinical presentation and predictors. *Frontiers in Psychiatry, 9*, 717.

Baribeau, D. A., Vigod, S., Pullenayegum, E., Kerns, C. M., Mirenda, P., Smith, I. M., Vaillancourt, T., Volden, J., Waddell, C., Zwaigenbaum, L., Bennett, T., Duku, E., Elsabbagh, M., Georgiades, S., Ungar, W. J., Zaidman-Zait, A., & Szatmari, P. (2020). Repetitive behavior severity as an early indicator of risk for elevated anxiety symptoms in autism spectrum disorder. *Journal of the American Academy of Child & Adolescent Psychiatry, 59*(7), 890–899.

Baribeau, D. A., Vigod, S. N., Pullenayegum, E., Kerns, C. M., Vaillancourt, T., Duku, E., Smith, I. M., Volden, J., Zwaigenbaum, L., Bennett, T., Elsabbagh, M., Zaidman-Zait, A., Richard, A. E., & Szatmari, P. (2023). Developmental cascades between insistence on sameness behaviour and anxiety symptoms in autism spectrum disorder. *European Child & Adolescent Psychiatry, 32*(11), 2109–2118.

Ben-Itzchak, E., Koller, J., & Zachor, D. A. (2020). Characterisation and prediction of anxiety in adolescents with autism spectrum disorders: A longitudinal study. *Journal of Abnormal Child Psychology, 48*(9), 1239–1249.

Barrett, S. L., Uljarević, M., Baker, E. K., Richdale, A. L., Jones, C. R., & Leekam, S. R. (2015). The Adult Repetitive Behaviours Questionnaire-2 (RBQ-2A): A self-report measure of restricted and repetitive behaviours. *Journal of Autism and Developmental Disorders, 45*, 3680–3692.

Bodfish, J. W., Symons, F. J., Parker, D. E., & Lewis, M. H. (2000). Varieties of repetitive behavior in autism: Comparisons to mental retardation. *Journal of Autism and Developmental Disorders, 30*, 237–243.

Borenstein, M., Higgins, J. P. T., Hedges, L. V., & Rothstein, H. R. (2017). Basics of meta-analysis: I^2 is not an absolute measure of heterogeneity. *Research Synthesis Methods, 8*(1), 5–18.

Boswell, J. F., Thompson-Hollands, J., Farchione, T. J., & Barlow, D. H. (2013). Intolerance of uncertainty: A common factor in the treatment of emotional disorders. *Journal of Clinical Psychology, 69*(6), 630–645.

Boulter, C., Freeston, M., South, M., & Rodgers, J. (2013). Intolerance of uncertainty as a framework for understanding anxiety in children and adolescents with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 44*(6), 1391–1402.

Cai, R. Y., Richdale, A. L., Dissanayake, C., & Uljarevic, M. (2017). Brief report: Inter-relationship between emotion regulation, intolerance of uncertainty, anxiety, and depression in youth with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 48*(1), 316–325.

Carleton, R. N., Norton, M. P. J., & Asmundson, G. J. (2007). Fearing the unknown: A short version of the Intolerance of Uncertainty Scale. *Journal of Anxiety Disorders, 21*, 105–117.

Cashin, A., & Yorke, J. (2018). The relationship between anxiety, external structure, behavioural history and becoming locked into restricted and repetitive behaviours in autism spectrum disorder. *Issues in Mental Health Nursing, 39*(6), 533–537.

Collis, E., Gavin, J., Russell, A., & Brosnan, M. (2022). Autistic adults' experience of restricted repetitive behaviours. *Research in Autism Spectrum Disorders, 90*, Article 101895.

Damiano, C.R. (2014). *Neural mechanisms of uncertainty processing in children with autism spectrum disorders.* [Doctoral thesis, University of North Carolina at Chapel Hill]. ProQuest Dissertations and Theses.

Dault, L. (2013). *Parent-reported behavioural symptoms of anxiety in children with autism spectrum disorders across levels of functioning.* [Master's thesis, Queen's University (Canada)]. ProQuest Dissertations and Theses.

Dominick, K.R. C. (2010). *Repetitive behaviours in autism spectrum disorder and the role of the striatum.* [Doctoral thesis, Boston University]. ProQuest Dissertations and Theses.

Dugas, M. J., Gagnon, F., Ladouceur, R., & Freeston, M. H. (1998). Generalized anxiety disorder: A preliminary test of a conceptual model. *Behaviour Research and Therapy, 36*(2), 215–226.

Duvekot, J., Van Der Ende, J., Verhulst, F. C., & Greaves-Lord, K. (2018). Examining bidirectional effects between the autism spectrum disorder (ASD) core symptom domains and anxiety in children with ASD. *Journal of Child Psychology & Psychiatry, 59*(3), 277–285.

Factor, R. S., Ryan, S. M., Farley, J. P., Ollendick, T. H., & Scarpa, A. (2017). Does the presence of anxiety and ADHD symptoms add to social impairment in children with autism spectrum disorder? *Journal of Autism and Developmental Disorders, 47*(4), 1122–1134.

Foundation for People with Learning Disabilities (n.d.). *Learning Disability Statistics: Autism. https://www.learningdisabilities.org.uk/learning-disabilities/help-information/statistics/learning-disability-statistics-/187690#:~:text=60%2D70%25%20of%20people%20who,disability%20or%20lower%20verbal%20IQ.&text=Autism%20spectrum%20conditions%20are%20associated%20with%20educational%20qualification.*

Gabriele, A. (2022). *Exploring the relationship between restricted and repetitive behaviours, anxiety, and aggression in children with autism spectrum disorder.* [Doctoral thesis, St. John's University]. ProQuest Dissertations and Theses.

Goodwin, J., Rob, P., Freeston, M., Garland, D., Grahame, V., Kernohan, A., Labus, M., Osborne, M., Parr, J. R., Wright, C., & Rodgers, J. (2022). Caregiver perspectives on the impact of uncertainty on the everyday lives of autistic children and their families. *Autism: The International Journal of Research and Practice, 26*(4), 827–839.

Glod, M., Riby, D. M., & Rodgers, J. (2019). Short report: Relationships between sensory processing, repetitive behaviours, anxiety, and intolerance of uncertainty in autism spectrum disorder and Williams syndrome. *Autism Research, 12*(5), 759–765.

Greco, V., & Roger, D. (2003). Uncertainty, stress, and health. *Personality and Individual differences, 34*(6), 1057–1068.

Higgins, J. P., Thompson, S. G., Deeks, J. J., & Altman, D. G. (2003). Measuring inconsistency in meta-analyses. *BMJ, 327*(7414), 557–560.

Hikmiah, Z. (2019). Heterogeneity in anxiety disorder and autism spectrum disorder: Difference and overlap. *Indonesian Journal of Disability Studies, 6*(2), 269–281.

Hwang, Y. I., Arnold, S., Srasuebkul, P., & Trollor, J. (2020). Understanding anxiety in adults on the autism spectrum: An investigation of its relationship with intolerance of uncertainty, sensory sensitivities and repetitive behaviours. *Autism: The International Journal of Research and Practice, 24*(2), 411–422.

Jenkinson, R., Milne, E., & Thompson, A. (2020). The relationship between intolerance of uncertainty and anxiety in autism: A systematic literature review and meta-analysis. *Autism: The International Journal of Research and Practice, 24*(8), 1933–1945.

Joosten, A. V., Bundy, A. C., & Einfield, S. L. (2008). Intrinsic and extrinsic motivation for stereotypic and repetitive behaviour. *Journal of Autism and Developmental Disorders, 39*(3), 521–531.
Joyce, C., Honey, E., Leekam, S. R., Barrett, S. L., & Rodgers, J. (2017). Anxiety, intolerance of uncertainty and restricted and repetitive behaviour: Insights directly from young people with ASD. *Journal of Autism and Developmental Disorders, 47*(12), 3789–3802.
Kalvin, C. B., Marsh, C. L., Ibrahim, K., Gladstone, T. R., Woodward, D., Grantz, H., Ventola, P., & Sukhodolsky, D. G. (2020). Discrepancies between parent and child ratings of anxiety in children with autism spectrum disorder. *Autism Research, 13*(1), 93–103.
Kanner, L. (1943). Autistic disturbances of affective contact. *Nervous Child, 2*, 217–250.
Kerns, C. M., & Kendall, P. C. (2014). Autism and anxiety: Overlap, similarities, and differences. *Handbook of Autism and Anxiety*, 75–89.
Kerns, C. M., Kendall, P. C., Berry, L., Souders, M. C., Franklin, M. E., Schultz, R. T., Miller, J., & Herrington, J. (2014). Traditional and Atypical Presentations of Anxiety in Youth with Autism Spectrum Disorder. *Journal of Autism and Developmental Disorders, 44*, 2851–2861.
Kerns, C. M., Kendall, P. C., Zickgraf, H., Franklin, M. E., Miller, J., & Herrington, J. (2015). Not to be overshadowed or overlooked: Functional impairments associated with comorbid anxiety disorders in youth with ASD. *Behavior Therapy, 46*(1), 29–39.
Kim, S. Y., Kim, Y. A., Song, D., Bong, G., Kim, J., Kim, J. H., & Yoo, H. J. (2021). State and trait anxiety of adolescents with autism spectrum disorders. *Psychiatry Investigation, 18*(3), 257–265.
Lai, M., Lombardo, M. V., Auyeung, B., Chakrabarti, C., & Baron-Cohen, S. (2015). Sex/gender differences and autism: Setting the scene for future research. *Journal of the American Academy of Child & Adolescent Psychiatry, 54*(1), 11–24.
Lau, B. Y., Leong, R., Uljarevic, M., Lerh, J. W., Rodgers, J., Hollocks, M. J., South, M., McConachie, H., Ozsivadjian, A., Van Hecke, A., Libove, R., Hardan, A., Leekam, S., Simonoff, E., & Magiati, I. (2020). Anxiety in young people with autism spectrum disorder: Common and autism-related anxiety experiences and their association with individual characteristics. *Autism; The International Journal of Research & Practice, 24*(5), 1111–1127.
Leekam, S., Tandos, J., McConachie, H., Meins, E., Parkinson, K., Wright, C., Turner, M., Arnott, B., Vittorini, L., & Le Couteur, A. (2007). Repetitive behaviours in typically developing 2-year-olds. *Journal of Child Psychology and Psychiatry, 48*(11), 1131–1138.
Leekam, S. R., Prior, M. R., & Uljarevic, M. (2011). Restricted and repetitive behaviours in autism spectrum disorders: A review of research in the last decade. *Psychological Bulletin, 137*(4).
Lewis, M., & Kim, S. (2009). The pathophysiology of restricted repetitive behaviour. *Journal of Neurodevelopmental Disorders, 1*(2), 114–132.
Lidstone, J., Uljarevic, M., Sullivan, J., Rodgers, J., McConachie, H., Freeston, M., Le Couteur, A., Prior, M., & Leekam, S. (2014). Relations among restricted and repetitive behaviours, anxiety and sensory features in children with autism spectrum disorders. *Research in Autism Spectrum Disorders, 8*(2), 82–92.
MacLennan, K., Rossow, T., & Tavassoli, T. (2021). The relationship between sensory reactivity, intolerance of uncertainty and anxiety subtypes in preschool-age autistic children. *Autism: The International Journal of Research & Practice, 25*(8), 2305–2317.
Magiati, I., Ong, C., Lim, X. Y., Tan, J. W., Ong, A. Y. L., Patrycia, F., Fung, D. S. S., Sung, M., Poon, K. K., & Howlin, P. (2016). Anxiety symptoms in young people with autism spectrum disorder attending special schools: Associations with gender, adaptive functioning and autism symptomatology. *Autism: The International Journal of Research & Practice, 20*(3), 306–321.
Maisel, M. E., Stephenson, K. G., South, M., Rodgers, J., Freeston, M. H., & Gaigg, S. B. (2016). Modelling the cognitive mechanisms linking autism symptoms and anxiety in adults. *Journal of Abnormal Psychology, 125*(5), 692–703.
Masjedi, N., Clarke, E. B., & Lord, C. (2022). Development of restricted and repetitive behaviors from 2-19 and internalizing symptom outcomes in a longitudinal study of autism. *medRxiv, 06*, 2022.
McEvoy, P. M., Hyett, M. P., Shihata, S., Price, J. E., & Strachan, L. (2019). The impact of methodological and measurement factors on transdiagnostic associations with intolerance of uncertainty: A meta-analysis. *Clinical Psychology Review, 73*, Article 101778.
McFayden, T. C., Antezana, L., Albright, J., Muskett, A., & Scarpa, A. (2020). Sex differences in autism spectrum disorder diagnosis: Are restricted repetitive behaviours and interests the key? *Review Journal of Autism and Developmental Disorders, 7*, 119–126.
Mingins, J. E., Tarver, J., Waite, J., Jones, C., & Surtees, A. D. R. (2021). Anxiety and intellectual functioning in autistic children: A systematic review and meta-analysis. *Autism, 25*(1), 18–32.
Moore, H. L., Brice, S., Powell, L., Ingham, B., Freeston, M., Parr, J. R., & Rodgers, J. (2021). The mediating effects of alexithymia, intolerance of uncertainty, and anxiety on the relationship between sensory processing differences and restricted and repetitive behaviours in autistic adults. *Journal of Autism and Developmental Disorders*, 1–13.
Muskett, A., Capriola-Hall, N. N., Radtke, S. R., Factor, R., & Scarpa, A. (2019). Repetitive behaviours in autism spectrum disorder: Associations with depression and anxiety symptoms. *Research in Autism Spectrum Disorders, 68*, Article 101449.
Neil, L., Olsson, N. C., & Pellicano, E. (2016). The relationship between intolerance of uncertainty, sensory sensitivities, and anxiety in autistic and typically developing children. *Journal of Autism and Developmental Disorders, 46*(6), 1962–1973.
Norr, A. M., Oglesby, M. E., Capron, D. W., Raines, A. M., Korte, K. J., & Schmidt, N. B. (2013). Evaluating the unique contribution of intolerance of uncertainty relative to other cognitive vulnerability factors in anxiety psychopathology. *Journal of Affective Disorders, 151*(1), 136–142.
Osmanağaoğlu, N., Creswell, C., & Dodd, H. F. (2018). Intolerance of uncertainty, anxiety, and worry in children and adolescents: A meta-analysis. *Journal of Affective Disorders, 225*, 80–90.
R Core Team (2021). *R: A language and environment for statistical computing*. R Foundation for Statistical Computing, Vienna, Austria. https://www.r-project.org/.
Rodgers, F., Farquhar, K., Mason, D., Brice, S., Wigham, S., Ingham, B., Freeston, M., & Parr, J. R. (2020). Development and initial evaluation of the anxiety scale for autism – adults. *Autism in Adulthood, 2*(1), 24–33.
Rodgers, J., Freeston, M., South, M., Wigham, S., & Boulter, C. (2012). Intolerance of uncertainty: Its relevance to autism spectrum disorder. In *British Association of Behavioural and Cognitive Psychotherapy Annual Meeting, Leeds, UK*.
Rodgers, J., Goodwin, J., Garland, D., Grahame, V., Isard, L., Kernohan, A., Labus, M., Osborne, M., Parr, J. R., Rob, P., Wright, C., & Freeston, M. (2022). Coping with uncertainty in everyday situations (CUES©) to address intolerance of uncertainty in autistic children: an intervention feasibility trial. *Journal of Autism and Developmental Disorders*, 1–15.
Rodgers, J., Riby, D., Janes, E., Connolly, B., & McConachie, H. (2012). Anxiety and repetitive behaviours in autism spectrum disorders and Williams syndrome: A cross-syndrome comparison. *Journal of Autism and Developmental Disorders, 42*(2), 175–181.
Rodgers, J., Wigham, S., McConachie, H., Freeston, M., Honey, E., & Parr, J. R. (2016). Development of the anxiety scale for children with autism spectrum disorder (ASC-ASD). *Autism Research, 9*(11), 1205–1215.
Russell, K. M., Frost, K. M., & Ingersoll, B. (2019). The relationship between subtypes of repetitive behaviours and anxiety in children with autism spectrum disorder. *Research in Autism Spectrum Disorders, 62*, 48–54.
Sáez-Suanes, G. P., García-Villamisar, D., Del Pozo Armentia, A., & Dattilo, J. (2020). Emotional dysregulation and uncertainty intolerance as transdiagnostic mediators of anxiety in adults with autism spectrum disorders and intellectual disability. *Research in Developmental Disabilities, 106*, Article 103784.
Scahill, L., Lecavalier, L., Schultz, R. T., Evans, A. N., Maddox, B., Pritchett, J., Herrington, J., Gillespie, S., Miller, J., Amoss, R. T., Aman, M. G., Bearss, K., Gadow, K., & Edwards, M. C. (2019). Development of the parent-rated anxiety scale for youth with autism spectrum disorder. *Journal of the American Academy of Child & Adolescent Psychiatry, 58*(9), 887–896.
South, M., Hanley, M., Normansell-Mossa, K., Russell, N. C. C., Cawthorne, T., & Riby, D. M. (2021). "Intolerance of uncertainty" mediates the relationship between social profile and anxiety in both Williams syndrome and autism. *Autism Research, 14*(9), 1986–1995.
South, M., & Rodgers, J. (2017). Sensory, emotional and cognitive contributions to anxiety in autism spectrum disorders. *Frontiers in Human Neuroscience, 11*, 20.
Stratis, E. A., & Lecavalier, L. (2013). Restricted and repetitive behaviours and psychiatric symptoms in youth with autism spectrum disorders. *Research in Autism Spectrum Disorders, 7*(6), 757–766.
Tarver, J., Pearson, E., Edwards, G., Shirazi, A., Potter, L., Malhi, P., & Waite, J. (2020). Anxiety in autistic individuals who speak few or no words: A qualitative study of parental experience and anxiety management. *Autism, 25*(2), 429–439.

Tindall, K.L. U. (2021). *Behavioural and neural correlates of sensory processing and anxiety in autistic children.* [Master's thesis, Brigham Young University]. ProQuest Dissertations and Theses.

Turner, M.A. (1996). *Repetitive behaviour and cognitive functioning in autism.* [Doctoral thesis, University of Cambridge]. ProQuest Dissertations and Theses.

Uljarevic, M. (2013). *Repetitive behaviours, anxiety and sensory problems in children with autism and correlates of anxiety in their parents.* [Doctoral thesis, Cardiff University]. ProQuest Dissertations and Theses.

Uljarevic, M., Hedley, D., Rose-Foley, K., Magiati, I., Cai, R. Y., Dissanayake, C., Richdale, A., & Trollor, J. (2020). *Anxiety and depression from adolescence to old age in Autism Spectrum Disorder, 50(9)), 3155–3165*.

Uljarevic, M., Jo, B., Frazier, T. W., Scahill, L., Youngstrom, E. A., & Hardan, A. Y. (2021). Using the big data approach to clarify the structure of restricted and repetitive behaviours across the most commonly used autism spectrum disorder measures. *Molecular Autism, 12*(1), 1–39.

van Steensel, F. J. A., Bögels, S. M., & Perrin, S. (2011). Anxiety disorders in children and adolescents with autistic spectrum disorders: A meta-analysis. *Clinical Child and Family Psychology Review, 14*, 302–317.

Vasa, R. A. (2019). Anxiety and intolerance of uncertainty in preschool children with ASD. *Journal of the American Academy of Child and Adolescent Psychiatry, 58*, S338.

Vasa, R. A., Kreiser, N. L., Keefer, A., Singh, V., & Mostofsky, S. H. (2018). Relationships between autism spectrum disorder and intolerance of uncertainty. *Autism Research, 11*(4), 636–644.

Vasa, R. A., & Mazurek, M. O. (2015). An update on anxiety in youth with autism spectrum disorders. *Current Opinion in Psychiatry, 28*(2), 83–91.

Wigham, S., Rodgers, J., South, M., McConachie, H., & Freeston, M. (2014). The interplay between sensory processing abnormalities, intolerance of uncertainty, anxiety and restricted and repetitive behaviours in autism spectrum disorder. *Journal of Autism and Developmental Disorders, 45*(4), 943–952.